Infratemporal Fossa Cellulitis Caused by a Remnant Iatrogenic Foreign Body After a Bimaxillary Operation

Do Yang Park, MD, Oak-Sung Choo, MD,† Sang Young Hong, MD,† and Hyun Jun Kim, MD†*

Abstract: Infratemporal fossa cellulitis is rare and mostly occurs because of sinusitis and dental procedures. Furthermore, cellulitis caused by iatrogenic foreign bodies is very rare.

A 28-year-old woman who had previously undergone cosmetic bimaxillary operation visited our hospital complaining of left facial swelling, oppressive pain, and nasal obstruction since 2 years. She had been attending another clinic, but despite having additional procedures and taking medications, her symptoms persisted. A subsequent operation was performed, during which we found a remnant surgical gauze from the previous operation, which was decomposed and trapped around the necrotic soft tissue and had eroded the bony structure around the pterygoid fossa. The material was successfully removed by endoscopic surgery, and the necrotic tissue was debrided. After the operation, all symptoms disappeared, and the patient was discharged without sequelae.

During any procedure, surgeons must meticulously check for remnant material. Additionally, physicians must carefully note patient history and perform a physical examination, even in patients without serious symptoms. We report a case of advanced infratemporal fossa cellulitis due to remnant gauze material during a previous operation that was undetected.

Key Words: Foreign bodies, nasopharynx, infratemporal fossa, complications, aesthetic surgery

Infratemporal cellulitis is rare but possibly life threatening. It can arise from several sources, especially from sinusitis and dental procedures, and may have unknown origins.[1–3] This disease causes nasal obstruction, sneezing, visual acuity, orbital abscess, and central nervous system rhinorrhea and infection.[1] However, due to anatomical difficulty, diagnosis can be delayed despite proper physical examination and radiological testing.[4] Severe complications can develop during this delay. Herein, we present a case of iatrogenic infratemporal cellulitis after a bimaxillary operation; in addition, we provide a literature review.

CLINICAL REPORT

A 28-year-old woman visited our outpatient department with a 2-year history of left facial swelling, oppressive pain, and nasal obstruction. Her medical history was unremarkable. Surgically, however, 5 years ago, she had undergone rhinoplasty with hump removal and dorsal augmentation at a local clinic. Additionally, 3 years ago, she had undergone bimaxillary operation with sagittal split osteotomy and Le Fort osteotomy at another local clinic for aesthetic reasons.

After the bimaxillary operation, the patient experienced left-side nasal obstruction, postnasal drip, ear fullness, and hypoacusis. She therefore visited an otolaryngologist from another clinic and had undergone a v-tube insertion in the left ear. Despite several medicines and procedures, her symptoms persisted. Therefore, she was referred to our Department of Otolaryngology–Head and Neck Surgery.

At the first visit, the patient's vital signs were within the normal ranges, except for a fever of 37.6°C. Gingiva swelling, heat sensation on the face, and mild trismus were also noted. In the left nasal cavity, pus discharge and mucosal swelling were observed. In the nasopharynx, tiny white material was noted, and a sample was taken for biopsy, confirming the presence of *Actinomyces* and surgical fabric.

On a facial computed tomography (CT) and paranasal sinus magnetic resonance image (PNS MRI), a 3.3-cm low-density material and inflammation was noted at the infratemporal fossa, and the inflammation was expanded to the pterygoid muscle, parapharyngeal space, and partially to the parotid gland. Therefore, infratemporal fossa cellulitis with an eroded pterygoid plate was diagnosed (Fig. 1).

Navigator-assisted endoscopic surgery was planned. Through the left nasal cavity, an incision was performed at the pterygoid level, compared to the inflamed soft tissue and foreign body lesion of navigator system. For the removal of necrotic tissue, ethmoidectomy, middle meatal antrostomy, and turbinectomy were performed. Necrotic soft tissue and surgical gauze material were noted at the nasopharyngeal lateral wall. Posterior mucosa of the medial pterygoid plate was debrided, and a partial pterygoid plate was drilled out. A surgical gauze was found and was removed using the navigation system; the gauze was found to be crushed and had adhered to the surrounding soft tissue by an inflammatory process (see Supplemental Digital Content, Video, http://links.lww.com/SCS/A114). The surgeon removed almost all the foreign body material around the medial and lateral pterygoid plate and the infratemporal fossa by endoscopic surgery. The operation was performed with nasal packing (Fig. 2).

Methicillin-resistant *Staphylococcus aureus* was harvested from the pus discharge in the pathologic tissue. Intravenous antibiotics and conservative care were provided during the patient's 6 days in the hospital. Immediately after the operation, the nasal obstruction, facial

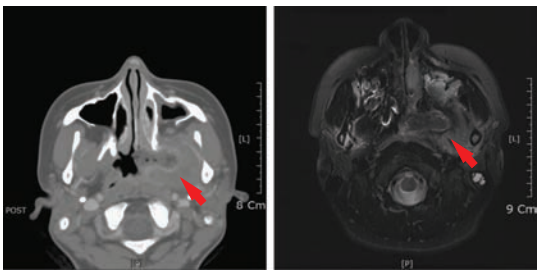

FIGURE 1. Preoperative facial CT and PNS MRI showing a hypodense inflamed lesion. CT indicates computed tomography; PNS MRI, paranasal sinus magnetic resonance image.

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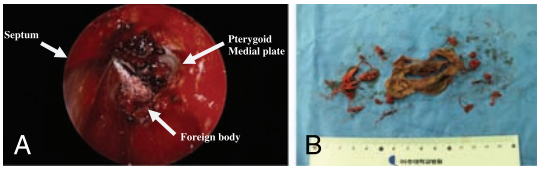

FIGURE 2. A, Operation finding showing a white fabric on endoscopic surgical view. B, Pathologic finding showing the foreign body materials. Almost all were removed.

pain, and trismus improved. One month later, the nasal obstruction and facial pain completely disappeared. No foreign body material or inflammation was noted at the follow-up facial CT (Fig. 3). At the 1-year follow-up visit, the patient did not present any symptoms.

DISCUSSION

Infratemporal fossa cellulitis mostly originates from sinusitis and dental procedures.[1,2] Fever, facial pain, swelling, and trismus probably occurred because of an inflamed masseter muscle.[5] Orbital invasion can lead to decreased visual acuity and orbital abscess; invasion through a skull base lesion[6] can lead to CNS rhinorrhea, infection, and abscess.[1]

A detailed medical interview, physical examination, and radiological examination were necessary when the patient first presented symptoms. However, early diagnosis can be difficult by radiological examination due to anatomical difficulty.[4] An MRI is a good tool to examine the lesion and extent of the disease. However, performing CT is necessary to evaluate the surgical extent and approach needed.[4]

To date, infratemporal fossa cellulitis due to iatrogenic surgical gauze has not been reported. However, the presence of a metallic foreign body in the nasal cavity, sinus, and orbit has been reported.[7] Iatrogenic remnant gauze packing and complications after adenoidectomy have also been reported.[8] Improvement in patient symptoms was observed after surgical foreign body removal in these cases. Essentially, in all cases reported, surgical abscess of the inflammation or abscess due to the presence of the foreign body was vital.[7–9]

In Oriental culture, bimaxillary operation is commonly performed for aesthetic purposes. The most common complication is temporal facial numbness.[10] In the current case, bleeding may have occurred and the surgical gauze may have been packed for hemostasis in the previous bimaxillary operation. The narrow view during the bimaxillary operation may have facilitated the remnant gauze.

Intraoperative hemorrhage from Le Fort I osteotomy may be either muscular (involving the pterygoid musculature), osseous (involving the palatine bone), or vascular (involving superior alveolar and descending palatine arteries) in origin.[11] In the present case, the foreign body may have migrated to the nasopharynx or to the nearest open space, and the intermediated anatomical structures were eroded by an inflammatory reaction. This inflammation affected the infratemporal fossa, nasopharynx, medial and lateral pterygoid plate, and masseter muscle, causing facial pain, serous otitis media, and trismus.

In the present case, the disease was difficult to diagnose for the aforementioned reasons; therefore, the treatment was delayed and complications developed. Surgeons must pay close attention when operating in this anatomical area. In this case, when the left-side serous otitis media was diagnosed, a detailed interview and physical examination can have aided early detection of the foreign body. A detailed medical interview, careful physical examination, and proper radiological examination are required to diagnose a foreign body-related disease. Surgical removal is the treatment of choice, and suitable antibiotics should also be administered.[12]

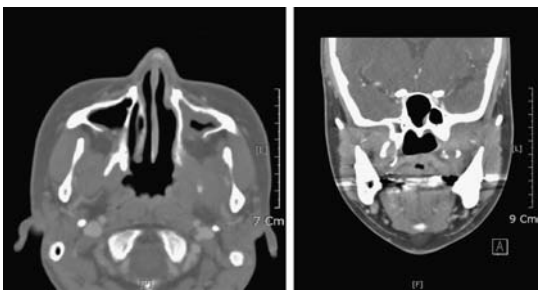

FIGURE 3. Postoperative computed tomography showing healing of the cellulitis and the absence of remnant material.

REFERENCES

1. de Oliveira Neto PJ, de Souza Maliska MC, Sawazaki R, et al. Temporal abscess after third molar extraction in the mandible. *Oral Maxillofac Surg* 2012;16:107–110
2. Weiss BR. Infratemporal fossa abscess unusual complication of maxillary sinus fracture. *Laryngoscope* 1977;87:1130–1133
3. Leventhal D, Schwartz DN. Infratemporal fossa abscess: complication of dental injection. *Arch Otolaryngol Head Neck Surg* 2008;134:551–553
4. Kamath M, Bhojwani K, Mahale A, et al. Infratemporal fossa abscess: a diagnostic dilemma. *Ear Nose Throat J* 2009;88:E23
5. Gallagher J, Marley J. Infratemporal and submasseteric infection following extraction of a non-infected maxillary third molar. *Br Dent J* 2003;194:307–309
6. Raghava N, Evans K, Basu S. Infratemporal fossa abscess: complication of maxillary sinusitis. *J Laryngol Otol* 2004;118:377–378
7. Kim JH, Kim SH, Kim JH, et al. A case of metallic foreign body retained in the naso-maxillo-ethmoido-orbital complex. *Korean J Otorhinolaryngol-Head Neck Surg* 2004;47:177–181
8. Ozer C, Ozer F, Sener M, et al. A forgotten gauze pack in the nasopharynx: an unfortunate complication of adenotonsillectomy. *Am J Otolaryngol* 2007;28:191–193
9. Shemen LJ, Schechter LS, Godfrey N. Needle-wire localization of an infratemporal fossa foreign body using computed tomography. *Arch Otolaryngol Head Neck Surg* 1992;118:1337–1339
10. Choi BK, Goh RC, Moaveni Z, et al. Patient satisfaction after zygoma and mandible reduction surgery: an outcome assessment. *J Plast Reconstr Aesthet Surg* 2010;63:1260–1264
11. Kang SH, Yoo JH, Yi CK. The efficacy of postoperative prophylactic antibiotics in orthognathic surgery: a prospective study in Le Fort I osteotomy and bilateral intraoral vertical ramus osteotomy. *Yonsei Med J* 2009;50:55–59
12. Kang SJ, Jeon SP. Surgical treatment of periorbital foreign body. *J Craniofac Surg* 2012;23:e603–e605

Unusual Long-Term Complication of Polyalkylimide Hydrogel Manifesting as Nasal Septal Abscess

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Abstract: Bio-Alcamid is a new synthetic polyacrylic hydrogel that contains alkylimide-amide groups and pyrogen-free water (96%) and has gained widespread use in cosmetic and reconstructive

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practice since being awarded a Conformite European certificate in 2001.

According to the research on the efficacy and safety of the long-acting filler Bio-Alcamid in the early phases of development, Bio-Alcamid is nearly nontoxic and nonallergenic and has long in vivo persistence. It has been widely used because of its superior durability compared with short-acting fillers.

Many published studies have examined only early-phase postoperative inflammatory responses. More recently, however, complications with delayed onsets ranging from several months to several years, such as inflammation and filler migration, have been emerging. Given the rapidly increasing application of long-term fillers, an increased incidence of complications is expected, and increasing awareness of its correct use and complication treatments is needed.

To address this problem, we reviewed the safety of long-acting fillers on the basis of a rare case of surgical incision and drainage of a nasal septal abscess that developed in a patient 2 years after Bio-Alcamid was injected into the nasal region.

Key Words: Bio-Alcamid, long acting filler, complication

Various fillers have been used in plastic and reconstructive surgery, especially for soft-tissue augmentation, because of their advantages of noninvasiveness and easy injectability. An ideal filler is nontoxic, noncarcinogenic, inert, anallergic, nonimmunogenic, nonpyrogenic, and nonmigrating and must be similar to the native tissue—durable, easily implanted, and painless for the patient.[1]

These injectables are referred to as "filler materials." Just like dermal autografts and synthetic implants, these liquid fillers are meant to substitute for the lost subcutaneous skin supporting tissues and, hence, reduce wrinkles.[2] There are 3 different categories of injectable filler materials: biodegradable fillers such as plain hyaluronic acid and bovine collagen, which are effective for up to a few months; semipermanent fillers such as lactate acid and cross-linked hyaluronic acid, which are effective from a few months up to a year; and finally, permanent fillers such as polyalkylimide and polyacrylamide that are meant to last in situ for many years.[3] Hydrogels are a class of polymers that are very similar to soft tissues owing to their high water content, mechanical properties (very low modulus and elasticity), softness, oxygen permeability, and high biocompatibility.[1]

Although Bio-Alcamid has received European Union Certificate and Korea Food & Drug Administration approvals, it still carries the risk for complications such as inflammation and infection because it is not a natural component of biologic tissues. Despite such potential risks, a lack of awareness exists about the possible complications of long-acting filler materials, such as Bio-Alcamid, that are used indiscriminately.[4] Here, we report a case of a 26-year-old woman who underwent surgical treatment because of nasal septal abscess that developed 2 years after she was treated with a Bio-Alcamid injection in the nasal tip for cosmetic purposes.

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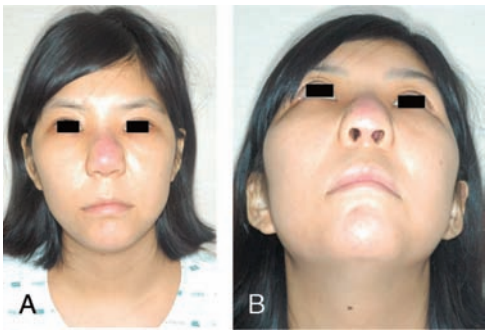

FIGURE 1. Facial photography (A, B) of a 26-year-old woman who presented with diffuse erythematous flare and swelling at the nasal tip and dorsum with progressive aggravating pain.

CLINICAL REPORT

A 26-year-old woman presented to our department with pain and swelling at the nasal tip that had manifested 3 days prior. This patient was referred from a local clinic because of exacerbated clinical manifestations after treatment with oral antibiotics. She had received a Bio-Alcamid filler injection into the nasal tip in a private clinic for cosmetic purposes 24 months before presenting to our department. She had no significant medical history. Physical examination revealed diffuse erythematous flare and swelling at the nasal tip and dorsum (Fig. 1) with concomitant pain, tenderness, and local heat. No lymph node enlargement or purulent rhinorrhea was observed.

The patient's white blood cell count was 12.0×10^3 cells/μL. No improvement was seen after the administration of cefazolin of 1 g intravenous 2 times a day and amikacin of 250 mg intramuscular 2 times a day for 3 days. A facial computed tomography scan revealed a nasal soft tissue swelling and septal abscess of $2.8 \times 1.7 \times 1.4\,cm^3$ (Fig. 2). Surgical incision and drainage were performed under general anesthesia. Methicillin-sensitive *Staphylococcus aureus* (*S. aureus*) strains were cultured at the time of surgery. When her symptoms and the lesion improved without any complications, she was discharged on the fourth postoperative day. Outpatient follow-ups conducted the next day and 1 week later showed no marked changes, and no recurrence has been encountered to date.

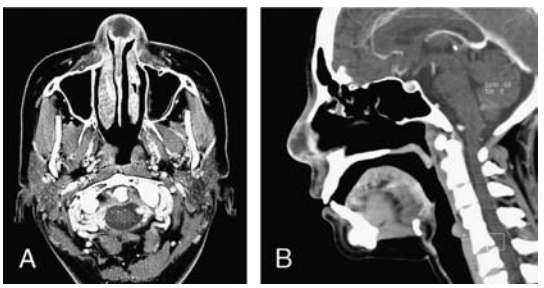

FIGURE 2. Facial computed tomography (A, B) imaging demonstrates a thin-walled, cystlike collection with peripheral enhancement involving the cartilaginous septum consistent with a nasal septal abscess.

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DISCUSSION

The adverse reactions associated with filler injection can be largely categorized into acute complications and delayed complications. Acute complications are those that present within a few days after treatment, such as pain, swelling, erythema, bleeding, purpura, and allergic reactions, and their etiologies may include infection by strains such as *S. aureus*, bulging masses induced by inadequate filler placement, pigmentation, and necrosis.[5,6] Delayed complications are those that present several weeks or even later after treatment and include filler migration, granulomatous infection, scarring, sterile abscesses induced by allergic reactions, and atypical mycobacterial infections.[7]

The foreign body response to polyalkylimide gel is thought to decrease with time and culminate in a low-grade host response at 6 months or no response at all at 2 years.

Polyalkylimide of 4% is viewed as an injectable liquid endoprosthesis. Hence, it is prone to the potential drawbacks of an endoprosthesis, such as excessive capsule formation, dislocation or migration, and infection. As with breast implants, injected polyalkylimide induces fibroblast proliferation and capsule formation. Capsule formation severity is unpredictable and varies among patients.[3] Infection can be triggered by additional revision procedures at or near the site of previous injections. However, complications may also arise spontaneously without any history of trauma or local infection.[8]

An early multicenter study including 2000 patients reported excellent cosmetic results with only 12 complications of *S. aureus* infection. This study also reported no observed cases of migration, dislocation, granulomas, allergic response, or implant intolerance.[3]

Recent reports have emerged from plastic surgery units that have become referral centers for Bio-Alcamid–related complications. Schelke et al conducted a retrospective review including 3196 patients treated with polyalkylimide in the Netherlands and reported a patient complication rate of 4.8%. Several other authors have emphasized the late appearance of abscesses months to years after material injection.[9]

In cases in which infection has occurred, more often than not, the foreign material must be removed surgically to adequately treat the infection. Surgical incision and drainage are usually required to remove all of the infected materials, resulting in undesirable facial scarring.

As these serious long-term complications of Bio-Alcamid emerge, several practitioners who previously advocated its use now strongly caution against it. Letters of concern regarding the use of Bio-Alcamid have been submitted to the National Institute of Clinical Excellence in the United Kingdom, although no recommendations have been published. The Dutch Society of Cosmetic Medicine considers the overall complication rate of polyalkylimide to be too high for cosmetic treatment and now advises against its use.[9]

Because we do not know how many patients have undergone additional treatments, we cannot estimate the occurrence rate of complications among this subgroup. Considering the potentially disfiguring therapy needed to treat these complications, we warn against the use of such permanent fillers for cosmetic indications.

Additional or secondary treatment in patients who previously had this nonresorbable filler material injected should be done under strict aseptic circumstances.[10]

CONCLUSIONS

Despite the safety and efficacy of recently developed fillers verified by early studies and their consequent wide use, much caution is required for the use of long-acting and permanent fillers for their long-term durability owing to the risk for complications. Therefore, physicians must explain the possible complications of these fillers to patients before treatment for reconstructive purposes. In addition, their cautious and limited use for esthetic purposes is recommended.

REFERENCES

1. Ramires PA, Miccoli MA, Panzarini E, et al. In vitro and in vivo biocompatibility evaluation of a polyalkylimide hydrogel for soft tissue augmentation. *J Biomed Mater Res* 2005;72:230–238
2. Pacini S, Ruggiero M, Cammarota N, et al. Bio-Alcamid, a novel prosthetic polymer, does not interfere with morphological and functional characteristics of human skin fibroblasts. *Plast Reconstr Surg* 2003;111:489–491
3. Karim RB, Hage JJ, van Rozelaar L, et al. Complications of polyalkylimide 4% injections (Bio-Alcamid): a report of 18 cases. *J Plast Reconstr Aesthet Surg* 2006;59:1409–1414
4. Liu HL, Cheung WY. Complications of polyacrylamide hydrogel (PAAG) injection in facial augmentation. *J Plast Reconstr Aesthet Surg* 2010;63:e9–e12
5. Xu JH, Zeng BW, Shen H, et al. A complication of polyacrylamide hydrogel injection in nasal dorsum augmentation. *Dermatol Surg* 2012;38:813–815
6. Goldan O, Georgiou I, Farber N, et al. Late-onset facial abscess formation after cosmetic soft tissue augmentation with bio-alcamid. *Aesthet Surg J* 2007;27:416–418
7. Chung WK, Park GH, Chang SE, et al. Case report: a case of mycobacterium chelonae infection with foreign body granuloma after injection of filler. *Korean J Dermatol* 2008;46:1521–1525
8. Schelke LW, van den Elzen HJ, Canninga M, et al. Complications after treatment with polyalkylimide. *Dermatol Surg* 2009;35 (suppl 2): 1625–1628
9. Salati SA, Aithan Ba. Multiple facial abscesses as an adverse outcome of cosmetic dermal filling: a case report. *East Cent Afr J Surg* 2011;16: 122–124
10. Nelson L, Stewart KJ. Early and late complications of polyalkylimide gel (Bio-Alcamid)(R). *J Plast Reconstr Aesthet Surg* 2011;64:401–404

Successful Treatment of Posttraumatic Arteriovenous Malformation of the Lower Lip

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Abstract: Arteriovenous malformations (AVMs) are most commonly reported in the brain. Head and neck AVMs are reported to occur in 0.1% of the general population. On the other hand, posttraumatic AVMs are quite rare. Traumatic AVMs are extremely

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rare in the head and neck and are mostly seen in the extremities. The management of such lesions may include selective embolization or surgical exploration with ligation.

A 13-year-old male adolescent visited our hospital for lower lip swelling, which developed 5 years ago after a lower lip laceration. The AVM was expanded and was graded as stage II. It was fed by the mandibular branch of both facial arteries and drained to the posterior facial vein. The radiologic intervention department performed an embolization before the operation. The main operation was performed 12 days after the embolization. A well-demarcated AVM lesion was found in the oral mucosa and was totally excised under general anesthesia. The authors easily performed the operation owing to the embolization making the AVM definitely demarcated and firmly palpable. The lip closure was done carefully considering the lip contour.

No sign of recurrence was seen during 6 months of follow-up. The excellent treatment result of the posttraumatic facial AVMs occurs largely because of a collaboration with the radiologic intervention department using the selective embolization.

Key Words: Arteriovenous malformation, trauma, head, and neck, embolization, surgical excision

Arteriovenous malformations (AVMs) are most commonly reported in the brain. Head and neck AVMs are reported to occur in 0.1% of the general population.[1] On the other hand, posttraumatic AVMs are quite rare.[1–4] Particularly, when it develops in the facial area, it can originate from the superficial temporal artery or the facial artery, which is close to the skin and superficial veins. In such cases, AVMs may cause a disfigurement of facial structures, and it can be quite challenging to achieve a good esthetic result after treatment.[1] Herein, we report a case of posttraumatic lower lip AVM, which developed after a lower lip laceration. The authors could get an excellent esthetic result because of the collaboration of embolization and surgical treatment.

BRIEF CLINICAL REPORT

A 13-year-old male adolescent visited our hospital with lower lip swelling, which developed 5 years ago after a lower lip laceration (Fig. 1). The mechanism of the injury was a penetration by own teeth. He did not take any treatment at that time, and the wound healed spontaneously. The size of the swelling increased during the past 5 years accompanied by a throbbing sensation. The volume of lower lip was enlarged temporarily by the Valsalva maneuver on physical examination. Our final diagnosis was confirmed by angiography (Fig. 2A). The AVM was expanded and graded as stage II (a clinical staging system), introduced by Schobinger in 1990.[5] It was fed by the mandibular branch of both facial arteries and being drained to the posterior facial vein. A tense and tortuous vein was observed, and the size of nidus was 3.1 × 4.4 cm. The radiologic intervention department performed an embolization before the operation. They superselected the mandibular branch of both facial arteries and injected 10-mL alcohol and 2-mL lipiodol on the right side and 4-mL alcohol and 1-mL lipiodol on the left side using a microcatheter (Fig. 2B). The AVM could not be visualized anymore after the embolization (Fig. 2C). Our operation was performed 12 days after embolization. A well-demarcated AVM in the oral mucosa was totally excised under general anesthesia (Fig. 3). The authors could perform the operation without any difficulties owing to the embolization that caused the AVM to definitely demarcate and to be firmly palpable due to fibrosis. Skin closure was carefully done considering the lip contour.

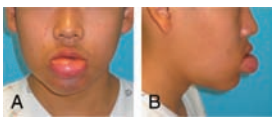

FIGURE 1. Preoperative photography. A 13-year-old male adolescent visited our hospital for lower lip swelling that developed 5 years ago after the treatment of a lower lip laceration.

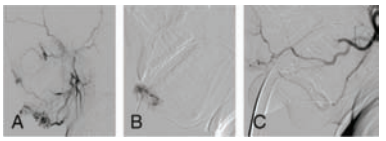

FIGURE 2. Angiography findings. A, Our final diagnosis confirmed an AVM due to angiography. The AVM was expanded and was graded as stage II. It was fed by the mandibular branch of both facial arteries and drained to the posterior facial vein. A tense and tortuous vein was observed, and the size of nidus was 3.1 × 4.4 cm. B, The radiologic intervention department superselected the mandibular branch of both facial arteries and injected via microcatheter 10-mL alcohol and 2-mL lipiodol at the right side as well as 4-mL alcohol and 1-mL lipiodol at the left side. C, After embolization, no AVM is visualized any more.

The final lower lip contour was esthetically excellent, and there was no development of any problem such as a circulation compromise, lip volume mismatch (overcorrection or undercorrection), or abnormal lip movement (Fig. 4). The patient was very satisfied, and no recurrence was seen during the 6 months of follow-up.

DISCUSSION

Head and neck AVMs are reported to occur in 0.1% of the population. On the other hand, posttraumatic AVMs in the head and neck region are quite rare.[1,6] An AVM is an endothelial-lined communication between an artery and vein without an interposed capillary bed. The communication channel can be single or multiple.[1] Of AVMs, the most common cause is congenital, and they usually occur in the head and neck area with multiple-channeled feeding vessels. In contrast, posttraumatic AVMs usually occur in extremities. In most cases of posttraumatic AVMs, feeding vessels have a single channel. So, the treatment of them may look easier than in congenital AVMs. However, if posttraumatic AVMs have been formed in the facial area, minimizing the disfigurement of esthetic unit must be considered so treatment could be rather difficult (Table 1).[1,6]

Of the posttraumatic AVMs, the most common feeding vessel is the superficial temporal artery (40%), followed by the facial artery (28%). Origins include postsurgical wounds (36%) and penetrating wounds (32%), and there was a report of formation of AVM by a

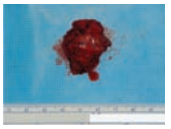

FIGURE 3. The excised AVM mass. A well-demarcated AVM that invaded the oral mucosa was totally excised under general anesthesia.

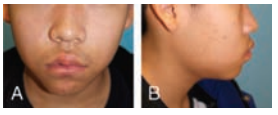

FIGURE 4. Postoperative photography. The final lower lip contour was esthetically excellent, and no problem related to the circulation of the lower lip was developed as concerned before. The patient was very satisfied with the result, and no recurrence was seen during 6 months of follow-up.

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TABLE 1. A Comparison of the Congenital AVMs and Traumatic AVMs

	Congenital AVMs	Traumatic AVMs
Location	Common in the head and neck	Rare in the head and neck
	Most common in the cranium	Most common in the extremities
Feeding vessel	Mostly multiple	Mostly single
Treatment	Selective embolization and surgical excision	Selective embolization and surgical excision

blunt trauma without an external wound.[1] In our presented case was the facial artery the feeder, and we think that having the original wound spontaneously healed instead of primary closure in a layer-by-layer manner could have some influence on the formation of this lesion. Therefore, a meticulous bleeding control and avoiding a rough closure are important during an operation near the superficial temporal artery or facial artery.

The symptoms of AVM include pulsatile tinnitus on the side of the lesion, progressive enlarging mass, throbbing sensation (aggravated by the Valsalva maneuver), bleeding, and palpable thrill without bruit.[1,6] The AVMs ought to be considered in the differential diagnosis if symptoms as mentioned previously appear after operation or trauma.

An AVM could be treated by a combination of surgical resection, embolization, and sclerotherapy according to the condition of the lesion. So far, an embolization followed by surgical resection has shown the highest cure rate.[7] Surgical resection is usually done within 72 hours after embolization. Otherwise, an inflammatory process can make the surgical approach tough.[7,8] So the surgery should be planned within 72 hours if solid materials such as Gelfoam, fat, muscles, or coils are used. On the other hand, temporary occlusive substances such as polyvinyl alcohol particles or 100% of alcohol also could be used for the preoperative embolization. In this situation, the excision should be done at least 72 hours after the embolization to achieve the maximal reduction in blood flow to the central portion of the vascular lesion.[9] In our case, we performed the operation after more than 72 hours because we used alcohol as the main embolization agent, and therefore, the lesion was found to be solid consolidated and well demarcated so that we could remove it with ease. There are various embolization agents that require a different interval time for operation. So, the patient can adjust his/her schedule by choosing the proper embolization agent with the medical team. Close communication between surgical team and intervention team is important for the selection of the proper agent.

The AVM lesion can be easily collapsed by tracting or pressing, which obscures the mass margin if the operation is performed without embolization. In addition, bleeding could interrupt the operation field, making an en bloc resection hard. On the contrary, an AVM does not easily collapse by tracting or pressing and can be palpated firmly like a hard mass, if the operation is performed after inducing fibrosis with alcohol such as in the presented case. In addition, the lesion becomes well demarcated, which makes the total excision easy.

If an AVM is located at an orbit or periorbital area, an embolization could obstruct the central retinal artery, which may result in blindness.[1] So, it must be done with caution. Furthermore, a neovascularization can be recruited from the adjacent arteries if an embolization is done proximal to the feeding vessel by mistake. It means competent intervention skills of the physician are also crucial for good treatment results.

In conclusion, traumatic AVMs are quite rare in the head and neck area and are most common in the extremities. The management of such lesions may include selective embolization or surgical exploration with ligation. The authors could get an excellent treatment result for a posttraumatic facial AVM that usually makes it troublesome for the operator. The result was reached because of collaboration with the radiologic intervention department using the selective embolization.

ACKNOWLEDGMENT

This study was carried out under the approval of institutional review board of the Catholic Medical Center of Korea.

REFERENCES

1. Holt GR, Holt JE, Cortez EA, et al. Traumatic facial arteriovenous-malformations. *Laryngoscope* 1980;90:2011–2020
2. Stocks HM, Sacksman S, Thompson P, et al. First documented case of a submandibular arteriovenous malformation in a male patient. *Am J Otolaryngol* 2007;28:275–276
3. Ogbole GO, Ogunseyinde AO, Obajimi MO, et al. Extracranial arteriovenous malformation of the scalp: value of computed tomographic angiography. *Internet J Radiol* 2006;5:1–4
4. Adme N Jr, Bayless P. Carotid arteriovenous fistula in the neck as a result of a facial laceration. *J Emerg Med* 1998;16:575–578
5. Mulliken JB. Vascular anomalies. In: Aston SJ, Beasly RW, Thorne CHM, eds. *Grabb and Smith's Plastic Surgery.* Philadelphia: Lippincott-Raven; 1997:191–204
6. Kao ST, Walker PW, Ferguson HW. Posttraumatic arteriovenous malformation of the face: a case report. *J Emerg Med* 2014;46:e5–e8
7. Manganaro AM, Will MJ, Rafetto LK. Rapidly enlarging lesion of the upper lip. *J Oral Maxillofac Surg* 2000;58:883–887
8. Han MH, Seong SO, Kim HD, et al. Craniofacial arteriovenous malformation: preoperative embolization with direct puncture and injection of n-butyl cyanoacrylate. *Radiology* 1999;211:661–666
9. Marx R. Oral and maxillofacial pathology: a rationale for diagnosis and treatment. 1st ed. Hanover Park: Quintessence Publishing Co, Inc; 2003

Frontal Endoscopic Myotomies for Chronic Headache

Raposio Edoardo, MD, PhD, and Caruana Giorgia, MD

Abstract: Recent insights into the pathogenesis of migraine headache substantiate a neuronal hyperexcitability and inflammation involving compressed peripheral craniofacial nerves, and these trigger points can be eliminated by surgery. The aim of this study was to describe a modified, innovative, minimally invasive endoscopic technique to perform selective myotomies of corrugator supercilii, depressor supercilii, and procerus muscles, which turned

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out to be an effective therapy for migraine and tension-type headaches.

Forty-three patients (18–75 years) who experienced 15 or more frontal migraine headaches without aura, tension-type headaches, or new daily persistent headaches each month were enrolled in the study between 2011 and 2013. Of 43 patients, 15 were followed for 2 years. Fourteen patients (93.3%) reported a positive response to the surgery: 5 (33.3%) observed complete elimination, 9 (60%) experienced significant improvement (at least 50% reduction in intensity or frequency), and 1 patient (6.6%) did not notice any change in their headaches. A statistically significant difference was found between our protocol compared with currently performed, more invasive technique (odds ratio, 1.9; 95% confidence interval, 1.151–3.13).

According to our data, the modified endoscopic procedure leads to better results, compared to previous techniques, together with eliminating the need for general anesthesia, reducing the invasiveness of the procedure and the number of postoperative scars.

Key Words: Migraine surgery, facial endoscopic surgery, headache, mininvasive plastic surgery

During the past decades, endoscopic techniques have provided surgical options applied to aesthetic plastic surgery in very different fields[1–5]; recent studies showed that endoscopic techniques also succeeded in improving migraine and tension-type headaches,[6,7] hence expanding the indications of this surgical approach.

Recent insights[8–10] into the pathogenesis of migraine headache substantiate the possible role of neuronal hyperexcitability and inflammation involving compressed peripheral craniofacial nerves; these trigger points could be eliminated by surgery, which thus might provide a therapeutic approach to migraine. The peripheral mechanism of migraine headache was first proposed by Guyuron et al[6]: based on patients whose migraine headaches disappeared after forehead rejuvenation, they first reported an association between forehead muscles resection and the elimination or significant improvement of migraine headaches.[8] The currently adopted procedure,[11,12] performed under sedo-analgesia, relies on 3 to 5 1.5-cm-long access incisions 1 cm behind the anterior hairline, and it is performed by means of an endoscope and a dissector. Since September 2011, we have been using a minimally invasive endoscopic selective myotomy technique, performed by a specifically modified endoscope and under local anesthesia, in an effort to reduce the invasiveness of the currently adopted techniques.

MATERIALS AND METHODS

Patient Selection

Patients in good health (age range, 18–75), with frontal migraine headaches without aura, tension-type, or new daily persistent headaches not responding to medications, were considered eligible for the study. All patients' condition had been diagnosed by board-certified neurologists as outlined by the International Classification of Headache Disorders II criteria.[13] The patients completed a comprehensive validated headache questionnaire before and after the surgery to obtain a comparison of outcome of surgical treatment using this procedure versus prior protocol. The pretreatment questionnaire collected data including age, sex, age at onset, headaches per month, and so on (see Supplemental Digital Content, Headache Questionnaire, http://links.lww.com/SCS/A136). The postoperative evaluation (6 months and 2 years after surgery) included additional information, such as degree of reduction of headache or medication usage (see Supplemental Digital Content, Headache Questionnaire, http://links.lww.com/SCS/A136). Statistical analysis was performed by means of odds ratio (OR) evaluation, standard error for the log OR (Woolf method), and 95% confidence interval of OR (Woolf method).

Surgical Procedures

All procedures were performed by a single surgeon (E.R.) under local anesthesia as a one-day surgery procedure. Skin markings were performed in the glabellar region, bilaterally along both midpupillary line and 1 cm medially. After bilateral selective supratrochlear and supraorbital nerve block, diluted carbocaine 1% + 8.4% sodium bicarbonate were infiltrated throughout the entire forehead. Our approach is to perform the whole procedure by only one 2-cm-long midline cutaneous incision just behind the anterior hairline (Fig. 1). After subgaleal dissection, 3 surgical sutures (1–0 nylon) were placed bilaterally in the superciliary region, at each side of both supratrochlear and supraorbital nerves, to lift the frontal skin to better visualize the nerves and surrounding muscles (Fig. 2). Endoscopically assisted section of the corrugator supercilii, depressor supercilii, and procerus muscles bilaterally (one myotomy per side of each nerve) was carried on by means of a modified endoscope (Fig. 3) to decompress the supraorbital and supratrochlear nerves, which were not injured during dissection (Figs. 4–6). Our modified endoscope (Karl Storz, Tuttlingen, Germany) consists of a 9-mm trocar, a straight Hopkins telescope with fiber light transmission, a Wittmöser operating sheath with a connection for high-frequency diathermy, and a specifically designed elliptical-tipped wire loop electrode for the electrocautery. Full-thickness (to reach the subcutaneous tissue) myotomies, always parallel and approximately 2 mm medially and

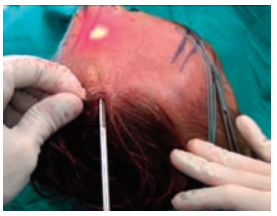

FIGURE 1. One 2-cm-long midline cutaneous incision endoscope entry.

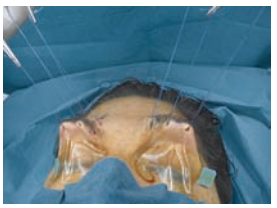

FIGURE 2. Three surgical sutures (on each side) placed in the superciliary region to lift the frontal skin.

FIGURE 3. The endoscope used to perform the minimally invasive myotomy procedure.

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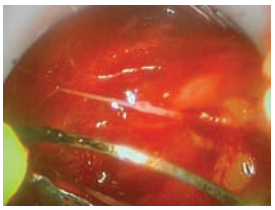

FIGURE 4. Approaching left superciliary muscles, between left supraorbital and supratrochlear nerves.

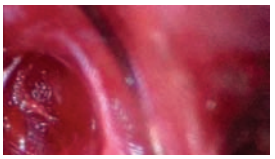

FIGURE 5. Myotomy lateral to supraorbital nerve.

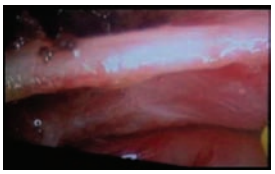

FIGURE 6. Decompressed right supraorbital nerve.

laterally to each nerve, were performed. After completion of the procedure, cutaneous sutures (4–0 nylon) were placed.

RESULTS

Follow-up

Of the 43 patients included in the study (range, 18–72 years), 38 were women and 5 were men. All the 43 patients underwent surgery: 43 patients were followed for 6 months and 15 of them for 2 years. After a 6-month-long follow-up, we obtained the following results: 17 (39.5%) of the 43 patients observed complete elimination, 18 (41.9%) experienced significant improvement (at least 50% reduction in intensity or frequency), and 8 (18.6%) did not notice any change. The minimally invasive approach yielded positive response in 35 (81.4%) of the 43 patients, confirming the validity of the procedure.

After a 2-year-long follow-up, our approach yielded positive response in 14 (93.3%) of 15 patients: 5 (33.3%) of 15 patients observed complete recovery, 9 (60%) of 15 patients experienced significant improvement (at least 50% reduction in intensity or frequency), and 1 (6.6%) did not notice any change.

TABLE 1. Comparison Between Our Surgery Protocol Outcomes and Previous Surgery Protocol Outcomes

	Benefit (%)	No Benefit (%)
Our protocol	93.3	6.6
Previous protocol	88	12

OR, 1.9; ES [log(OR)], 0.256; 95% confidence interval (OR), 1.151–3.13.

We compared our 2-year results with data obtained by Guyuron et al[14] after a 5-year follow-up. According to our statistical analysis, our technique led up to a statistically significant increased possibility of obtaining a positive response, compared with the potential of not reporting any benefit from surgery (OR, 1.9; 95% confidence interval, 1.151–3.13; Table 1). No statistically significant difference was found comparing the adverse effects between the 2 surgical approaches (Table 1).

These results mimic what has been reported in other previous studies[14] and show a statistically significant improvement compared with them; at any rate, we believe that more studies with a major number of patients are needed to assess statistical results with a proper and more consistent population.

DISCUSSION

This study was designed to compare our results with those reported by a previous study[14] as well as to assess the long-term success of surgical deactivation of migraine headache trigger points. One reason why not every patient reached full recovery might be that the operation was performed on a single predominant trigger site, which could be not the only one. Furthermore, there is the possibility that some scarring tissue might compress again the nerves, causing the failure of surgery.

Since the operation did not cause any complication, it can be recommended to patients with chronic migraine, particularly patients whose headaches are poorly controlled by pharmacologic means. The minimally invasive procedure we described is easy, fast, and cost-effective, relying on the use of a single instrument, also reducing the numbers of postoperative scars to only one.

REFERENCES

1. Caleel RT. Transumbilical endoscopic breast augmentation: submammary and subpectoral. *Plast Reconstr Surg* 2000;106:1177–1182
2. Hansen TB, Majeed HG. Endoscopic carpal tunnel release. *Hand Clin* 2014;30:47–53
3. Swift RW, Nolan WB, Aston SJ, et al. Endoscopic brow lift: objective results after 1 year. *Aesthet Surg J* 1999;19:287
4. Niamtu J 3rd. Endoscopic brow and forehead lift: a case for new technology. *J Oral Maxillofac Surg* 2006;64:1464
5. Papadopulos NA, Eder M, Weigand C, et al. A review of 13 years of experience with endoscopic forehead-lift. *Arch Facial Plast Surg* 2012;14:336–341
6. Guyuron B, Varghai A, Michelow BJ, et al. Corrugator supercilii muscle resection and migraine headaches. *Plast Reconstr Surg* 2000;106:429–434
7. Bearden WH, Anderson RL. Corrugator superciliaris muscle excision for tension and migraine headaches. *Ophthal Plast Reconstr Surg* 2005;21:418–422
8. Guyuron B, Kriegler JS, Davis J, et al. Comprehensive surgical treatment of migraine headaches. *Plast Reconstr Surg* 2005;115:1–9
9. Dirnberger F, Becker K. Surgical treatment of migraine headaches by corrugator muscle resection. *Plast Reconstr Surg* 2004;114:652–657
10. Poggi JT, Grizzel BE, Helmer SD. Confirmation of surgical decompression to relieve migraine headaches. *Plast Reconstr Surg* 2008;122:115–122
11. Guyuron B, Tucker T, Davis J. Surgical treatment of migraine headaches. *Plast Reconstr Surg* 2002;109:2183–2189
12. Guyuron B, Reed D, Friegler JS, et al. A placebo controlled surgical trial for the treatment of migraine headaches. *Plast Reconstr Surg* 2009;124:461–468
13. Headache Classification Committee of the International Headache Society. Classification and diagnostic criteria for headache disorders, cranial neuralgias and facial pain. *Cephalgia* 1988;8 (suppl 7):1–96
14. Guyuron B, Kriegler J, Davis J, et al. Five-year outcome of surgical treatment of migraine headaches. *Plast Reconstr Surg* 2011;127:603

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Alternatives for Rehabilitation of Cleft Patients With Severe Maxillomandibular Discrepancy

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Abstract: Rehabilitation of cleft patients is a process that occurs from birth to adult life and involves a team of many professionals. Reconstructive plastic surgery, despite its functional and aesthetic benefits to the patient, can restrict the normal anterior displacement that occurs in the growth of the maxilla, which, in turn, can lead to a concave profile that requires correction. This study aimed to demonstrate an alternative rehabilitation treatment for cleft patients who have severe maxillomandibular discrepancy and choose not to undergo orthognathic surgery. A retrospective review and case reports of rehabilitation treatment of cleft patients were performed, with an emphasis on prosthetic rehabilitation without orthognathic surgical procedures. Prosthetic rehabilitation is a fast and reversible option for cleft patients that provides facial harmony and facilitates the reintegration of these patients into society.

Key Words: Oral rehabilitation, removable partial denture, cleft lip, cleft palate

Rehabilitation of cleft patients is a process that occurs from birth to adult life and involves a team of many professionals.[1–3]

Appropriate facial aesthetics is a concern of dentists because most cleft patients carry the stigma of a congenital deformity that affects the aesthetic, functional, and psychologic aspects of daily life.[2,4]

Reconstructive plastic surgery (eg, cheiloplasty and palatoplasty), despite its functional and aesthetic benefits to the patient, can restrict the normal anterior displacement that occurs in the growth of the maxilla, which can, in turn, lead to a concave profile that requires correction.[5]

Orthodontics is a key area for the rehabilitation of cleft patients who require repositioning of the teeth in the bony bases, thereby allowing subsequent surgery to reposition the bony bases, restoring the function and the oral health of the patient, and improving the aesthetics.[5,6]

Significant factors that must be taken into consideration include the patient's ability to participate in all stages of treatment and delayed demand for treatment, both of which can prevent timely completion of surgery; thus, it is necessary to develop alternative forms of treatment beyond the ideal method of rehabilitation.

In an attempt to circumvent these complications, overlay prostheses may be used; these are intended to correct the maxillomandibular discrepancies present in individuals who express a desire for a simplified treatment or do not wish to undergo orthognathic surgery and/or orthodontic treatment.[7,8]

The aim of this study was to disseminate alternative possibilities for the treatment of cleft patients to the scientific community; these alternatives include prosthetic treatment, which has achieved satisfactory cosmetic and functional results.

METHODOLOGY

This study was reviewed and approved by the institutional review board.

A retrospective study and clinical case reports on rehabilitation treatment of cleft patients were performed, with an emphasis on prosthetic rehabilitation without orthognathic surgical procedures.

All patients were informed about the use of their images and signed an informed consent form that permitted the use of their images for scientific purposes.

CLINICAL REPORT AND DISCUSSION

Correction of maxillomandibular discrepancies in cleft patients is usually performed through orthodontic treatment that may or may not be supplemented with orthognathic surgery.[9,10]

The ideal protocol is to start treatment early in life, perform all of the corrective surgical steps, and finish the dental rehabilitation with orthodontia or dental prostheses.[6]

Rehabilitation of edentulous areas can be achieved with conventional fixed partial dentures, removable partial dentures, implant-supported prostheses, or overlays. The choice is based on the size of the edentulous space, the condition of the adjacent teeth, as well as the patient's periodontal health and opinion.[11]

Patient 1: Finished With Orthodontics and Orthognathic Surgery

Patient 1 underwent orthodontic treatment before surgical correction of maxillomandibular discrepancy. The quality of the outcome with regard to aesthetics and occlusion was satisfactory (Figs. 1–2).

Orthodontic preparation should be performed before orthognathic surgery; the opposite sequence of treatment is not feasible, and an alternative treatment is necessary to correct the maxillomandibular discrepancy and rehabilitate the edentulous area.[12,13]

FIGURE 1. Initial appearance.

FIGURE 2. Final frontal view.

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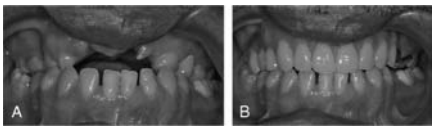

FIGURE 3. Frontal view: A, Without prosthesis. B, With prosthesis.

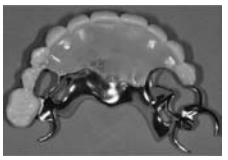

FIGURE 4. Overlay prosthesis: occlusal view.

Patient 2: Finished With Overlay Prosthesis

Patient 2, who had a considerable maxillomandibular discrepancy, chose nonsurgical rehabilitation, and the occlusion was corrected with an overlay prosthesis (Figs. 3–5).

The so-called conventional removable partial denture can only be used to restore partially edentulous regions. However, overlays have the ability to restore an edentulous arch and to correct severe maxillomandibular discrepancies because they allow the mounting of the restorations over the remaining teeth. Thus, it was possible to return the patient to a normal overbite and overjet as well as to improve the proportionality of the patient's facial appearance.[7]

Patient 3: Finished With Overlay Prosthesis

Because of limitations in surgical and orthodontic treatment based on the extent of the cleft palate, the rehabilitation of patient 3 was finished with only an overlay. On the basis of the young age of the patient, a temporary overlay was chosen, and it was planned to be changed periodically to avoid preventing bone growth. The prosthesis was combined with palatal extension to ameliorate velopharyngeal dysfunction[14] (Figs. 6–8).

In some cases, the discrepancy is so great that surgery alone is not sufficient to fully correct the patient's occlusion and an overlay prosthesis is required even after orthognathic surgery.

Patient 4: Finished With Orthognathic Surgery and Overlay Prosthesis

Patient 4 underwent all of the steps of corrective surgery at the appropriate times, but owing to the large extent of the patient's maxillomandibular discrepancy, an overlay was required to rehabilitate his occlusion even after orthognathic surgery (Figs. 9–11).

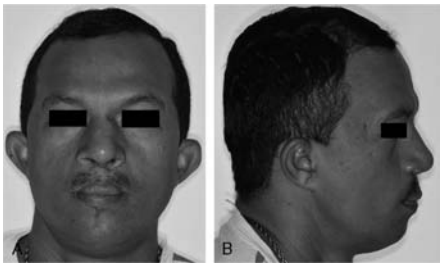

FIGURE 5. Final appearance. A, Frontal view. B, Lateral view.

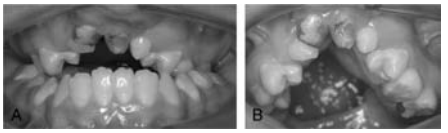

FIGURE 6. Initial appearance: A, Frontal view. B, Palatal view.

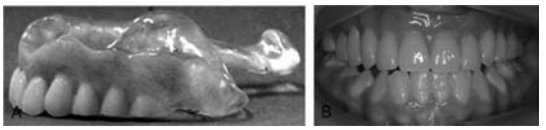

FIGURE 7. A, Overlay prosthesis with palatal extension. B, Final frontal view.

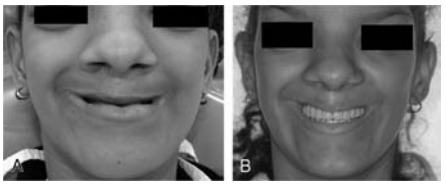

FIGURE 8. Final appearance. A, Without prosthesis. B, With prosthesis.

FIGURE 9. Initial lateral view. Note the extent of the discrepancy.

Similar types of treatment have been reported in the literature. Fragoso et al[15] reported a case in which an overlay was placed over the lingual part of the anterior teeth to restore the vertical dimension of occlusion, thus improving the patient's masticatory function and comfort.

One thing that must be considered when using an overlay prosthesis is the patient's oral hygiene. Once the teeth are covered, the susceptibility to cavities increases considerably, so the patient must be made aware of his/her new condition and be informed that he/she will have to redouble his/her care with brushing and flossing; otherwise, the teeth may be lost.[16]

CONCLUSIONS

Cleft patients experience the stigma of having a physical problem that affects not only aesthetics and function but also psychologic factors.

Aesthetic dental rehabilitation, especially with overlays, provides facial harmony that is typically only achieved with highly complex surgeries, which can restrict some patients from being rehabilitated. The main advantages of the option of using overlays

FIGURE 10. Final frontal view.

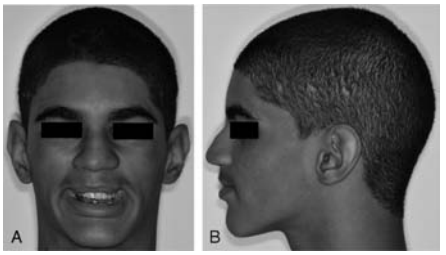

FIGURE 11. Final appearance. A, Frontal view. B, Lateral view.

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are that the treatment is fast as well as reversible and that it does not prevent these individuals from being treated with different modalities at any time throughout their lives.

REFERENCES

1. Salyer KE, Xu H, Portnof JE, et al. Skeletal facial balance and harmony in the cleft patient: principles and techniques in orthognathic surgery. *Indian J Plast Surg* 2009;42:149–167
2. Freitas JA, das Neves LT, de Almeida AL, et al. Rehabilitative treatment of cleft lip and palate: experience of the Hospital for Rehabilitation of Craniofacial Anomalies/USP (HRAC/USP)—part 1: overall aspects. *J Appl Oral Sci* 2012;20:9–15
3. Klassen AF, Tsangaris E, Forrest CR, et al. Quality of life of children treated for cleft lip and/or palate: a systematic review. *J Plast Reconstr Aesthet Surg* 2012;65:547–557
4. Wehby GL, Cassell CH. The impact of orofacial clefts on quality of life and healthcare use and costs. *Oral Dis* 2010;16:3–10
5. Silva Filho OG, Ferrari Junior FM, Carvalho RM, et al. A cirurgia ortognatica na reabilitação do paciente portador de fissura unilateral completa de lábio e palato. *Rev Dent Press Ortodon Ortop Facial* 1998;3:51–70
6. Cavassan AO, Silva Filho OG. Abordagem ortodôntica. In: Trindade IEK, Silva Filho OG, eds. *Fissuras Labiopalatinas: Uma Abordagem Interdisciplinar.* 1st ed. Sao Paulo: Editora Santos, 2007:213–238.
7. Rizoto A, Ribeiro MCM, Ferreira AR, et al. Reabilitação de paciente portador de fissura através de prótese de recobrimento—relato de caso clínico. *Rev Bras Prót Clín Lab* 2000;2:22–26
8. Lopes MMW, Bonfante G, Lopes JFS, et al. Próteses parciais Removíveis Não—Convencionais Empregadas no Hospital de Anomalias Craniofaciais-USP-Bauru. *Rev Bras Prót Clín Lab* 2002;4:420–426
9. Manna F, Pensiero S, Clarich G, et al. Cleft lip and palate: current status from the literature and our experience. *J Craniofac Surg* 2009;20:1383–1387
10. Ruiter A, Van der Bilt A, Meijer G, et al. Orthodontic treatment results following grafting autologous mandibular bone to the alveolar cleft in patients with a complete unilateral cleft. *Cleft Palate Craniofac J* 2010;47:35–42
11. Pinto JHN, Lopes JFS. Reabilitação oral com prótese dentária. In: Trindade IEK, Silva Filho OG, eds. *Fissuras Labiopalatinas: Uma Abordagem Interdisciplinar.* 1st ed. Sao Paulo: Editora Santos; 2007:261-274.
12. Proffit WR, Sarver DM, White RP. *Tratamento contemporâneo de deformidades dentofaciais.* Porto Alegre: Artmed, 2005:783.
13. Phillips JH, Nish I, Daskalogiannakis J. Orthognathic surgery in cleft patients. *Plast Reconstr Surg* 2012;129:535e–548e
14. Lopes JFS, Pinto JHN, de Almeida AL, et al. Cleft palate obturation with Brånemark protocol implant-supported fixed denture and removable obturator. *Cleft Palate Craniofac J* 2010;47:211–215
15. Fragoso WS, Troia MGJ, Valdrighi HC, et al. Reabilitação oral com prótese parcial removível overlay. *RGO* 2005;53:243–246
16. Preshaw PM, Walls AW, Jakubovics NS, et al. Association of removable partial denture use with oral and systemic health. *J Dent* 2011;39: 711–719

To Computer-Aided Design and Manufacturing or Not to Computer-Aided Design and Manufacturing? Free Fibula Flap With Computer-Aided Technique for Mandibular Reconstruction

Emanuele Zavattero, MD, Paolo Garzino-Demo, MD, Massimo Fasolis, MD, and Guglielmo Ramieri, MD, DDS

Aim: The purpose of this study was to analyze the accuracy of computer-assisted free fibula flap for reconstruction of large mandibular defects for benign tumors.

Materials and Methods: Between December 2012 and January 2014, a total of 4 free osteocutaneous computer-assisted fibula flaps have been used in an equal number of patients for reconstruction of the mandible at the Division of Maxillofacial Surgery, Città della Scienza e della Salute Hospital, University of Turin. Inclusion criteria were large mandibular defects due to benign tumors. The computer-assisted fibula flap was chosen when 2 or more osteotomies were requested. Intraoperative complication, mean ischemia time, operative time, and morphologic outcomes were analyzed in all cases. After surgery, a postoperative computed tomography compared the virtual plan with the surgical results.

Results: All 3 flaps were harvested and transplanted successfully. For the free flaps examined in this work, no intraoperative complications were noted. Postoperative computed tomography showed high correspondence in terms of bone contour according to the virtual plan.

The immediate and long-term morphologic results were satisfactory.

Conclusions: Microsurgical mandible reconstruction using a computer-assisted fibula flap technique is the best available method to manage complex defects.

Key Words: Mandible reconstruction, fibula flap, CAD/CAM, computer-aided surgery

Mandible reconstruction remains one of the challenges faced by the reconstructive maxillofacial surgeon. The most common indication for mandibular reconstruction remains ablative surgery for tumors involving the mandible.[1]

Several methods have been proposed for reconstruction using nonvascularized bone grafts or alloplastic implants.[1–6]

However, with the advent of microsurgery and since the pioneering work of Hidalgo[7] in 1989, the fibula free flap has become the workhorse for microvascular mandible reconstruction. The fibula free flap had some advantages including a long vascular pedicle and the possibility of multiple osteotomies to recreate the mandibular contour.

In the 1990s, computer-generated three-dimensional models were introduced for the preoperative planning and plate prebending, but only over the last decade, virtual reality technique was introduced to manipulate bone segments.[8–10]

Recently, computer-aided design and manufacturing (CAD/CAM) technologies were introduced for osseous free flap

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reconstruction with the advantages of obtaining more precise reconstruction operating time, especially in complex defects.[11]

The purpose of this study was to investigate the accuracy of computer-assisted fibula free flap technique for large complex mandibular defects.

Our attention was focused on the reliability of the virtual planning and on the predictability of the outcome.

MATERIALS AND METHODS

Four consecutive patients who underwent computer-assisted free osteocutaneous fibula flaps for reconstruction of the mandible were retrospectively reviewed.

The etiology of the osseous defect included ameloblastoma, fibrous dysplasia, ossifying fibroma, and keratocystic tumor.

Inclusion criteria were mandibular benign tumors with complex defects, where 2 or more osteotomies were required.

Surgical Planning

Spiral computed tomographic (CT) data sets (SOMATOM Sensation 16 scanner; Siemens, Erlangen, Germany), with 1-mm slice thickness and 0-degree gantry tilt, were acquired for preoperative planning and surgery on a computer.

Images in Digital Imaging and Communications in Medicine format were sent using a compact disc to the manufacturer (Synthes Maxillofacial) for processing.

In a Web conference with the engineer, the surgeon planned virtual tumor resection and virtual reconstruction with the aid of software ProPlan CMF.

Virtual reconstruction was carried out by placing fibular osteotomies to recreate the native facial contour.

Once the virtual planning was completed, custom-made surgical guides for the planned resection and fibular cutting guides were designed.

A prebent titanium plate was also generated, precisely matching the contours of the native mandible and interposed fibula flap.

The surgeon reviewed and approved the guides.

Surgical Procedure

The fibula flap was harvested according to the standard lateral approach described by Gilbert.[13]

During surgery, the sterilized cutting guide was temporarily fixed to the harvested fibula bone using monocortical screws and a reciprocating saw blade was inserted into slots in the cutting guide to make osteotomies at the lengths and angles required to replicate the virtual plan (Figs. 1A, B). The tumor was also resected using cutting guides to have correct shape and angle to fix the flap (Fig. 1C).

The vascular pedicle was then divided and then transferred.

The fibular segments were then fixated with the prebent titanium plate at the mandible as planned (Fig. 2).

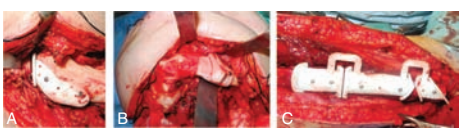

FIGURE 1. A and B, Intraoperative placement of cutting guides, which was temporarily fixed to the bone using 6-mm monocortical screws. A reciprocating saw blade was inserted into slots in the cutting guide to make osteotomies at the lengths and angles required to replicate the virtual plan. C, Intraoperative placement of cutting guides, which was temporarily fixed to the bone using 6-mm monocortical screws. A reciprocating saw blade was inserted into slots in the cutting guide to make osteotomies at the lengths and angles required to replicate the virtual plan.

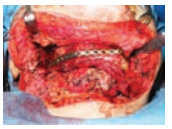

FIGURE 2. Intraoperative placement of a prebent titanium plate to fix the fibular segments.

The data collection included general information about the patient, surgical indications, location, and extension of bone defect, intraoperative complications, mean ischemia time (time from pedicle division to reanastomosis) and operative time (time from skin incision to closure), postoperative complications, healing, and follow-up.

Perioperative and long-term outcomes were assessed through clinical examination and CT scans (axial, coronal, and three-dimensional view).

The minimum follow-up period was 6 months.

This study was performed in agreement with the local institutional review board. This study followed the Declaration of Helsinki guidelines.

RESULTS

Patient's characteristic, defect classification, and planned reconstruction are summarized in Table 1.

All the flaps were harvested and transplanted successfully. All patients had a 2-day average intensive care unit stay.

All patients were discharged from the hospital within 25 days. No intraoperative complications were observed.

Patient 3 had delayed wound healing at the donor site.

Mean ischemia time was 75 minutes and mean operative time was 6 hours.

The postoperative CT images showed that the bone continuity was good in all cases and that the contour was resected at all the virtual reconstruction planned (Figs. 3,4).

The virtual planning was reliable, and no changes had to be made in the operating room.

The immediate and long-term morphologic results were satisfactory.

These cases highlight the potential ability of CAD/CAM technologies applied to microsurgical technique to more

TABLE 1. Patient's Characteristic, Defect Classification, and Planned Reconstruction

Patient	Age (yr)	Sex	Diagnosis	Tumor site	Planned resection	Planned reconstruction
1	37	F	Ossifying fibroma			
2	28	F	Fibrous displasia			
3	38	F	Ameloblastoma			
4	48	F	Keratocystic tumor			

*P value shows the results of the χ^2 test.

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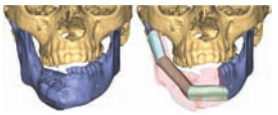

FIGURE 3. Preoperative CT scan of a 38-year-old woman presenting with a mandibular ameloblastoma and virtual planning.

comprehensively restore complex large mandibular defects using all the bone flaps available and improving the contour shape.

DISCUSSION

Mandibular reconstruction after trauma and pathology is one of the cornerstones of maxillofacial surgery.

Historically, various nonvascularized bone graft and alloplastic materials were responsible for inadequate functional and aesthetic results, showing a high rate of failure due to infection, reabsorption, and poor recipient vascular bed.[1,2,4,5]

Although in the literature, there are many donor sites for mandibular reconstruction. The fibula osteocutaneous free flap is the workhorse donor site in our center. This flap was originally described in 1989 by Hidalgo,[7] and it has become the flap of choice for the restoration of mandibular defects. Indications for the fibula osteocutaneous flap include total or subtotal mandibulectomy, bone-only defects, and reconstruction of the subcondylar-condylar complex.

The technique of flap harvest is relatively straightforward. As summarized by Gbara et al,[11] the advantages in harvesting this flap are the following: (1) its straight shape and high resistance to pressure and torsion; (2) rapid healing due to excellent perfusion; (3) its high content of cortical bone; (4) its great length, which allows the bringing of large defects; (5) the possibility of osteomizing it in various points; (6) its convenient location and 2-team approach can be done; (7) adjacent soft tissue available; (8) it is indicated for all anterior mandibular defects and the most lateral ones; (9) the low morbidity of the donor region; and (10) it can be elevated with skin and muscle as an osteomyocutaneous flap.

The fibula provides up to 25 cm of bicortical bone that can be osteotomizing in multiple locations to create a curvilinear contour in the neomandible without devascularizing any segments.[1]

Even if the procedure is standardized, shaping the neomandible with conventional technique still remains a challenge, especially when the defect is large and 2 or more osteotomies were required.

Recently, CAD/CAM technologies were introduced for osseous free flap reconstruction with the purpose of obtaining more precise reconstruction spearing time, especially in complex defects.[12]

The CAD/CAM technique allowed to manage a high number of segmental osteotomies, and this is indicative of a greater level of reconstructive accuracy.[14]

As Seruya et al[15] reported, the major advantages of CAD/CAM technologies are as follows: (1) preplanning of segmental osteotomies, (2) performance of osteotomies via cutting guides both at the tumor site and at the donor site, (3) generation of a prebent reconstruction plate to facilitate insetting of the neomandible, and (4) the possibility to use stereolithographic model.

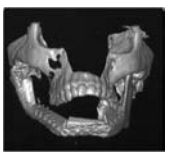

FIGURE 4. Immediate postoperative CT scan demonstrating comprehensive restoration of the mandibular defect (patient 3).

Several reports showed that fibula flap ischemia time using CAD/CAM technologies was lower than that using the conventional technique. This is valid if the osteotomies were performed free handedly in the back table. We usually perform osteotomies at the donor site with the flap still hinged to its pedicle, so in our hand, the ischemia time is not shorter with the use of CAD/CAM technology.[12,14,15]

In the cases we report on, the mean ischemia time was 75 minutes. These data are similar to what we reported in our work in 2012 that analyzed outcomes using conventional free fibula flap for mandibular reconstruction.[16]

Some authors reported a significant decrease in mean operative time for patients undergoing mandibular reconstruction with CAD and rapid prototype modeling compared with traditional reconstruction.[12] Even if our work is not a control-matched study, we did not find a decrease in mean operative time.

The virtual planning was reliable in all 3 cases. In 2 cases, no additional adjustments were needed. In 1 case, minor adjustment was needed.

In conclusion, there are 2 main advantages of the present technique: (1) the preoperative model allows the surgeon to plan the resection and reconstruction before surgery; and (2) resection and reconstruction via cutting guides and prebent plate allow the restoring of the complex structure of the resected bone to obtain a greater level of reconstructive accuracy.

Therefore, this method is indicated in selected cases in which complex reconstruction is required owing to large defects. Conventional methods might be preferable for cost-related reasons when the resected area is small and does not require more than 2 osteotomies.

The main limitation of this study is the small sample. More experience and further long-term follow-up studies are needed to evaluate a much larger patient population with better control over the variables.

REFERENCES

1. Cordeiro PG, Disa JJ, Hidalgo DA, et al. Reconstruction of the mandible with osseous free flaps: a ten-year experience with 150 consecutive patients. *Plast Reconstr Surg* 1999;104:1314
2. Cordeiro PG, Hidalgo DA. Conceptual considerations in mandibular reconstruction. *Clin Plast Surg* 1995;22:61
3. Schusterman MA, Harris SW, Raymond AK, et al. Immediate free flap mandibular reconstruction: Significance of adequate surgical margins. *Head Neck* 1993;15:204–207
4. August M, Tompach P, Chang Y, et al. Factors influencing the long-term outcome of mandibular reconstruction. *J Oral Maxillofac Surg* 2000;58:731–737
5. Phillips CM. Primary and secondary reconstruction of the mandible after ablative surgery. Report of twenty-four cases using stainless stell prostheses. *Am J Surg* 1967;114:601–604
6. Swartz WM, Banis JC, Newton ED, et al. The osteocutaneous scapular flap for mandibular and maxillary reconstruction. *Plast Reconstr Surg* 1986;77:530–545
7. Hidalgo DA. Fibula free flap: a new method of mandible reconstruction. *Plast Reconstr Surg* 1989;84:71–79
8. Rose EH, Norris MS, Rosen JM. Application of high-tech three-dimensional imaging and computer-generated models in complex facial reconstructions with vascularized bone grafts. *Plast Reconstr Surg* 1993;91:252–264
9. Ueda K, Tajima S, Tanaka Y, et al. Mandibular reconstruction using computer-generated three-dimensional solid models. *J Reconstr Microsurg* 1994;10:291–296
10. Hallermann W, Olsen S, Bardyn T, et al. A new method for computer-aided operation planning for extensive mandibular reconstruction. *Plast Reconstr Surg* 2006;117:2431–2437

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11. Gbara A, Darwich K, Li L, et al. Long-term results of jaw reconstruction with microsurgical fibula graft and dental implants. *J Oral Maxillofac Surg* 2007;65:1005–1009
12. Hanasono MM, Skoracki RJ. Computer-assisted design and rapid prototype modeling in microvascular mandible reconstruction. In: *Laryngoscope* 2013;123:597–604.
13. Gilbert A. Vascularised transfer of the fibular shaft. *Int J Microsurg* 1979;1:100
14. Modabber A, Legros C, Rana M, et al. Evaluation of computer-assisted jaw reconstruction with free vascularized fibular flap compared to conventional surgery: a clinical pilot study. *Int J Med Robot* 2012;8:215–220
15. Seruya M, Fisher M, Rodriguez ED. Computer-assisted versus conventional free fibula flap technique for craniofacial reconstruction: an outcomes comparison. *Plast Reconstr Surg* 2013;132:1219–1228
16. Ramieri GA, Zavattero E, Tosco P, et al. Fibula free flap recontruction for segmental mandibular defects. *Italian J Maxillofac Surg* 2012;23:15–25

Nasal Septal Perforation Reconstruction With Superior-Based Pedicled Nasolabial Island Flap in a Patient With the Raynaud Phenomenon

Musa Kemal Keles, MD, Hakan Cepni, MD,* Faruk Cicekci, MD,† and Alper Yenigün, MD‡*

Abstract: Perforation of the nasal septum may have multiple causes, and there are lots of options for reconstruction. We discuss a septal perforation case with Raynaud phenomenon.

Key Words: Nasolabial island flap, Raynaud phenomenon, septal perforation

Perforation of the nasal septum may have multiple causes: traumatic, iatrogenic, infectious, degenerative, overuse of vasoconstrictors, abuse of cocaine, and, more recently, chemotherapy agents.[1] Nasal septal perforation repair is a challenging issue. It leads to several symptoms. Septal perforation does not close spontaneously.[2] Surgical treatment is the most accurate treatment option. Success rates differs from 20% to 100%.[2] There are lots of etiological factors for septal perforation.[1–3]

Closure may become more difficult with accompanying rheumatic diseases. Raynaud phenomenon (RP) is a clinical condition. It can be primary or secondary to an underlying disease. Primary RP occurs without an underlying cause, and secondary RP occurs in association with an underlying disease such as systemic lupus erythematosus, polyarteritis nodosa, and scleroderma.[4]

To the best of our knowledge, we present the first case in the literature of a septal perforation repair with nasolabial island flap.

CLINICAL REPORT AND MANAGEMENT

In January 2014, a 34-year-old woman presented with crusting, dryness, nasal obstruction, and whistling in her nose. Examination results revealed a 3×3-cm posterior nasal septal perforation (Figs. 1A, B). She had a history of open rhinoplasty operation and RP. The diagnosis of RP was established by the internal medicine department with clinical findings. Under general anesthesia, 6-mL local anesthetic (lidocaine HCl, 20 mg/mL; epinephrine HCl, 0.0125 mg/mL) was applied to reduce the bleeding. Open technique was performed using the previous reversed V-shaped columellar incision and ipsilateral rim incision. Perforated septal area was exposed. Afterward, a 3×3.5-cm left-sided angular artery–based superior pedicled nasolabial flap was dissected. Ipsilateral alar incision was performed on existing scars to extend the island flap to perforated septal area through a tunnel under the nasal mucosa (Figs. 2A, B). Flap was adapted to the defective septum with 4–0 absorbable sutures. All nasal and flap donor-site incisions were closed, and silicone nasal packs were placed. The base of the island flap was left to epithelize spontaneously. Nasal packs were removed 7 days after the surgery. No early complication was encountered. At the 6-month follow-up, all symptoms of the patient decreased and closure of defect was accomplished (Fig. 3).

DISCUSSION

Lots of surgical techniques have been described to reconstruct the nasal septal perforation.[5] Miscellaneous flaps and grafts have been used to repair the septal defect.[6,7] The defects smaller than 2 cm can be reconstructed with mucoperichondrial/mucoperiosteal rotation flaps with a success rate of 90% to 100%.[7] Previous surgical scars and accompanying diseases may impair vascular supply of the nasal mucosa and adjacent tissues.[8] The nasal mucosa can also be atrophic, and it might not be suitable to cover large septal defects.[9] In this case, we prefered to use a well-vascularized tissue because scar tissues arose from previous surgery and accompanying

FIGURE 1. Nasal septal perforation: endoscopic view (A) and intraoperative view (B).

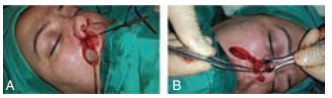

FIGURE 2. Nasolabial flap: dissection of flap (A) and adaptation through submucosal tunnel (B).

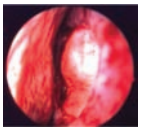

FIGURE 3. Postoperative view.

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rheumatologic disease. Nasolabial flaps can give enough tissue to close the septal defect. Besides, the septal defect was too large to reconstruct with local mucosal flaps. Owing to posterior localization of the perforation, we did not prefer oral mucosal flaps to avoid pedicle shortness.

Nasal septal perforation can be encountered in patients with various rheumatic diseases.[10] In a previous study, nasal septal perforation was reported in 12 patients with rheumatologic diseases and 11 of these 12 patients had RP.[3] Raynaud phenomenon affects 5 times more women than men, with prevalence up to 3%.[11] It may occur in association with autoimmune disorders: 90% of patients with systemic sclerosis (presenting symptoms in 30% of cases) as well as 20% of patients with systemic lupus erythematosus and frequently develops in patients with rheumatoid arthritis. It usually occurs before 30 years of age. Our patient was 34 years old when she presented with nasal septal perforation.

In past studies, 2-stage cheek flap was used; however, we repaired a large nasal defect using nasolabial island flap in 1 stage because an axial nasolabial island flap has a constant blood supply.[12,13]

Even the scar was acceptable for our patient. This technique is not routinely applicable owing to donor-site scarring. Also, rising a nasolabial island flap requires advanced surgical technical knowledge. Additional incisions that may be needed are another disadvantage of the technique. Although we made the incision on the scar generated by the previous surgeon, more incision may be needed in all patients.

Raynaud phenomenon is associated with several pathologies; however, this is the first report on a septal perforation repair with nasolabial island flap with RP.

REFERENCES

1. Mocella S, Muia F, Giacomini PG, et al. Innovative technique for large septal perforation repair and radiological evaluation. *Acta Otorhinolaryngol Ital* 2013;33:202–214
2. Rokkjær MS, Barrett TQ, Petersen CG. Good results after endonasal cartilage closure of nasal septal perforations. *Dan Med Bull* 2010;57:A4196
3. Willkens RF, Roth GJ, Novak A, et al. Perforation of nasal septum in rheumatic diseases. *Arthritis Rheum* 1976;19:119–121
4. Senel S, Zorlu P, Dogan B, et al. Successfully treated fingertip necrosis in an infant with primary Raynaud phenomenon. *Indian J Pediatr* 2014;81:1399–1400
5. Re M, Paolucci L, Romeo R, et al. Surgical treatment of nasal septal perforations. Our experience. *Acta Otorhinolaryngol Ital* 2006;26:102–109
6. Yenigun A, Meric A, Verim A, et al. Septal perforation repair: mucosal regeneration technique. *Eur Arch Otorhinolaryngol* 2012;269: 2505–2510
7. Heller JB, Gabbay JS, Trussler A, et al. Repair of large nasal septal perforations using facial artery musculomucosal (FAMM) flap. *Ann Plast Surg* 2005;55:456–459
8. Seyhan T, Kircelli BH, Caglar B. Correction of septal and midface hypoplasia in maxillonasal dysplasia (Binder's syndrome) using high-density porous polyethylene. *Aesthetic Plast Surg* 2009;33:661–665
9. Yildirim G, Onar V, Sayin I, et al. The reconstruction of nasal septal perforation with high density porous polyethylene covered with fascia lata: an experimental study on rabbit model. *Clin Exp Otorhinolaryngol* 2011;4:137–141
10. Avcin T, Silverman ED, Forte V, et al. Nasal septal perforation: a novel clinical manifestation of systemic juvenile idiopathic arthritis/adult onset Still's disease. *J Rheumatol* 2005;32:2429–2431
11. Bartelink ML, Wollersheim H, vandeLisdonk E, et al. Prevalence of Raynaud's phenomenon. *Neth J Med* 1992;41:149–152
12. Ohlsén L. Closure of nasal septal perforation with a cutaneous flap and a perichondrocutaneous graft. *Ann Plast Surg* 1988;21:276–288
13. Romo T 3rd, Sclafani AP, Falk AN, et al. A graduated approach to the repair of nasal septal perforations. *Plast Reconstr Surg* 1999;103:66–75

Relationship Between Activity of Gluthatione Peroxidase and Nitric Oxide in Synovial Fluid and the Progression of Temporomandibular Joint Internal Derangement

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Abstract: The purposes of this study were to measure the activity of glutathione peroxidase (GPX) and nitric oxide (NO) in the synovial fluid of patients with temporomandibular joint (TMJ) internal derangement (ID) and to indicate the relationship between the activity of GPX and NO and the progress of the ID.

Twenty-six patients with TMJ ID were identified and classified according to Wilkes staging through clinical and radiologic examinations. Levels of GPX were determined indirectly by a coupled reaction with glutathione reductase. Levels of NO were measured colorimetrically.

The activity of GPX and NO was observed to be progressively increasing as the stage of the TMJ ID progressed. There were significant correlations between the 2 substances and the Wilkes stages.

Oxidative stress may have a role in the pathogenesis of TMJ ID. In synovial fluid, GPX and NO activities are increased as the stage of the disease increased. Increase in the activities of GPX might not be enough to prevent progression of the TMJ ID.

Key Words: Temporomandibular joint, internal derangement, free radical, antioxidant, GPX, NO

Temporomandibular joint (TMJ) *internal derangement* (ID) is a common TMJ disorder that can be defined as an abnormal relationship among mandibular condyle, articular eminence, and articular disk. Limited mandibular motion, joint noise, jaw pain, irregular jaw movement, and joint tenderness are the typical symptoms of ID.[1–4] Recent studies have shown that ID is a progressive disorder, which can be classified.[5] Wilkes[6] defined the subgroups as early, early intermediate, intermediate, late

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intermediate, and late stages on the basis of clinical, anatomic, and radiologic examinations.

Despite intensive clinical and experimental studies that showed not only the morphologic process but also the biochemical agent cause ID, the mechanisms involved in such progressive symptom-generating processes in ID are not clear yet.[5] In recent years, the biochemical analysis of the contents of synovial fluid (SF), obtained either by direct aspiration or by lavage, which is used as a treatment modality particularly of intermadiate or late stages of the TMD ID, has provided new insights into the pathophysiologic nature of TMJ diseases.[7–9] The SF of the TMJ has been analyzed for the presence of various mediators and free radicals that may be used as markers of joint disease.[10–12]

It is known that some cytokines, cytokine receptors (interleukin [IL]-1, IL-1β, IL-6, IL-8, IL-11, and tumor necrosis factor α), as well as antioxidant and antioxidant enzymes (superoxide dismutase [SOD], methionine reductase, catalase, and glutathione peroxidase (GPX); vitamin A, β-carotene, as well as vitamins C and E; the trace mineral selenium; and the hormone melatonin) exist in synovial tissue or fluids of patients with TMJ ID.[3,5,10,12–14]

Nitric oxide (NO), the short-lived free radical, is synthesized from L-arginine. It is considered as an important endogenous mediator for neurotransmission, vasodilatation, nerve function, and immune defense mechanisms. Physiologic functions involving a change in flow or shear stress, a change in oxygen tension, and a variety of vasoactive substances cause the release of NO from endothelial cells.[2,12,14–16]

Low levels of NO production are important in protecting an organ such as the liver from ischemic damage. On the other hand, sustained levels of increased NO production result in direct tissue damage.[12,15,16]

Glutathione peroxidase is the general name of an enzyme family with peroxidase activity whose main biological role is to protect the organism from oxidative damage. It catalyzes the reduction of hydroperoxides using glutathione, thereby protecting mammalian cells against oxidative damage. An imbalance in the production or activation of antioxidant enzymes can lead to generation of oxidative stress. It is well documented that oxidative stress is one of the reasons for apoptotic cell death, and GPX can play a pivotal role in preventing apoptosis.[17–19]

The presence of elevated levels of certain mediators in certain conditions in TMJ ID increases the possibility that a lowering of mediators may destroy joint tissues or an enhancement of protective mediators levels may have therapeutic value.

This study was undertaken to evaluate the relation of the progression of the TMJ ID according to the Wilkes classification[6] and levels of GPX and NO in the SF of the patients.

MATERIALS AND METHODS

This study included 26 patients (22 females and 4 males) who presented to the Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, University of Ankara and who were diagnosed with internal derengement. The age of our patient population ranged from 18 to 47 years (30.44 ± 7.91). Exclusion criteria applied to patients with systemic arthropathy and/or history of trauma.

To control consistent surgical procedure and treatment protocol as well as conservative treatment of the patients, 1 clinician (O.G.) was involved. According to clinical examination and magnetic resonance findings, all patients included in the study were diagnosed with internal derangement. A visual analog scale (VAS)-marked scores 0 through 100 preoperatively was used to assess pain with end points. Arthrocentesis was performed on 27 TMJs of these 26 patients. The maximum interincisal mouth opening was measured preoperatively with a ruler to the nearest millimeter.

After subcutaneous local anesthesia was administered with 3% Citanest Octapressin solution (Dentsply, Surrey, United Kingdom) containing prilocaine hydrochloride, 30 mg/mL, TMJ SF samples were obtained through arthrocentesis, which was performed according to the technique descrbed by Nitzan et al.[20] To allow the saline solution to mix with SF, saline solution was reaspirated into the syringe for the biochemical analysis. The SF samples were frozen at -80 C^{o} until assayed. The measurement of biochemical parameter NO and GPX was analyzed using commercial kits (Cayman Chemical Company). Levels of GPX, as an index of oxidative stress, were determined indirectly through a coupled reaction with glutathione reductase. Levels of NO were measured colorimetrically. Although arthrocentesis is a simple procedure that can be performed under local anesthesia, the study was carried out without control samples for ethical reasons.

The results of clinical and radiologic examinations were used to classify the patients according to Wilkes[6] staging:

Stage I (early stage) - No significant mechanical symptoms, no pain or limitation of motion.

Stage II (early-intermediate stage) - First, few episodes of pain, occasional joint tenderness and related temporal headaches, increase in intensity of clicking, joint sounds; later, in opening movement, beginning transient subluxations or joint locking.

Stage III (intermediate stage) - Multiple episodes of pain, joint tenderness, temporal headaches, locking, closed locks, restriction of motion, painful chewing, anterior disk displacement, moderate to marked disk thickening, normal osseous contour.

Stage IV (intermediate-late stage) – Chronic and episodic pain, headaches, variable restriction of motion, anterior disk displacement, marked disk thickening, and abnormal bone contours.

Stage V (late stage) - Crepitus on examination, variable and episodic pain, anterior disk displacement with disk perforation and gross deformity, and degenerative osseous changes, chronic restriction of motion, difficulty to function.

All data were recorded using a standard data form and analyzed using SPSS version 20.0 (SPSS, Chicago, IL).

RESULTS

Twenty-six patients and 27 TMJs who had undergone intervention were involved, and the amount of GPX and NO in SF was classified according to Wilkes[6] classification in this study.

Table 1 summarizes the clinical and pathologic preoperative data of the patients studied. Twenty-seven TMJs that had undergone intervention were grouped according to the Wilkes stages. The distributions of the patients according to the Wilkes classification were as follows: 10 (7 women and 3 men; mean age, 30.90 ± 8.56 y) TMJs were stage III, 10 (9 women and 1 man; mean age, 28.80 ± 8.64 y) TMJs were stage IV, and 7 (7 women; mean age, 32.14 ± 6.41 years) TMJs were stage V. There were no statistically significant differences in the ages among the stages. Maximum interincisal opening (MIO) for stage III was 17.90 ± 5.20; that for stage IV, 19 ± 3.74, and that for stage V, 23.14 ± 4.78. The MIO seemed to be decreased as the stage of disease increased ($P < 001$). The VAS scores were 74.80 ± 5.01 in stage III, 81.5 ± 4.74 in stage IV, and 81.43 ± 5.29 in stage V. The VAS scores were significantly higher in the stage IV and V patients than in the stage III patients. Table 2 shows the results of GPX and NO in SFs of the patients with ID classified according to Wilkes[6] staging. We compared GPX and NO levels in SF and between Wilkes stage III, stage IV, and stage V. There were significant correlations between the 2 substances and Wilkes stages. The values and activities of GPX and NO seemed to be progressively increased as the stage of the disease increased ($P < 0.001$). The results obtained by the analysis of the data using Kruskal-Wallis test are given in Table 2. Spearman correlation shows that there is no correlation between GPX and NO (Table 3).

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TABLE 1. The Clinical Data of the Patients

Stages (Wilkes)	Age	Sex	Diseased Side	VAS	MIO
Stage III	28	F	R	74	23
Stage III	29	F	R	74	17
Stage III	30	F	R	71	10
Stage III	18	F	L	80	20
Stage III	35	F	R	75	10
Stage III	20	F	R	69	25
Stage III	32	M	R	86	22
Stage III	47	F	R	70	14
Stage III	40	M	R	74	20
Stage III	30	M	L	75	18
Stage IV	18	F	R	80	15
Stage IV	19	F	R	78	24
Stage IV	38	F	R	83	17
Stage IV	31	F	L	91	14
Stage IV	39	F	R	88	18
Stage IV	32	F	L	81	21
Stage IV	40	F	R	81	22
Stage IV	29	F	R	80	20
Stage IV	23	M	L	76	15
Stage IV	19	F	L	77	24
Stage V	20	F	L	86	21
Stage V	32	F	R	82	19
Stage V	30	F	R	84	23
Stage V	35	F	R	75	31
Stage V	37	F	L	84	24
Stage V	40	F	R	73	27
Stage V	31	F	R	86	17

Analog scale; MIO (in millimeters).
F indicates female; L, left; M, male; R, right.

DISCUSSION

The TMJ is a load-bearing synovial joint subject to breakdown by damaging molecular events triggered by overloading or systemic disease.[14] In the current study, patients with systemic arthropathy and/or history of trauma were not included.

There is clinical evidence supporting the existence of disk displacement in TMJ ID.[14] However, recent concepts suggest that a change in the position of the disk is not the primary factor in dysfunction and pain of the TMJ.[14] In addition, more recently, studies using magnetic resonance imaging suggest that the articular disk is displaced in 35% of asymptomatic volunteers.[21,22]

Overloading of TMJ results in a pathologic state if the intrinsic healing or adaptive capacity of the joint is exceeded. Thus, disk displacement results from a failure of the lubricant system that is initiated by free radicals.[14] Previous studies have shown that NO is an important mediator of inflammation and tissue injury.[13,14,23] In our study, we found out that NO level was the lowest in the early stage of the ID. The highest level of the NO was present in the late stage. The level of NO significantly increased progressively as the stage of the disease increased ($P < 0.001$). It is suggested that these occur with 2 mechanisms: First, mechanical loads can result in the production of highly reactive molecules, which are free radicals including NO.[2–4] Free radicals damage important molecules of the articular tissues as well as SF and trigger celluler responses. Second, mechanical loads can stimulate sensory neurons supplying the nerve-rich regions of the TMJ, resulting in release of neuropeptides and NO, which reacts with superoxides to form the potent oxidant peroxynitrite, which is implicated in cellular injury. Thus, it contributes to inflammation. An additional explanation (on the other way) is that, in the TMJ, focal regions may become hypoxic if intra-articular pressures exceed the end capillary perfusion pressures of the blood vessels supplying affected articular tissues. Under this condition, oxygen-derived free radicals are generated and these damage joint tissues.[13,14]

TABLE 2. The Results of GPX and NO in Synovial Fluids of the Patients With Internal Derangement Classified According to Wilkes Staging

Stage	GPX (nmol/min/ml)	NO (nmol/min/ml)
III (n = 10)	10.91 ± 3.13	8.82 ± 1.06
IV (n = 10)	17.52 ± 3.26	12.02 ± 3.09
V (n = 7)	29.26 ± 5.37	18.79 ± 6.82

Values are expressed as mean ± standard deviation.
There were significant correlations between the 2 substances (GPX and NO) and Wilkes stages.
$*P < 0.001$.

TABLE 3. Noncorrelation Between GPX and NO

	r	P	n
Wilkes stage 3	0.159	0.661	10
Wilkes stage 4	0.384	0.273	10
Wilkes stage 5	−0.107	0.819	7

Our results suggest that enhancement of the NO level causes the progression of the TMD ID. That means tissue damaging is enhanced, abnormal bone contouring occurs, and disk degenerations including perforation and deformity occur. Parallel to these findings, the pain will be more felt by the patients and the TMJ system will work painfully with restriction of motion.

Like our findings, Alpaslan et al[13] reported that painful TMD, including disk derangements, is associated with a highly significant increase in the level of NO in the TMJ SF. In a patient with TMJ ID, pain might be associated with NO, which causes vasodilatation and vascular permeability.[13] Kaneyama et al[24] reported that the concentrations of the proinflammatory cytokines were higher in patients with TMJ ID than that in controls. This reflects the activated immunologic network of the proinflammatory cytokines in the pathogenesis of TMD. Takahashi et al[12,25] reported that the levels of NO in SF from the patients with ID or osteoarthritis (OA) were significantly higher than those from the control group. However, there were no differences in the levels of NO between the ID and OA groups.

There are some in vivo data about the levels of antioxidant enzymes in the SF of patients with OA.[26–28] There are still controversial results, especially concerning whether SF is being either increasing or decreasing antioxidant mechanism. Sutipornpalangkul et al[26] did not find a significant decrease in antioxidant molecules (glutathione), as reported by Regan et al,[27] and an increased antioxidant enzyme activity (GPX and superoxide dismutase [SOD] activity), as reported by Ostalowska et al.[28] During the antioxidant mechanism, superoxide radicals (O2?) are converted to hydrogen peroxide (H_2O_2) by SOD enzyme and H_2O_2 is further detoxified and metabolized by catalase or GPX.[28] Ostalowska et al[28] found a highly significant increase in the level of GPX in the TMJ SF. Sutipornpalangkul et al[26] did not observe significant changes in SF GPX activities. Ediz et al[29] reported no significant differences in terms of the mean whole blood and serum antioxidative activity of GPX. Tercic and Bozic[30] found decreased GPX activities in SF from patients, as compared with normal SF. Carlo and Loeser[31] reported that these antioxidant enzymes are rarely present in extracellular fluids, such as SF, which contain little or no GPX activity. In the current study, the values and activities of GPX progressively

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increased as the stage of the disease increased ($P < 0.001$). It is observed that increase of the activities of GPX is not enough (itself) to prevent progression of the TMJ ID. Also, there is no correlation between GPX and NO levels. There might be some other molecules. Further research is needed to get a suitable solution for such cases.

In conclusion, this study is the first report demonstrating that there is a correlation between SF GPX and NO activities and Wilkes stages. It is shown that, as the stage of the disease increased, GPX and NO activities are increased. Nitric oxide may cause harmful effects on TMJ tissues and may worsen the disorder. We found that high levels of NO in SF can induce high activity of GPX to protect articular cartilage from the harmful effects of the oxidative strees and free radicals. Increase in the activities of GPX might not be enough (itself) to prevent the destroying effect of the NO and progression of the TMJ ID. Further studies and long-term evaluations are needed to better understand the pathogenesis of the ID and to effectively treat the patients with TMJ ID.

REFERENCES

1. Tang YL, Zhu GQ, Hu L, et al. Effects of intra-articular administration of sodium hyaluronate on plasminogen activator system in temporomandibular joints with osteoarthritis. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2010;109:541–547
2. Gulen H, Ataoglu H, Haliloglu S, et al. Proinflammatory cytokines in temporomandibular joint synovial fluid before and after arthrocentesis. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2009;107:1–4
3. Matsumoto K, Honda K, Ohshima M, et al. Cytokine profile in synovial fluid from patients with internal derangement of the temporomandibular joint: a preliminary study. *Dentomaxillofac Radiol* 2006;35:432–441
4. Bouloux GF. Temporomandibular joint pain and synovial fluid analysis: a review of the literature. *J Oral Maxillofac Surg* 2009;67:2497–2504
5. Hamada Y, Kondoh T, Holmlund AB, et al. Cytokine and clinical predictors for treatment outcome of visually guided temporomandibular joint irrigation in patients with chronic closed lock. *J Oral Maxillofac Surg* 2008;66:29–34
6. Wilkes CH. Internal derangements of the temporomandibular joint. Pathological variations. *Arch Otolaryngol Head Neck Surg* 1989;115:469–477
7. Fredriksson L, Alstergren P, Kopp S. Tumor necrosis factor-(in temporomandibular joint synovial fluid predicts treatment effects on Pain by intra-articular glucocorticoid treatment. *Mediators Inflamm* 2006;6:1–7
8. Arinci A, Ademoglu E, Aslan A, et al. Molecular correlates of temporomandibular joint disease. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2005;99:666–670
9. Tozoglu S, Al-Belasy FA, Dolwick MF. A review of techniques of lysis and lavage of the TMJ. *Br J Oral Maxillofac Surg* 2011;49:302–309
10. Güven O, Tekin US, Durak I, et al. Superoxide dismutase activity in synovial fluids in patients with temporomandibular joint internal derangement. *J Oral Maxillofac Surg* 2007;65:1940–1943
11. Fujimura K, Segami N, Yoshitake Y, et al. Electrophoretic separation of the synovial fluid proteins in patients with temporomandibular joint disorders. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2006;101:463–468
12. Takahashi T, Kondoh T, Kamei K, et al. Elevated levels of nitric oxide in synovial fluid from patients with temporomandibular disorders. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1996;82:505–509
13. Alpaslan C, Bilgihan A, Alpaslan GH, et al. Effect of arthrocentesis and sodium hyaluronate injection on nitrite, nitrate, and thiobarbituric acid-reactive substance levels in the synovial fluid. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2000;89:686–690
14. Laskin DM, Greene CS, Hylander WL. Temporamandibular disorders: an evidence-based approach to diagnosis and treatment Chicago: Quintessence; 2006:441–481
15. Blake DR, Unsworth J, Outhwaite JM, et al. Hypoxic-reperfusion injury in the inflamed joint. *Lancet* 1989;333:289–293
16. Taylor BS, Kim YM, Wang Q, et al. Nitric oxide down-regulates hepatocyte-inducible nitric oxide synthase gene expression. *Arch Surg* 1997;132:1177–1183
17. Kalpakcioglu B, Senel K. The interrelation of glutathione reductase, catalase, glutathione peroxidase, superoxide dismutase, and glucose-6-phosphate in the pathogenesis of rheumatoid arthritis. *Clin Rheumatol* 2008;27:141–145
18. Chorazy PA, Schumacher HR Jr, Edlind TD. Role of glutathione peroxidase in rheumatoid arthritis: analysis of enzyme activity and DNA polymorphism. *DNA Cell Biol* 1992;11:221–225
19. Karatay S, Kiziltunc A, Yildirim K, et al. Effects of different hyaluronic acid products on synovial fluid NO levels in knee osteoarthritis. *Clin Rheumatol* 2005;24:497–501
20. Nitzan DW, Dolwick MF, Martinez GA. Temporomandibular joint arthrocentesis: a simplified treatment for severe, limited mouth opening. *J Oral Maxillofac Surg* 1991;49:1163–1167
21. Katzberg RW, Westesson PL, Tallents RH, et al. Anatomic disorders of the temporomandibular joint disc in asymptomatic subjects. *J Oral Maxillofac Surg* 1996;54:147–153
22. Ribeiro RF, Tallents RH, Katzberg RW, et al. The prevalence of disc displacement in symptomatic and asymptomatic volunteers aged 6 to 25 years. *J Orofac Pain* 1997;11:37–47
23. Emshoff R, Puffer P, Rudisch A, et al. Temporomandibular joint pain: relationship to internal derangement type, osteoarthrosis, and synovial fluid mediator level of tumor necrosis factor-α. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2000;90:442–449
24. Kaneyama K, Segami N, Nishimura M, et al. Osteoclastogenesis inhibitory factor/osteoprotegerin in synovial fluid from patients with temporomandibular disorders. *Int J Oral Maxillofac Surg* 2003;32:404–407
25. Takahashi T, Kondoh T, Fukuda M, et al. Proinflammatory cytokines detectable in synovial fluids from patients with temporomandibular disorders. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1998;85:135–141
26. Sutipornpalangkul W, Morales NP, Charoencholvanich K, et al. Lipid peroxidation, glutathione, vitamin E, and antioxidant enzymes in synovial fluid from patients with osteoarthritis. *Int J Rheum Dis* 2009;12:324–328
27. Regan EA, Bowler RP, Crapo JD. Joint fluid antioxidants are decreased in osteoarthritic joints compared to joints with macroscopically intact cartilage and subacute injury. *Osteoarthritis Cartilage* 2008;16:515–521
28. Ostalowska A, Birkner E, Wiecha M, et al. Lipid peroxidation and antioxidant enzymes in synovial fluid of patients with primary and secondary osteoarthritis of the knee joint. *Osteoarthritis Cartilage* 2006;14:139–145
29. Ediz L, Hiz O, Ozkol H, et al. Relationship between anti-CCP antibodies and oxidant and anti-oxidant activity in patients with rheumatoid arthritis. *Int J Med Sci* 2011;9:139–147
30. Tercic D, Bozic B. The basis of the synovial fluid analysis. *Clin Chem Lab Med* 2001;39:1221–1226
31. Carlo MD Jr, Loeser RF. Increased oxidative stress with aging reduces chondrocyte survival: correlation with intracellular glutathione levels. *Arthritis Rheum* 2003;48:3419–3430

Blood Parameters as Indicators of Upper Airway Obstruction in Children With Adenoid or Adenotonsillar Hypertrophy

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Adenotonsillar hypertrophy (ATH) is the most common cause of obstructive sleep apnea in children. This study aimed to evaluate the blood parameters of children with ATH who underwent surgery.

Methods: The study included a review of the medical records of 130 children who underwent adenoidectomy or adenotonsillectomy with a diagnosis of adenoid hypertrophy and/or chronic tonsillitis. Patients were classified into 3 groups: group 1 (n = 69) underwent adenoidectomy, group 2 (n = 61) underwent adenotonsillectomy, and group 3 consisted of 82 healthy children. White blood cell count, platelet count, hemoglobin levels, mean platelet volume, and platelet distribution width values were the primary outcome measures.

Results: Mean platelet volume, platelet distribution width and hemoglobin values decreased in the groups that underwent surgery. Whereas the decrease in group 1 was insignificant, it was significant in group 2. White blood cell count values increased in both group 1 (adenoidectomy) and group 2 (adenotonsillectomy), but the increase in group 2 was significant. No significant difference in platelet count was detected before versus after the operation.

Conclusions: Upper airway obstruction caused by ATH remarkably changes the blood parameters related to chronic hypoxia. Significant improvement can be achieved after adenotonsillectomy rather than adenoidectomy alone.

Key Words: Mean platelet volume, platelets, upper airway obstruction, adenotonsillar hypertrophy, adenoidectomy, tonsillectomy

Adenotonsillar enlargement is the most common cause of upper airway obstruction (UAO) in the pediatric population. It has been suggested that cardiopulmonary complications such as pulmonary hypertension, cor pulmonale, and even heart failure can occur in this population, secondary to obstructive lesions.[1,2] Long-standing hypoxia secondary to adenotonsillar hypertrophia was found to be related to decreased somatic growth and impaired neurocognitive functions of children.[3,4] Upper airway obstruction stemming from adenotonsillar hypertrophia has been associated with increased inflammatory responses.[5]

Platelet indices and white blood cell (WBC) count are the main blood markers of systemic inflammation in all ages.[6,7] As a known platelet parameter, mean platelet volume (MPV) has been reported as an indicator of platelet activation and has an important role in the pathophysiology of cardiovascular diseases.[8] Larger platelets are metabolically and enzymatically more active and have greater prothrombotic potential with higher levels of thromboxane A_2 and increased levels of procoagulant surface proteins.[9] It had previously been reported that MPV increases in patients with obstructive sleep apnea (OSA).[10] Furthermore, it has been shown that continuous positive airway therapy (CPAP) decreases platelet activation.[11,12]

Adenotonsillectomy is a common procedure in children with adenotonsillar pathology. However, the effects of adenotonsillectomy on cardiovascular risks associated with UAO in the pediatric age group are still inconclusive.

To date, there are no data in the relevant literature that focuses on the effects of adenoidectomy and adenotonsillectomy on the blood parameters of children. As a result, this retrospective clinical study aimed to investigate the change in blood markers of inflammation in cases of adenotonsillar hypertrophia who underwent adenoidectomy or adenotonsillectomy.

METHODS

This retrospective clinical study was conducted at 3 centers: (1) ENT Department, Kirikkale University Faculty of Medicine, Kirikkale, Turkey; (2) Ministry of Health, Yesilyurt State Hospital, ENT Clinics, Malatya, Turkey; and (3) Clinics of Social Pediatrics, Dr. Sami Ulus Maternity and Children's Training and Research Hospital, Ministry of Health, Ankara, Turkey. It was conducted over a period of 2 years, from January 1, 2012, to December 31, 2013. The study was completed in accordance with the basic principles of the Declaration of Helsinki.

Subjects

The study included 130 children who underwent adenoidectomy (group 1) or adenotonsillectomy (group 2) with a diagnosis of adenoid hypertrophy (AH) and/or chronic tonsillitis (CT) within the study period. An additional 82 children, who were admitted to outpatient pediatrics clinics without any complaints of upper respiratory tract obstructions, systemic diseases, or acute/chronic infections or diseases, served as the control group (group 3).

To be included, the study group had to meet the following criteria: (1) AH causing 50% or more obstruction in nasopharynx in transnasal flexible nasopharyngoscopic examination, (2) history of peritonsillar abscess or repeated attacks of acute tonsillitis totaling more than 4 or 5 per year, (3) no history of previous upper airway surgery, (4) no accompanying comorbid medical or surgical conditions related to UAO, and (5) completion of 3 months of follow-up.

Methods

Demographic properties and blood parameters of all participants were investigated electronically, and required data were obtained from the patients' medical files. The blood parameters, including MPV (fL), platelet distribution width (PDW) (fL), WBC ($\times 10^3/\mu L$), platelet (PLT) ($\times 10^3/\mu L$), and hemoglobin (Hb) (g/dL) values of patients who underwent adenoidectomy or adenotonsillectomy during admission and postoperative third month control, were compared with the values of the control group participants.

Venous blood samples were taken into tubes containing K3 EDTA. The Improvacuter blood collection tube (Guangzhou Improve Medical Instruments Co, Ltd, Guangzhou, China) auto analyzer was used for the total blood count analyzed in the hospitals' central laboratories. Mean platelet volume, PDW, Hb, PLT, and WBC values were individually assessed.

Sample Size

A sample size of 60 patients per group provides approximately 80% power at the 5% 2-sided significance level to detect at least 5% decrease in the MPV values of patients with OSA treated with surgery, assuming that the surgical treatment of these cases significantly improved the hypoxia and therefore decreased the platelet volume.

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TABLE 1. Mean Ages and Sex Distribution of Groups*

		Group 1 (Adenoidectomy) (n = 69)	Group 2 (Adenotonsillectomy) (n = 61)	Group 3 (Control) (n = 82)	P
Age, mean (SD), y		8.1 ± 3.6	8.1 ± 2.2	8.2 ± 2.2	0.992
Sex, n (%)	Male	39 (56.5)	32 (52.5)	39 (47.6)	0.544
	Female	30 (43.5)	29 (47.5)	43 (52.4)	

* One way ANOVA and χ^2 tests were used.

Statistical Analysis

All study data were evaluated using the Statistical Packages for Social Sciences 16.0 software (SPSS Inc, Chicago, IL). For the statistical analysis, the values are expressed as mean ± SD whenever appropriate. The normalities of distributions were tested using the Kolmogorov-Smirnov test. Normally distributed variables were compared by 1-way analysis-of-variance (ANOVA) test, whereas Kruskal-Wallis tests were used to compare the non–normally distributed variables between the groups. For multiple comparisons of groups, Tukey and Conover tests were used where appropriate. Categorical variables were compared with a χ^2 test. A paired-sample *t* test and Wilcoxon signed rank test were used to compare the preoperative and postoperative blood parameters within each group. $P < 0.05$ was considered statistically significant.

RESULTS

Group 1 included 69 children, group 2 included 61 children, and group 3 included 82 children. The groups were similar in terms of their demographic characteristics (Table 1).

Table 2 presents the comparison of preoperative and postoperative blood parameters of the study group with control subjects. The changes in MPV and PDW values after the operation were compared in groups 1 and 2. The decrease in MPV was insignificant after adenoidectomy ($P = 0.179$); however, in cases of adenotonsillectomy, MPV values significantly decreased after the operation ($P < 0.001$). Platelet distribution width also changed slightly after adenoidectomy ($P = 0.852$), whereas in cases of adenotonsillectomy, the change in PDW was significant ($P < 0.001$). The decrease in Hb was insignificant after adenoidectomy ($P = 0.644$); however, in cases of adenotonsillectomy, Hb values significantly decreased after operation ($P < 0.001$). No significant difference was observed in operated cases with regard to platelet counts ($P = 0.536$ for adenoidectomy; $P = 0943$ for adenotonsillectomy). The change in WBC was insignificant after adenoidectomy ($P = 0.584$); however, in cases of adenotonsillectomy, WBC values significantly changed after operation ($P < 0.001$). Figures 1 and 2 illustrate blood parameters and platelet values, respectively.

DISCUSSION

The results obtained from the current study revealed that children with AH and/or CT have significantly increased MPV and PDW values compared with control subjects. In addition, the presence of both AH and CT is associated with markedly increased Hb values. After surgical treatment, the abnormal hemogram findings related to UAO returned within the reference range of healthy subjects ($P < 0.05$). These results not only confirm the current knowledge concerning the relationship between UAO and chronic hypoxia; they also add to the current literature that children having both AH and CT are exposed to much more hypoxia, and resection of hypertrophic adenoid and tonsil tissues together enhance significant healing of hypoxia, which showed markedly decreased MPV, PDW, and Hb values after the operation.

Various studies show platelet activation in patients with chronic obstructive airway disease.[13–18] Most of them state a clear association between the obstructive airway pathologies and increased platelet activity. Previously, Varol et al[10] showed that MPV, an indicator of platelet activation, was significantly higher in adult patients with severe OSA when compared with control subjects. Similarly, Kucur et al[14] reported that MPV levels were significantly higher in children with AH than in control subjects and were significantly lower in these patients after adenoidectomy. Steiropoulos et al[16] also reported that patients with chronic obstructive pulmonary disease had relatively high MPV values. It has been suggested that chronic hypoxia causes catecholamine-dependent platelet activation and that this leads to changes in platelet shape with an increase in platelet swelling, leading to an increase in MPV and PDW.[19,20] In contrast, Cengiz et al[13] suggested that MPV values in patients with AH-CT were significantly lower than those in control subjects.

In children, AH and CT, which cause nocturnal hypoxia, are the most frequent causes of OSA.[21,22] Children with AT and/or CT suffer from snoring and sleep with open mouths and experience OSA episodes; in addition, if left untreated, they can experience abnormal

TABLE 2. The Comparison of Preoperative and Postoperative Blood Parameters of Groups*

		Group 1 (Adenoidectomy) (n = 69)	Group 2 (Adenotonsillectomy) (n = 61)	Group 3 (Control) (n = 82)	
		Mean ± SD	Mean ± SD	Mean ± SD	P
MPV, fL	Preop	8.20 ± 1.07	8.28 ± 0.77	7.76 ± 0.77†	*0.001*
	Postop	8.06 ± 1.02‡	7.55 ± .074‡	7.76 ± 0.77	*0.004*
		Median (Min-Max)	Median (Min-Max)	Median (Min-Max)	
PDW, fL	Preop	16.1 (15.3–18.5)	16.2 (15.5–17.9)	13.8 (9.3–19.5)†	*<0.001*
	Postop	16.2 (14.8–18.5)‡	15.5 (14.0–17.6)‡	13.8 (9.3–19.5)‡	*<0.001*
Hb, g/dL	Preop	12.8 ± 1.3	13.6 ± 0.7†	12.4 ± 0.9	*0.001*
	Postop	12.7 ± 1.1	12.7 ± 0.7	12.4 ± 0.9	0.113
PLT, $\times 10^3/\mu L$	Preop	293 ± 70	283 ± 64†	318 ± 79	*0.011*
	Postop	299 ± 102	284 ± 50‡	318 ± 79‡	*0.042*
WBC, $\times 10^3/\mu L$	Preop	8.1 ± 3.0†	7.0 ± 1.2	7.1 ± 1.7	*0.003*
	Postop	8.4 ± 2.9	8.9 ± 2.1	7.1 ± 1.7†	*<0.001*

* One way ANOVA test and Kruskal-Wallis *H* tests were used.
† Significantly different from others.
‡ Significantly different from each other.
P values in italics are statistically significant.

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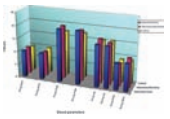

FIGURE 1. Blood parameters in groups 1 to 3. ∗MPV values are given as mean, and the other values are given as median.

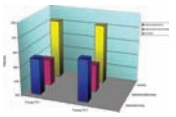

FIGURE 2. Platelet values in groups 1 to 3. ∗PLT values are given as median.

orofacial anatomy, poor appetite, growth retardation, aggressive behavior, anxiety, impaired attention, depression, and somatization disorders over the long term.[1–4] Although adenoidectomy and adenotonsillectomy are common procedures performed in ear-nose-throat practices, studies investigating the benefits of surgical treatment in pediatric UAO by objective methods are quite limited. Varol et al[11] showed that 6 months of CPAP therapy caused significant reductions in the MPV values in patients with severe OSA who had significantly higher MPV values before treatment. Similarly, Oga et al[23] have reported that ADP-induced platelet aggregability increased significantly in patients with moderate to severe OSA compared with patients with no to mild OSA, and CPAP treatment improved platelet aggregability at 90 days.

This study also revealed that children with UAO secondary to AT-CT had significantly increased platelet activation and that this abnormal activity may return to normal with adenoidectomy and/or adenotonsillectomy. In addition, patients with both AH and CT had significantly higher preoperative Hb values, and the adenotonsillectomy procedure significantly decreased the MPV values compared with children who underwent only adenoidectomy. This finding may be due to the fact that relief of airway obstruction might be more pronounced after adenotonsillectomy, instead of adenoidectomy alone.

This is presumably the first time such observations have been reported in pediatric cases with UAO. The results of this study might help justify adenotonsillectomy in children with AH and CT instead of adenoidectomy alone. According to the study findings, parents of children with AT and CT should be informed during the preoperative interview that the removal of both hypertrophic adenoid and tonsils offers better results than adenoidectomy alone.

The present study has clear limitations, including its retrospective design and the relatively small number of patients surveyed in each group. A prospective controlled clinical setting with participation of multiple institutions would enhance clear results that indicate the association of blood parameters and AH and/or CT and the effects of surgical treatment on these parameters.

Finally, it is possible to report that UAO resulting from AH and/or CT in children causes platelet activation and increased Hb values secondary to chronic hypoxia and that the elimination of UAO by surgical treatment might break abnormal platelet activation cascade, which is more prominent in cases that underwent adenotonsillectomy. Because increased platelet activity could contribute to long-term cardiovascular complications, timely surgical treatment in pediatric OSA patients may confer some cardioprotective effects through the reduction of platelet activation.

REFERENCES

1. Tatlipinar A, Duman D, Uslu C, et al. The effects of obstructive sleep apnea syndrome due to adenotonsillar hypertrophy on the cardiovascular system in children. *Turk J Pediatr* 2011;53:359–363
2. Li HY, Lee LA. Sleep-disordered breathing in children. *Chang Gung Med J* 2009;32:247–257
3. Vontetsianos HS, Davris SE, Christopoulos GD, et al. Improved somatic growth following adenoidectomy and tonsillectomy in young children. Possible pathogenetic mechanisms. *Hormones (Athens)* 2005;4:49–54
4. Ezzat WF, Fawaz S, Abdelrazek Y. To what degree does adenotonsillectomy affect neurocognitive performance in children with obstructive sleep apnea hypopnea syndrome due to adenotonsillar enlargement? *ORL J Otorhinolaryngol Relat Spec* 2010;72:215–219
5. Shen Y, Xu Z, Shen K. Urinary leukotriene E4, obesity, and adenotonsillar hypertrophy in Chinese children with sleep disordered breathing. *Sleep* 2011;34:1135–1141
6. Akelma AZ, Mete E, Cizmeci MN, et al. The role of mean platelet volume as an inflammatory marker in children with chronic spontaneous urticaria. *Allergol Immunopathol (Madr)* 2015;43:10–13
7. Zahorec R. Ratio of neutrophil to lymphocyte counts-rapid and simple parameter of systemic inflammation and stress in critically ill. *Bratisl Lek Listy* 2001;102:5–14
8. Sen N, Basar N, Maden O, Ozcan F, et al. Increased mean platelet volume in patients with slow coronary flow. *Platelets* 2009;20:23–28
9. Bath PM, Butterworth RJ. Platelet size: measurement, physiology and vascular disease. *Blood Coagul Fibrinolysis* 1996;7:157–161
10. Varol E, Ozturk O, Gonca T, et al. Mean platelet volume is increased in patients with severe obstructive sleep apnea. *Scand J Clin Lab Invest* 2010;70:497–502
11. Varol E, Ozturk O, Yucel H, et al. The effects of continuous positive airway pressure therapy on mean platelet volume in patients with obstructive sleep apnea. *Platelets* 2011;22:552–556
12. Ray RM, Bower CM. Pediatric obstructive sleep apnea: the year in review. *Curr Opin Otolaryngol Head Neck Surg* 2005;13:360–365
13. Cengiz C, Erhan Y, Murat T, et al. Values of mean platelet volume in patients with chronic tonsillitis and adenoid hypertrophy. *Pak J Med Sci* 2013;29:569–572
14. Kucur C, Kulekci S, Zorlu A, et al. Mean platelet volume levels in children with adenoid hypertrophy. *J Craniofac Surg* 2014;25:e29–e31
15. Wang RT, Li JY, Cao ZG, et al. Mean platelet volume is decreased during an acute exacerbation of chronic obstructive pulmonary disease. *Respirology* 2013;18:1244–1248
16. Steiropoulos P, Papanas N, Nena E, et al. Mean platelet volume and platelet distribution width in patients with chronic obstructive pulmonary disease: the role of comorbidities. *Angiology* 2013;64:535–539
17. Varol E. Role of mean platelet volume in acute exacerbation of chronic obstructive pulmonary disease. *Pol Arch Med Wewn* 2012;122:380–381
18. Biljak VR, Pancirov D, Cepelak I, et al. Platelet count, mean platelet volume and smoking status in stable chronic obstructive pulmonary disease. *Platelets* 2011;22:466–470
19. Boos CJ, Lip GY. Assessment of mean platelet volume in coronary artery disease—what does it mean? *Thromb Res* 2007;120:11–13
20. Ziegler MG, Nelesen R, Mills P, et al. Sleep apnea, norepinephrine release rate, and daytime hypertension. *Sleep* 1997;20:224–231
21. Izu SC, Itamoto CH, Pradella-Hallinan M, et al. Obstructive sleep apnea syndrome (OSAS) in mouth breathing children. *Braz J Otorhinolaryngol* 2010;76:552–556
22. Muzumdar HV, Sin S, Nikova M, et al. Changes in heart rate variability after adenotonsillectomy in children with obstructive sleep apnea. *Chest* 2011;139:1050–1059
23. Oga T, Chin K, Tabuchi A, et al. Effects of obstructive sleep apnea with intermittent hypoxia on platelet aggregability. *J Atheroscler Thromb* 2009;16:862–869

Relief of Pain at Rest and During Swallowing After Modified Cautery-Assisted Uvulopalatopharyngoplasty: Bupivacaine Versus Lidocaine

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Objectives: We investigated the efficacy of bupivacaine, lidocaine, and saline infiltrations to peritonsillar region and uvula and soft palate regions for pain relief after tonsillectomy and modified cautery-assisted uvulopalatopharyngoplasty (MCAUP) in patients with obstructive sleep apnea.

Methods: In this prospective study, 91 patients (32–65 years old) with obstructive sleep apnea underwent tonsillectomy and MCAUP and were divided into 3 groups. In group 1 patients (n = 31), 0.25% bupivacaine HCl + 1/200,000 epinephrine (10 mL); in group 2 patients (n = 31), 1% lidocaine HCl + 1/200,000 epinephrine (10 mL); and in group 3 patients (n = 29), 0.9% saline (10 mL) were injected to peritonsillar region and uvula and soft palate regions. Operation duration, amount of bleeding, and analgesic requirement and visual analog scale for pain at rest and at swallowing were evaluated in all groups.

Results: Mean body mass index values were between 27.0 and 27.3 kg/m^2 in all groups. Their apnea-hypopnea index values were between 15.3 and 16.9 per hour, and there were no significant differences between their body mass index and apnea-hypopnea index values. Duration of operation of patients in the bupivacaine group was significantly lower than that of patients in the lidocaine and saline groups. In addition, in the lidocaine group, operation duration was significantly lower than that in the saline group. Bleeding amount and postoperative analgesic requirement of the saline group were significantly higher than those of the bupivacaine and lidocaine groups. Although both these measures (operation duration and amount of bleeding) were statistically significant, a mean operating time of 44.3 versus 46.0 minutes and 64.4-mL versus 68.4-mL blood loss for the bupivacaine and lidocaine groups were clinically irrelevant and not too important. In terms of visual analog scale for pain at rest and/or swallowing, Bupivacaine provided more relief than lidocaine and saline injections. The lidocaine provides pain relief less than bupivacaine and more than saline at rest and/or swallowing.

Conclusion: We recommend the use of bupivacaine injections in peritonsillar, uvular, and soft palate regions during tonsillectomy + MCAUP operations. It reduces operation duration and provides more pain relief postoperatively. When patients had cardiac problems, lidocaine may also be recommended because of its cardiac depressant and antiarrhythmic effects and positive effects for pain relief compared with saline injections.

Key Words: Bupivacaine, lidocaine, tonsillectomy, modified cautery-assisted uvulopalatopharyngoplasty (MCAUP), pain at rest, pain at swallowing

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Uvulopalatopharyngoplasty (UPPP) is a common method for the treatment of moderate to severe obstructive sleep apnea (OSA) syndrome,[1] but wound-related pain is severe following UPPP, making adequate postoperative analgesia necessary.[2] Traditional postoperative analgesia is most frequently performed with opioids, and although the analgesic effects of such agents are well known, their potential adverse reactions (such as excessive sedation and respiratory depression) limit their application in postoperative UPPP analgesia.[3]

In tonsillectomy operations, peritonsillar infiltrations were studied by different researchers.[4–7] Moss et al[6] studied the effects of pretonsillectomy injection of local anesthetics with and without clonidine for reducing pain following tonsillectomy in children. The lidocaine plus bupivacaine plus clonidine group was significantly less likely to need intravenous pain medication in the recovery room compared with children in the placebo group, and again it was shown that the placebo group achieved a significantly more advanced diet and had less pain on postoperative days 1 and 3.

Ozmen et al[7] compared the effects of levobupivacaine hydrochloride, bupivacaine hydrochloride, and saline injections in alleviating posttonsillectomy pain. The pain scores in the levobupivacaine group were lower than those in the saline group at 13 hours ($P < 0.017$). The pain scores in the bupivacaine and levobupivacaine groups were significantly lower than those in the saline group from 17 to 21 hours until day 6 ($P > 0.017$). There was no difference between the levobupivacaine and bupivacaine groups ($P > 0.017$). Local infiltration of levobupivacaine is a relatively safe and effective method and is equivalent to use of bupivacaine for posttonsillectomy pain.

As UPPP operations have been performed for OSA patients, severe postoperative pain in these patients was noticed.[8] Patients usually suffer severe pain, especially during swallowing, which consequently results in morbidities and delayed return to normal activities after UPPP.[9] Opioid-based analgesic protocols can easily lead to respiratory depression and life-threatening conditions in patients with OSA syndrome.[10,11] Therefore, it is necessary to find a safe postoperative analgesic method for these patients.

In the present study, we performed tonsillectomy and modified cautery-assisted UPPP (MCAUP) in 91 OSA patients. We investigated the effects of bupivacaine, lidocaine, and saline infiltrations to the peritonsillar region and the uvula and soft palate regions. The outcomes were demonstrated in terms of visual analog scale (VAS) for pain at rest and VAS values for pain at swallowing.

MATERIALS AND METHODS

This prospective study was conducted in Adana Numune Training and Research Hospital between February 2012 and January 2014 according to the principles of Helsinki Declaration.[12] Ethics committee approval of Adana Numune Training and Research Hospital was also obtained. Patients were included into the study after signing informed consent.

Subjects

Ninety-seven patients were admitted to the ENT Department with the complaint of snoring and underwent tonsillectomy and MCAUP (uvulopalatal flap technique). Six patients were excluded from the study because the sutures were switched off. Therefore, 91 patients were included into the study. They were between 32 and 65 years old. Their body mass indexes (BMIs) were calculated, and polysomnography was performed preoperatively.

Patients were randomized into 3 groups: upper, lateral, and lower (tongue root) peritonsillar region, root of uvula, and soft palate:

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group 1 (bupivacaine group) (n = 31): 0.25% bupivacaine HCl + 1/200,000 epinephrine (10 mL) was injected
group 2 (lidocaine group) (n = 31): 1% lidocaine HCl + 1/200,000 epinephrine (10 mL) was injected
group 3 (control group) (n = 29): 0.9% saline (10 mL) was injected.

The surgeons knew which solution was injected to the patients. Inclusion criteria were as follows:

- obstructive sleep apnea patients with apnea-hypopnea index (AHI) >5 and <30
- obstructive sleep apnea patients whose BMI was less than 30 kg/m^2
- subjects with ≥2+ tonsillar hypertrophy[13]
- patients with retropalatal obstruction

Exclusion criteria were as follows:

- patients who underwent palatal surgery previously
- patients with chronic diseases preventing surgery.

Operation

All patients underwent tonsillectomy and MCAUP under general anesthesia. After mouth opener was inserted, 5 mL of the solution (different in groups 1, 2, and 3) was injected as 2.5 mL to the right tonsil and 2.5 mL to the left tonsil on upper pole, lateral of tonsil plica, and to the root of the tongue for deep pole. The remaining 5-mL solution was applied to the root of the uvula and soft palate region. Ten minutes after the injections, tonsillectomy has been applied. Bleeding control was achieved with bipolar cautery. Considering the projection that occurred at folding of uvula to the soft palate, the mucosal and submucosal tissues related to the uvula and the region of the soft palate were excised with the aid of electrocautery. Also, the amount was excised from the end portion of the uvula. After control of bleeding, horizontal incisions were made to the upper part of the posterior plicas of the tonsils. Uvula was folded, and in new position, it was fixed using 3-0 Vicryl sutures. Starting from the horizontal incision, tonsil posterior plicas were sutured to the root of the tongue (Fig. 1).

Methods

Patients' operation duration and amount of bleeding were recorded. After the surgery, at the 2nd, 4th, 8th, 16th, 24th, and 48th hours and third to seventh days, pain at rest and during swallowing was measured by VAS.[1–10] Therefore, VAS for pain at rest and VAS for pain at swallowing were recorded. At postoperative period, acetaminophen (250 mg/5 mL) was used in 10 to 15 mg/kg per dose (4 times a day).[14] In case of pain, the patients were permitted to take additional acetaminophen, which did not exceed the maximum daily dose of 4 g, and these doses were recorded as postoperative pain medication need. In none of the patients was primary or secondary hemorrhage or velopharyngeal insufficiency observed.

Statistical Analysis

All study data were evaluated using the Statistical Packages for Social Sciences version 16.0 software (SPSS Inc, Chicago, IL). A χ^2 test and Kruskal-Wallis H test with Bonferroni correction were used for analysis. $P < 0.05$ was considered statistically significant.

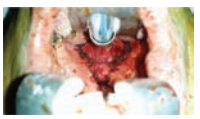

FIGURE 1. Tonsillectomy and MCAUP. Postoperative view, after applying of the sutures.

TABLE 1. Gender Distribution of Groups 1 to 3

		Groups								
		Bupivacaine (Group 1) (n = 31)		Lidocaine (Group 2) (n = 31)		Saline (Group 3) (n = 29)		Total		
		n	%	n	%	n	%	n	%	P^*
Sex	Male	19	61.3	17	54.8	17	58.6	53	58.2	$P = 0.874$,
	Female	12	38.7	14	45.2	12	41.4	38	41.8	$\chi^2 = 0.267$
	Total	31	100.0	31	100.0	29	100.0	91	100.0	

* P value shows the results of the χ^2 test.

RESULTS

Sex distributions of groups 1 to 3 are shown in Table 1. In the bupivacaine group, there were 19 males (61.3%) and 12 females (38.7%). In the lidocaine group, there were 17 males (54.8%) and 14 females (45.2%). In the saline group, there were 17 males (58.6%) and 12 females (41.4%). There were no significant differences between sexes in the groups by χ^2 test ($P > 0.05$).

Mean ages of groups 1 to 3 were 45.2 ± 7.9, 46.5 ± 7.5, and 46.9 ± 8.4 years, respectively. There were no significant differences between groups by Kruskal-Wallis H test ($P > 0.05$) (Table 2).

Body mass index values of the groups were 27.0 ± 1.8, 27.1 ± 1.7, and 27.3 ± 1.6 kg/m^2, respectively. There were no significant differences between groups by Kruskal-Wallis H test ($P > 0.05$) (Table 2).

Apnea-hypopnea index values of the groups were 16.4 ± 6.8, 16.9 ± 7.0, and 15.3 ± 5.7 per hour. There were no significant differences between groups by Kruskal-Wallis H test ($P > 0.05$) (Table 2).

There were significant differences between operation duration, amount of bleeding, and analgesic requirement of the groups ($P < 0.05$) (Table 2). Operation duration of bupivacaine group was significantly lower than those of the lidocaine and saline groups. In addition, in the lidocaine group, operation duration was significantly lower than that in the saline group ($P = 0.0001$) (Table 2). Bleeding amount and analgesic requirement of the saline group were significantly higher than those of the bupivacaine and lidocaine groups ($P = 0.0001$) (Table 2).

Although both these measures (operation duration and amount of bleeding) were statistically significant, a mean operating time of 44.3 versus 46.0 minutes and 64.4- versus 68.4-mL blood loss for the bupivacaine and lidocaine groups were clinically irrelevant and not too important.

For all measurement times (2nd, 4th, 8th, 16th, 24th, and 48th hours and third to seventh days), there was statistically significant difference between VAS-pain at rest scores of the groups ($P < 0.05$) (Table 3, Fig. 2). Visual analog scale for pain at rest scores of bupivacaine group was significantly lower than that of the lidocaine and saline groups at 2nd, 4th, 8th, 16th, and 24th hours and third and sixth days. In addition, in the lidocaine group, VAS scores for pain at rest was significantly lower than the saline group at the 2nd, 4th, 8th, 16th, and 24th hours and third and sixth days ($P = 0.0001$) (Table 3).

At 48 hours, VAS values for pain at rest of the bupivacaine and lidocaine groups were significantly lower than that of the saline

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TABLE 2. Age, BMI, AHI, Operation Duration, Amount of Bleeding, and Analgesic Requirement of Groups 1 to 3

								Kruskal-Wallis Variance Analysis		
	Groups	n	Mean	Median	Min	Max	SD	*H*	*P**	Pairwise Comparisons
Age, y	Bupivacaine	31	45.2	44.0	32.0	62.0	7.9	0.565	0.754	—
	Lidocaine	31	46.5	47.0	34.0	64.0	7.5			
	Saline	29	46.9	45.0	36.0	65.0	8.4			
	Total	91	46.2	45.0	32.0	65.0	7.9			
BMI	Bupivacaine	31	27.0	27.3	23.0	29.7	1.8	0.262	0.877	—
	Lidocaine	31	27.1	27.0	23.7	29.9	1.7			
	Saline	29	27.3	27.4	24.4	29.9	1.4			
	Total	91	27.1	27.3	23.0	29.9	1.6			
AHI	Bupivacaine	31	16.4	14.7	7.2	29.6	6.8	0.593	0.743	—
	Lidocaine	31	16.9	16.0	7.1	28.8	7.0			
	Saline	29	15.3	14.0	7.6	27.6	5.7			
	Total	91	16.2	15.3	7.1	29.6	6.5			
Operation duration, min	Bupivacaine	31	44.3	44.0	35.0	60.0	5.5	30.713	0.0001	1–2
	Lidocaine	31	46.0	45.0	36.0	60.0	5.0			1–3
	Saline	29	51.3	50.0	45.0	60.0	3.4			2–3
	Total	91	47.1	48.0	35.0	60.0	5.5			
Amount of bleeding, mL	Bupivacaine	31	64.4	55.0	40.0	160.0	24.5	38.37	0.0001	1–3
	Lidocaine	31	68.4	60.0	50.0	210.0	28.7			
	Saline	29	94.1	85.0	60.0	220.0	27.1			2–3
	Total	91	75.2	70.0	40.0	220.0	29.6			
Analgesic requirement	Bupivacaine	31	1.7	2.0	0.0	4.0	1.0	79.06	0.0001	1–3
	Lidocaine	31	4.1	4.0	3.0	6.0	0.8			
	Saline	29	9.0	8.0	6.0	12.0	1.6			2–3
	Total	91	4.8	4.0	0.0	12.0	3.2			

* *P* value shows the results of Kruskal-Wallis *H* test with Bonferroni correction.

group ($P = 0.0001$) (Table 3). Moreover, at the fourth, fifth, and seventh days, VAS values for pain of each of the lidocaine and saline groups were significantly lower than that of the bupivacaine group ($P = 0.0001$) (Table 3).

For all measurement times (2nd, 4th, 8th, 16th, 24th, and 48th hours and third to seventh days), there was statistically significant difference between VAS-pain at swallowing scores of the groups ($P < 0.05$) (Table 4, Fig. 3).

At all measurement times, VAS values for pain at swallowing of the bupivacaine group were significantly lower than that of the saline group. At the 2nd, 4th, 8th, 16th, 24th, and 48th hours and third day, VAS values for pain at swallowing of the lidocaine group were significantly lower than that of the saline group. At the 4th, 8th, 16th, 24th, 48th hours and third and sixth days, VAS values for pain at swallowing of the bupivacaine group were significantly lower than those of the lidocaine group ($P = 0.0001$) (Table 4).

DISCUSSION

A modified UPPP consisted of a classic tonsillectomy or residual tonsil resection and additional shortening of the uvula. The natural mucosal fold between the uvula and the upper pole of the tonsils was carefully preserved. A wide opening to the rhinopharynx was created by asymmetric suturing of the glossopalatine and pharyngopalatine arches. Postoperative pain is the one of the complaints after UPPP. The other problems were dryness of the mouth, slight difficulty in swallowing, discrete speech disturbances, and slight pharyngeal dysesthesias with feeling of a lump in the throat and compulsive clearing of the throat.[15]

Xie et al[16] reported that local anesthesia combined with intravenous parecoxib can (1) compensate for the short analgesic effect of local anesthetic alone, (2) avoid excessive sedation and respiratory depression caused by systemic opioids, and (3) provide well-tolerated postoperative analgesia in patients with OSA syndrome undergoing UPPP.

Uvulopalatal flap technique, commonly combined with necessary tonsillectomy, has been used for snoring and OSA treatment in recent years. During the surgical excision of mucosa and submucosa in the uvulopalatal region, scalpel or electrocautery can be used. When electrocautery is used, operation can be conducted as bloodless. However, postoperative pain can restrict the use of it in these operations. Cincik et al[17] reported that postoperative pain was mostly seen in laser-assisted uvulopalatoplasty and UPPP cases. The cautery-assisted uvulopalatoplasty procedure was easy and the least painful. In our study, we investigated the effects of local anesthetics (bupivacaine or lidocaine) infiltrations in terms of increasing the comfort of the patients postoperatively compared with saline.

In the current study, tonsillectomy and MCAUP under general anesthesia were performed. Infiltrations to peritonsillar region and the uvula and soft palate regions were performed at the beginning of the operations. Ten minutes after the injections, tonsillectomy was performed. Bleeding control was achieved with bipolar cautery. Mean BMI values were between 27.0 and 27.3 kg/m^2 in all groups. Their AHI values were between 15.3 and 16.9 per hour, and there were no significant differences between their BMI and AHI values.

Operation duration of the bupivacaine group was significantly lower than those of the lidocaine and saline groups. In addition, in the lidocaine group, operation duration was significantly lower than that in the saline group. Bleeding amount and postoperative analgesic requirement of the saline group were significantly higher than those of the bupivacaine and lidocaine groups. In terms of VAS for pain at rest and/or swallowing, bupivacaine provided more relief

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TABLE 3. Postoperative Pain at Rest of the Groups

								Kruskal-Wallis Variance Analysis		
Pain	**Groups**	**n**	**Mean**	**Median**	**Min**	**Max**	**SD**	***H***	***P****	**Pairwise Comparisons**
2nd hour	Bupivacaine	31	2.6	3.0	0.0	6.0	1.2	67	0.0001	1–2
	Lidocaine	31	3.8	4.0	3.0	5.0	0.7			1–3
	Saline	29	6.4	6.0	5.0	8.0	0.8			2–3
	Total	91	4.2	4.0	0.0	8.0	1.8			
4th hour	Bupivacaine	31	2.8	3.0	0.0	6.0	1.2	64.1	0.0001	1–2
	Lidocaine	31	4.5	4.0	3.0	7.0	1.1			1–3
	Saline	29	6.8	7.0	5.0	10.0	1.0			2–3
	Total	91	4.6	5.0	0.0	10.0	2.0			
8th hour	Bupivacaine	31	3.5	4.0	0.0	6.0	1.4	55.7	0.0001	1–2
	Lidocaine	31	5.0	5.0	3.0	7.0	1.0			
	Saline	29	6.9	7.0	5.0	10.0	1.1			1–3
	Total	91	5.1	5.0	0.0	10.0	1.8			2–3
16th hour	Bupivacaine	31	3.6	4.0	0.0	6.0	1.5	51	0.0001	1–2
	Lidocaine	31	4.7	5.0	2.0	7.0	1.2			1–3
	Saline	29	6.8	7.0	5.0	10.0	1.1			2–3
	Total	91	5.0	5.0	0.0	10.0	1.8			
24th hour	Bupivacaine	31	2.7	3.0	0.0	7.0	1.6	41	0.0001	1–2
	Lidocaine	31	3.9	4.0	1.0	8.0	1.5			
	Saline	29	5.8	6.0	4.0	10.0	1.4			1–3
	Total	91	4.1	4.0	0.0	10.0	2.0			2–3
48th hour	Bupivacaine	31	2.4	2.0	0.0	5.0	1.5	32.8	0.0001	1–3
	Lidocaine	31	3.1	3.0	1.0	6.0	1.4			
	Saline	29	5.1	5.0	2.0	10.0	1.6			2–3
	Total	91	3.5	3.0	0.0	10.0	1.9			
3rd day	Bupivacaine	31	1.7	2.0	0.0	5.0	1.3	35.2	0.0001	1–2
	Lidocaine	31	2.6	3.0	1.0	6.0	1.1			1–3
	Saline	29	4.0	4.0	2.0	7.0	1.2			2–3
	Total	91	2.8	3.0	0.0	7.0	1.5			
4th day	Bupivacaine	31	2.1	2.0	0.0	5.0	1.5	10.7	0.005	1–2
	Lidocaine	31	2.7	3.0	1.0	5.0	1.1			
	Saline	29	3.3	3.0	1.0	7.0	1.3			1–3
	Total	91	2.7	3.0	0.0	7.0	1.4			
5th day	Bupivacaine	31	1.9	2.0	0.0	4.0	1.1	18.1	0.0001	1–2
	Lidocaine	31	2.8	3.0	1.0	5.0	1.3			
	Saline	29	3.4	4.0	1.0	7.0	1.4			1–3
	Total	91	2.7	3.0	0.0	7.0	1.4			
6th day	Bupivacaine	31	1.4	1.0	0.0	3.0	1.1	28.6	0.0001	1–2
	Lidocaine	31	2.2	2.0	0.0	5.0	1.2			
	Saline	29	3.4	3.0	1.0	7.0	1.4			1–3
	Total	91	2.3	2.0	0.0	7.0	1.5			2–3
7th day	Bupivacaine	31	1.0	1.0	0.0	3.0	0.9	16.9	0.0001	1–2
	Lidocaine	31	1.9	2.0	0.0	5.0	1.1			
	Saline	29	2.3	2.0	1.0	7.0	1.3			1–3
	Total	91	1.7	2.0	0.0	7.0	1.2			

* *P* value shows the results of Kruskal-Wallis *H* test with Bonferroni correction.

than lidocaine and saline injections. Lidocaine provides pain relief at rest and/or swallowing less than does bupivacaine, but more than does saline.

Bupivacaine is a local anesthetic drug belonging to the amino amide group. Bupivacaine binds to the intracellular portion of sodium channels and blocks sodium influx into nerve cells, which prevents depolarization. Because pain-transmitting nerve fibers tend to be thinner and either unmyelinated or lightly myelinated, the agent can diffuse more readily into them than into thicker and more heavily myelinated nerve fibers such as those involved in touch and proprioception. (Myelin is nonpolar/lipophilic.) Bupivacaine also blocks specific potassium channels, an effect contributing to resting membrane potential depolarization.[18]

Lidocaine is a local anesthetic and cardiac depressant used as an antiarrhythmic agent. Its actions are more intense, and its effects

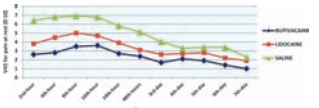

FIGURE 2. VAS values for pain at rest of the Bupivacaine, Lidocaine and Saline Groups.

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TABLE 4. Postoperative Pain at Swallowing of Groups 1 to 3

								Kruskal-Wallis Variance Analysis		
Pain With Swallowing	**Groups**	**n**	**Mean**	**Median**	**Min**	**Max**	**SD**	***H***	***P****	**Pairwise Comparisons**
2nd hour	Bupivacaine	31	4.2	4.0	1.0	8.0	1.5	47	0.0001	1–3
	Lidocaine	31	4.4	4.0	3.0	7.0	1.1			2–3
	Saline	29	6.8	7.0	5.0	8.0	0.8			
	Total	91	5.1	5.0	1.0	8.0	1.7			
4th hour	Bupivacaine	31	4.3	4.0	1.0	7.0	1.5	47.7	0.0001	1–2
	Lidocaine	31	5.5	5.0	3.0	8.0	1.3			1–3
	Saline	29	7.4	8.0	6.0	10.0	1.0			2–3
	Total	91	5.7	6.0	1.0	10.0	1.8			
8th hour	Bupivacaine	31	4.7	5.0	1.0	7.0	1.5	48.1	0.0001	1–2
	Lidocaine	31	6.1	6.0	4.0	8.0	1.0			1–3
	Saline	29	7.6	8.0	6.0	10.0	0.9			2–3
	Total	91	6.1	6.0	1.0	10.0	1.6			
16th hour	Bupivacaine	31	4.7	5.0	1.0	7.0	1.6	45.3	0.0001	1–2
	Lidocaine	31	5.9	6.0	3.0	8.0	1.0			1–3
	Saline	29	7.4	8.0	6.0	10.0	1.0			2–3
	Total	91	6.0	6.0	1.0	10.0	1.6			
24th hour	Bupivacaine	31	4.0	4.0	1.0	8.0	1.7	38.8	0.0001	1–2
	Lidocaine	31	5.6	6.0	2.0	8.0	1.3			1–3
	Saline	29	6.8	6.0	5.0	10.0	1.2			2–3
	Total	91	5.4	6.0	1.0	10.0	1.8			
48th hour	Bupivacaine	31	3.5	3.0	1.0	6.0	1.7	34.9	0.0001	1–2
	Lidocaine	31	5.1	5.0	2.0	8.0	1.4			1–3
	Saline	29	6.2	6.0	3.0	10.0	1.3			2–3
	Total	91	4.9	5.0	1.0	10.0	1.8			
3rd day	Bupivacaine	31	2.8	3.0	1.0	6.0	1.4	31.3	0.0001	1–2
	Lidocaine	31	3.8	4.0	1.0	6.0	1.3			1–3
	Saline	29	5.2	5.0	3.0	8.0	1.3			2–3
	Total	91	3.9	4.0	1.0	8.0	1.7			
4th day	Bupivacaine	31	2.9	3.0	1.0	7.0	1.6	17.5	0.000	1–3
	Lidocaine	31	3.7	4.0	1.0	6.0	1.5			
	Saline	29	4.7	5.0	2.0	7.0	1.5			
	Total	91	3.8	4.0	1.0	7.0	1.7			
5th day	Bupivacaine	31	3.0	3.0	0.0	7.0	1.5	15	0.001	1–3
	Lidocaine	31	3.7	4.0	1.0	6.0	1.4			
	Saline	29	4.6	5.0	1.0	8.0	1.6			
	Total	91	3.7	4.0	0.0	8.0	1.6			
6th day	Bupivacaine	31	2.1	2.0	0.0	4.0	1.2	22.9	0.0001	1–2
	Lidocaine	31	3.1	3.0	0.0	6.0	1.5			
	Saline	29	4.2	4.0	1.0	8.0	1.6			
	Total	91	3.1	3.0	0.0	8.0	1.7			
7th day	Bupivacaine	31	1.7	1.0	0.0	4.0	1.3	11.9	0.003	1–3
	Lidocaine	31	2.4	2.0	0.0	6.0	1.4			
	Saline	29	3.0	3.0	1.0	7.0	1.5			
	Total	91	2.4	2.0	0.0	7.0	1.5			

* *P* value shows the results of Kruskal-Wallis *H* test with Bonferroni correction.

more prolonged than those of procaine, but its duration of action is shorter than that of bupivacaine or prilocaine. Lidocaine stabilizes the neuronal membrane by inhibiting the ionic fluxes required for the initiation and conduction of impulses, thereby effecting local anesthetic action. Lidocaine alters signal conduction in neurons by blocking the fast voltage-gated sodium (Na^+) channels in the neuronal cell membrane that are responsible for signal propagation. With sufficient blockage, the membrane of the postsynaptic neuron will not depolarize and will thus fail to transmit an action potential.[19]

In our study, considering VAS scores for postoperative pain at rest and swallowing, it is determined that peritonsillar, uvular, and soft palate injections with bupivacaine provide maximum relaxation and pain relief in the patients. However, lidocaine also provided pain relief at rest and during swallowing compared with saline postoperatively. The difference between bupivacaine and lidocaine

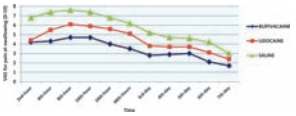

FIGURE 3. VAS values for pain at swallowing of the Bupivacaine, Lidocaine and Saline Groups.

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may be related to shorter duration of action of lidocaine compared with bupivacaine.[18] The important topic may be said that there was lower narcotic requirement for the longer-acting anesthetic.

On the other hand, if the foreground cardiac problems are present in patients with OSA, use of lidocaine, which is an agent for cardiac depressant and antiarrhythmic, should be more appropriate.

Both UPPP and tonsillectomy are associated with intense postoperative pain.[20] In tonsillectomy operations, peritonsillar infiltrations were performed to improve hemostasis during operation and to reduce of postoperative pain.[4] Broadman et al[4] reported that they were unable to evaluate the beneficial effects of peritonsillar infiltration performed with bupivacaine upon the reduction of the severity of pain and the requirement for narcotic analgesics following tonsillectomy. They suggested that peritonsillar infiltrations should be performed solely for the purpose of reducing operative blood loss. In addition, they recommended that such infiltrations should be performed with either normal saline containing epinephrine (1:200,000) or lidocaine containing epinephrine (1:200,000), whereas Wong et al[5] reported that peritonsillar infiltration of bupivacaine 0.5% with 1:200,000 epinephrine provides better posttonsillectomy pain control in the immediate postoperative period than does bupivacaine spray or placebo. In their study, group 1 received 0.5 mL/kg normal saline spray; group 2 received 2 mg/kg bupivacaine 0.5% with 1:200,000 epinephrine peritonsillar infiltration in a similar volume to that in group 1; and group 3 received 2 mg/kg bupivacaine 0.5% with 1:200,000 epinephrine spray to both tonsillar beds. The patients in each group were compared postoperatively with regard to the quality of pain control using an objective pain score and their analgesic requirements. Bameshki et al[21] reported that injection of epinephrine and bupivacaine before or after tonsillectomy is effective in reducing pain and bleeding. By this treatment, swallowing pain also decreases in the hours immediately after surgery. Ergil et al[22] suggested that levobupivacaine (0.25%) has a vasoconstrictive and a consistent analgesic effect, which may be beneficial in tonsillectomy patients.

In UPPPs, there are few studies about infiltration anesthesia and postoperative pain condition. The majority of studies have focused on reducing posttonsillectomy pain and bleeding control. Therefore, there are no studies in the literature similar to our study, which was focused on effectiveness of bupivacaine or lidocaine on postoperative pain conditions in patients who underwent tonsillectomy and MCAUP.

Patients with OSAS are at a significantly increased risk for hypertension. Therefore, in UPPP patients, nasal continuous positive airway pressure eliminated the postoperative risk of hypoxemia, which allowed the use of adequate parenteral or oral analgesics.[23] Ponstein et al[24] reported that continuous lesser palatine nerve block via infusion of ropivacaine 0.2% at 2 mL/h may be a useful regional anesthetic technique in the multimodal postoperative pain management of opioid-sensitive OSA patients undergoing UPPP.

Our results showed that bupivacaine had beneficial effects in tonsillectomy + MCAUP operations in OSA patients. It reduces the operation duration compared with the lidocaine and saline groups. Both bupivacaine and lidocaine injections reduced bleeding during the operation and postoperative analgesic requirement compared with saline. Although both these measures (operation duration and amount of bleeding) were statistically significant, a mean operating time of 44.3 versus 46.0 minutes and 64.4- versus 68.4-mL blood loss for the bupivacaine and lidocaine groups were clinically irrelevant and not too important.

For postoperative pain relief at rest and swallowing, bupivacaine was more effective than lidocaine and saline. In addition, lidocaine reduced pain at rest and swallowing compared the saline injections. Lower narcotic requirement was detected for the longer-acting anesthetic.

In conclusion, we recommend the use of bupivacaine injections in peritonsillar, uvular, and soft palate regions during tonsillectomy + MCAUP operations. It reduces operation duration and provides more pain relief postoperatively. If patients have cardiac problems, lidocaine may also be recommended because of its cardiac depressant and antiarrhythmic effects and positive effects for pain relief compared with saline injections.

REFERENCES

1. Lundkvist K, Januszkiewicz A, Friberg D. Uvulopalatopharyngoplasty in 158 OSAS patients failing non-surgical treatment. *Acta Otolaryngol* 2009;129:1280–1286
2. Gross JB, Bachenberg KL, Benumof JL, et al. Practice guidelines for the perioperative management of patients with obstructive sleep apnea: a report by the American Society of Anesthesiologists Task Force on Perioperative Management of patients with obstructive sleep apnea. *Anesthesiology* 2006;104:1081–1093
3. Kim JA, Lee JJ, Jung HH. Predictive factors of immediate postoperative complications after uvulopalatopharyngoplasty. *Laryngoscope* 2005;115:1837–1840
4. Broadman LM, Patel RI, Feldman BA, et al. The effects of peritonsillar infiltration on the reduction of intraoperative blood loss and post-tonsillectomy pain in children. *Laryngoscope* 1989;99 (6 pt 1):578–581
5. Wong AK, Bissonnette B, Braude BM, et al. Post-tonsillectomy infiltration with bupivacaine reduces immediate postoperative pain in children. *Can J Anaesth* 1995;42:770–774
6. Moss JR, Cofer S, Hersey S, et al. Comparison of clonidine, local anesthetics, and placebo for pain reduction in pediatric tonsillectomy. *Arch Otolaryngol Head Neck Surg* 2011;137:591–597
7. Ozmen S, Ozmen OA, Kasapoglu F. Effects of levobupivacaine versus bupivacaine infiltration on postoperative analgesia in pediatric tonsillectomy patients: a randomized, double-blind, placebo-controlled study. *Ann Otol Rhinol Laryngol* 2011;120:489–493
8. Douglas NJ, ed. *Clinicians' Guide to Sleep Medicine. Clinicians' Guide to Sleep Medicine*. London, UK: Arnoid; 2002
9. Hendolin H, Kansanen M, Koski E, et al. Propofolnitrous oxide versus thiopenthone-nitrous oxide anesthesia for uvulopalatopharyngoplasty in patients with sleep apnoea. *Acta Anaesthesiol Scand* 1994;38:694–698
10. Clyburn PA, Rosen M, Vickers MD. Comparison of the respiratory effects of i.v. infusions of morphine and regional analgesia by extradural block. *Br J Anaesth* 1990;64:446–449
11. Nikanne E, Virtaniemi J, Aho M, et al. Ketoprofen for postoperative pain after uvulopalatopharyngoplasty and tonsillectomy: two-week follow-up study. *Otolaryngol Head Neck Surg* 2003;129:577–581
12. 52nd, WMA., General, Assembly. World Medical Association Declaration of Helsinki: ethical principles for medical research involving human subjects. *JAMA* 2000;284:3043–3049
13. Brodsky L. Modern assessment of tonsils and adenoids. *Pediatr Clin North Am* 1989;36:1551–1569
14. Goldman RD, Scolnik D. Underdosing of acetaminophen by parents and emergency department utilization. *Pediatr Emerg Care* 2004;20:89–93
15. Röösli C, Schneider S, Häusler R. Long-term results and complications following uvulopalatopharyngoplasty in 116 consecutive patients. *Eur Arch Otorhinolaryngol* 2006;263:754–758
16. Xie GL, Chu QJ, Liu CL. Application of parecoxib in post-uvulopalatopharyngoplasty analgesia. *J Int Med Res* 2013;41:1699–1704
17. Cincik H, Cekin E, Cetin B, et al. Comparison of uvulopalatopharyngoplasty, laser-assisted uvulopalatoplasty and cautery-assisted uvulopalatoplasty in the treatment of primary snoring [published online ahead of print February 6, 2006]. *ORL J Otorhinolaryngol Relat Spec* 2006;68:149–155
18. Bupivacaine. Wikipedia. Published February 28, 2014. Available at: http://en.wikipedia.org/wiki/Bupivacaine. Accessed on July 13, 2014.
19. Lidocaine. DrugBank. Open Data Drug & Drug Target Database. Published February 28, 2014. Available at: http://www.drugbank.ca/drugs/DB00281. Accessed on July 13, 2014.
20. Virtaniemi J, Kokki H, Nikanne E, et al. Ketoprofen and fentanyl for pain after uvulopalatopharyngoplasty and tonsillectomy. *Laryngoscope* 1999;109:1950–1954

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21. Bameshki AR, Razban M, Khadivi E, et al. The effect of local injection of epinephrine and bupivacaine on post-tonsillectomy pain and bleeding. *Iran J Otorhinolaryngol* 2013;25:209–214
22. Ergil J, Akkaya T, Gozaydin O, et al. Vasoconstrictive and analgesic efficacy of locally infiltrated levobupivacaine in tonsillectomy patients. *Int J Pediatr Otorhinolaryngol* 2012;76:1429–1433
23. Riley RW, Powell NB, Guilleminault C, et al. Obstructive sleep apnea surgery: risk management and complications. *Otolaryngol Head Neck Surg* 1997;117:648–652
24. Ponstein NA, Kim TW, Hsia J, et al. Continuous lesser palatine nerve block for postoperative analgesia after uvulopalatopharyngoplasty. *Clin J Pain* 2013;29:e35–e38

Geometry of Anterior Open Bite Correction

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Abstract: Correction of anterior open bite is a frequently encountered and challenging problem for the craniomaxillofacial surgeon and orthodontist. Accurate clinical evaluation, including cephalometric assessment, is paramount for establishing the diagnosis and appropriate treatment plan. The purposes of this technical note were to discuss the basic geometric principles involved in the surgical correction of skeletal anterior open bites and to offer a simple mathematical model for predicting the amount of posterior maxillary impaction with concomitant mandibular rotation required to establish an adequate overbite. Using standard geometric principles, a mathematical model was created to demonstrate the relationship between the magnitude of the open bite and the magnitude of the rotational movements required for correction. This model was then validated using a clinical case. In summary, the amount of open bite closure for a given amount of posterior maxillary impaction depends on anatomic variables, which can be obtained from a lateral cephalogram. The clinical implication of this relationship is as follows: patients with small mandibles and steep mandibular occlusal planes will require greater amounts of posterior impaction.

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The diagnosis and surgical correction of an anterior open bite are challenging clinical problems for orthodontists and craniomaxillofacial surgeons.[1–4] Although patients often present with an obvious clinical deformity, the underlying skeletal anomalies and dental compensations necessitate careful surgical planning for definitive correction. Improper dental decompensation (ie, extrusion of the anterior teeth) and errant treatment planning can result in incorrect diagnosis, unnecessary or incorrect operations, unstable reconstruction, and, ultimately, relapse.[3,4]

The most common causes of anterior open bite are posterior vertical maxillary excess and a steep mandibular plane angle. Consequently, the basic principle underlying correction of anterior open bite involves changing the maxillary occlusal plane (ie, posterior shortening or anterior lengthening), allowing for rotation of the mandible (either as autorotation or an osteotomy with counterclockwise rotation), with resultant flattening of the mandibular plane. The amount of posterior shortening needed to achieve open bite closure can dictate whether operative correction is required.

Although surgical correction of anterior open bite is a common operation performed by craniomaxillofacial surgeons, there is a dearth of the literature regarding the geometry of the correction and empiric basis for correlating the magnitude of the open bite to the magnitude of the movement required for correction. For most craniomaxillofacial procedures correcting skeletal deformities, the three-dimensional skeletal movements are complex and mathematical modeling is beyond reasonable comprehension by most clinicians. This is particularly true for bilaterally asymmetric deformities. However, anterior open bite constitutes a bilaterally symmetric deformity that can often be corrected by a Le Fort I–level osteotomy with rotation of the mandible about a single axis through the condyles. These relatively simple movements can be modeled mathematically, allowing for an understanding of the degree of movements required to correct the open bite.

The purpose of this technical note was to describe a mathematical model that simulates the most common skeletal movements performed in surgical correction of an open bite: posterior maxillary impaction and mandibular rotation. Specifically, this model describes the geometric relationship between posterior maxillary impaction and anterior open bite closure and defines the variables contributing to this relationship. The mathematical model was then applied to a clinical case for validation.

TECHNIQUE

The assessment of a patient with anterior open bite begins with clinical and radiographic examinations. As such, a mathematical model describing the dynamic geometry of the maxillomandibular complex can be created on the basis of the sagittal jaw relationship captured on a standard lateral cephalogram. Three cephalometric points are used: the condylion (*Co*), the lower incisor tip (*LI*), and the most posterior molar contact (*M*). Two hypothetical circles are then drawn, each centered at *Co*. The 2 circles intersect points *M* and *LI*, respectively (Fig. 1).

The mathematical model uses novel cephalometric measurements including a surrogate of mandibular size, mandibular plane angle, and amount of mandibular rotation to predict the amount of posterior vertical impaction required to achieve a given vertical increase in the lower incisor tip position (Table 1). The mathematical model is described in Figures 2 and 3 as well as in Table 2.

The mathematical model described was embedded in an electronic spreadsheet (Excel 2011; Microsoft Corp, Redmond,

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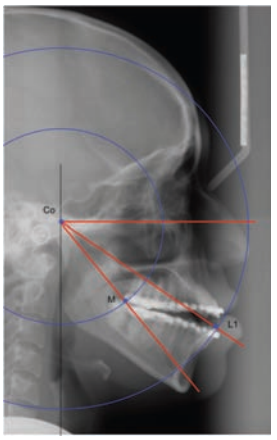

FIGURE 1. Schematic planning for mathematical modeling of open bite correction. Three cephalometric points were used: the condylion (*Co*), the lower incisor tip (*L1*), and the most posterior molar contact (*M*). Two circles centered around *Co* are shown passing through *M* and *L1*.

WA), and variables *Co-M*, *Co-L1*, α_{L1}, α_M, and the amount of rotation were varied. The trends were as follows:

1. As the radius increases, the vertical change for a given rotation increases.
2. As the position on the circle approaches the zero-degree position (decreased α), there is a greater vertical change for a given rotation.
3. The ratio of vertical change at *L1* relative to *M* increases with increasing α discrepancy (α_M-α_{L1}) and increasing *Co-L1*/*Co-M* ratio.

The previously mentioned patterns carry implications for clinical practice. The radius *Co-L1* is a surrogate for the mandibular size, and α_{L1} is a surrogate for the mandibular plane angle. Of note, many patients with anterior open bites also have hypoplastic mandibles with steep mandibular plane angles, both of which decrease the amount of vertical change at the lower incisor tip for a given mandibular rotation (Fig. 3).

An illustrative case is used herein to demonstrate the mathematical model created. A 15-year-old adolescent girl was referred by her orthodontist for evaluation of an anterior open bite. Examination results revealed a symmetric face with incompetent lips at rest. She had 5 mm of open bite at the central incisors (overjet = 0) and Angle class III molars bilaterally. The mandibular dental midline was 2 mm to the left of the maxillary dental midline (Fig. 4).

Cephalometric analysis was notable for a normal *Sella-nasion-A point* (83 degrees), but a short maxillary length (condylion to *anterior nasal spine* of 83.9 mm). The mandible was normal in size (*Sella-nasion-B point*, 81 degrees; condylion to gonion, 121 mm) but with a steep mandibular plane angle (35 degrees). Overbite and overjet were consistent with clinical measurements. The upper and lower incisors were protrusive (*upper incisor tip–Sella-nasion*, 115 degrees; *L1-Nasion-B point*, 43.6 degrees). The

TABLE 1. Cephalometric Variables and Definitions

Cephalometric Variable	Definition
M	Posterior molar contact
L1	Lower incisor tip
Co	Superior aspect of the condyle
α	Angle the radius makes with the horizontal axis (Fig. 2)
h	Vertical distance from a cephalometric point to horizontal passing through *Co*.
r	Radius from *Co* to the cephalometirc point.
Δh	Change in the vertical position

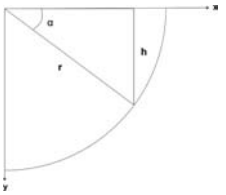

FIGURE 2. Measurement of vertical height (*h*) from the horizontal axis as a function of the angle of condylar rotation (α) and radius (*r*), $h = r \times sin(\alpha)$. The vertical position of *M* or *L1* relative to the horizontal axis through *Co* is computed by the equation $h = r \times sin(\alpha)$, where *h* is the vertical distance from the horizontal axis through the condylion, *r* is the radius or distance from the condylion, and α is the angle the radius makes with the horizontal axis.

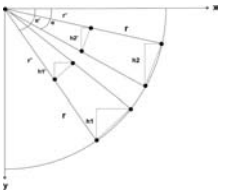

FIGURE 3. Schematic representation of changes in vertical height as a function of position along the mandibular arc of rotation (α) and distance from the condyle (*r*). For a given rotation, change in vertical position increases with decreasing α and increasing *r*.

ratio of the posterior facial height to the anterior facial height was decreased (posterior facial height/anterior facial height, 54%; 2.7 SD below normal), indicating a skeletal etiology for the anterior open bite (Fig. 5A).

The skeletal diagnoses included posterior vertical maxillary excess, maxillary sagittal hypoplasia, and anterior open bite. The surgical plan executed included a Le Fort I osteotomy with 4 mm of posterior maxillary impaction and 5 mm of sagittal maxillary advancement with autorotation of the mandible to achieve an overbite of 2 mm (Fig. 5B).

The index case described herein had *Co-M* of 66 mm, *Co-L1* of 104 mm, α_M of 44 degrees, and α_{L1} of 35 degrees. According to our mathematical model, for an estimated 5-degree rotation, 4.3 mm of posterior impaction would be required at point *M* to achieve 7.4 mm

TABLE 2. The Mathematical Model

Steps	Geometric/Trigonometric Equations
1. Calculate the vertical position of the most posterior molar contact and the lower incisor tip relative to the horizontal axis through the condyle.	$h = r \times sin(\alpha)$ $h_M = Co - M \times sin(\alpha_M)$ $h_{L1} = Co - L1 \times sin(\alpha_{L1})$
2. Determine the change in the vertical position for a given rotation for both *M* and *L1*.	$\Delta h = r \times sin(\alpha) - r \times sin(\alpha - rotation)$ $= r(sin(\alpha) - sin(\alpha - rotation))$ $\Delta h_M = CoM(sin(\alpha_M) - sin(\alpha_M - rotation))$ $\Delta h_{L1} = CoL1(sin(\alpha_{L1}) - sin(\alpha_{L1} - rotation))$
3. Determine the increase in the vertical position at the lower incisor tip relative to the vertical increase at the posterior molar contact.	$\frac{CoL1(sin(\alpha_{L1}) - sin(\alpha_{L1} - rotation))}{CoM(sin(\alpha_M) - sin(\alpha_M - rotation))}$

First, the vertical position of *M* or *L1* relative to the horizontal axis through *Co* is computed (step 1). Second, the change in the vertical position for a given rotation is calculated. Substituting *Co-M* and *Co-L1* for radii yielded vertical height change at the posterior molar contact and the lower incisor, respectively (step 2). Finally, the ratio of vertical height change at the posterior molar contact and the lower incisor can be computed (step 3). On the basis of the previous equations, the amount of vertical change in position depends on the radius, the position on the circle (α), and the amount of rotation.

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FIGURE 4. A to C, Preoperative occlusion, demonstrating an anterior open bite of 5 mm, Angle class III molar and canine relationships, as well as an overjet of 0 mm.

FIGURE 5. A, Preoperative lateral cephalogram demonstrating a class III skeletal relationship, anterior open bite, and steep mandibular plane angle. B, Postoperative lateral cephalogram after Le Fort I osteotomy, posterior impaction of 4 mm, and advancement of 5 mm, resulting in the shortening of posterior facial height, flattening of the mandibular occlusal plane, and closure of the open bite.

of vertical movement at *L1*. This model is consistent with the empiric treatment rendered, which included 4-mm posterior impaction with rotation achieving 7 mm of overbite closure (+2 mm from a preoperative measurement of −5 mm).

DISCUSSION

On the basis of our calculations, the ratio of vertical height change at *L1* versus *M* was 7.4/4.3, or 1.72. In a 2008 study evaluating posterior occlusal adjustment as an approach to treat open-bite malocclusion, Janson et al[5] found a 2.38-mm increase in overbite for a 1.29-mm reduction of enamel at the second molars. This corresponds to a ratio of 1.84 mm of open bite closure for every 1 mm of posterior maxillary impaction, which is consistent with the findings herein.

An additional phenomenon was identified from this model. Sagittal maxillary advancement along the Frankfort horizontal (FH) plane in a patient with an occlusal plane that diverges from FH allows for mandibular autorotation, even without any posterior impaction (Fig. 6). In the context of the model presented, this will increase the total amount of rotation and the amount of open bite closure. This should be taken into account in a patient with maxillary sagittal hypoplasia and anterior open bite.

Although this model is based on a lateral cephalometric radiograph and carries attendant limitations of magnification, parallax, distortion, and superimposition of structures, this method is important in light of concerns regarding the effects of radiation associated with computed tomographic (CT) scans. When CT scans are used for virtual treatment planning, however, the mathematical formulations derived herein remain applicable.[6] This is because, although the mandible is able to move with multiple degrees of freedom around its diarthrodial joint system, the movement of the mandible while in centric relation (reference position used for surgical planning) is limited to an arc of rotation in the sagittal plane, to which our geometric model applies. The disadvantage of the virtual environment is the inability to "see" the movements in the same manner as one would on a hinge-type articulator when doing model surgery. As orthognathic surgical treatment planning becomes increasingly virtual, a firm understanding of the mathematical basis for complex movements will serve the surgeon well.

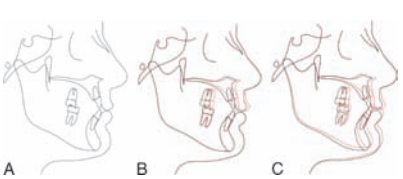

FIGURE 6. Maxillary advancement along the FH plane rather than along the occlusal plane allows for closure of an anterior open bite. Note the increased distance between the posterior molars after straight advancement without impaction (A, B), which allows for rotation of the mandible (C).

Of note, this model does not serve to predict the precise skeletal movements required to correct the open bite. Only the surgical manipulation in the operating room setting can determine the exact movements. Preoperative determination of the amount of posterior impaction required can be obtained using model surgery or specialized CT or cephalometric analysis software. The mathematical model described previously does not serve to replace these simulations. Practically, preoperative simulation for single-jaw procedures is largely academic because the maxilla can only be moved to occlude with the uncut mandible. The maxillomandibular complex can then be rotated around the condyles to achieve the appropriate maxillary incisor show. Nevertheless, the mathematical model provides insight for the surgeon when first examining patients with anterior open bite. A surgeon examining 2 patients with the same amount of open bite, one with a large mandible and normal mandibular occlusal plane and the other with a short mandible and steep mandibular occlusal plane, should anticipate that more impaction will be required for the second patient.

In summary, the amount of open bite closure for a given amount of posterior maxillary impaction depends on anatomic variables, which can be obtained from a lateral cephalogram. The clinical implication of this relationship is as follows: patients with small mandibles and steep mandibular occlusal planes will require greater amounts of posterior impaction. Clinical examinations and cephalometric radiographs can easily identify these features and help assist in treatment planning and patient counseling.

REFERENCES

1. Reyneke JP, Ferretti C. Anterior open bite correction by Le Fort I or bilateral sagittal split osteotomy. *Oral Maxillofac Surg Clin North Am* 2007;19:321–338
2. Sherwood K. Correction of skeletal open bite with implant anchored molar/bicuspid intrusion. *Oral Maxillofac Surg Clin North Am* 2007;19:339–350
3. Solano-Hernández B, Antonarakis GS, Scolozzi P, et al. Combined orthodontic and orthognathic surgical treatment for the correction of skeletal anterior open-bite malocclusion: a systematic review on vertical stability. *J Oral Maxillofac Surg* 2013;71:98–109
4. Greenlee GM, Huang GJ, Chen SS, et al. Stability of treatment for anterior open-bite malocclusion: a meta-analysis. *Am J Orthod Dentofacial Orthop* 2011;139:154–169
5. Janson G, Crepaldi MV, de Freitas KM, et al. Evaluation of anterior open-bite treatment with occlusal adjustment. *Am J Orthod Dentofacial Orthop* 2008;134:10–11
6. Caloss R, Atkins K, Stella JP. Three-dimensional imaging for virtual assessment and treatment simulation in orthognathic surgery. *Oral Maxillofac Surg Clin North Am* 2007;19:287–309

Recurrent Cherubism in an Adult Patient

Nihat Demirtas, DDS, Oya Barut, PhD, DMD,† Ilknur Ozcan, PhD, DMD,‡ Suzan Bayer, DDS,* and Hakki Oguz Kazancioglu, PhD, DDS**

Abstract: Cherubism is an uncommon, nonneoplastic, fibro-osseous disorder of the jaws in childhood and adolescence. It affects the jaw bones by deforming the cortical shell. Clinical features include

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progressive painless and mostly bilateral expansion of the mandible and/or maxilla. Because fibrous connective tissue replaces osseous tissue, radiographic features generally include expansile osteolytic lesions and a ground-glass appearance. Several treatment protocols for cherubism have been recommended in the literature; however, despite surgical curettage treatment, recurrences may occur.

Our aim was to emphasize the high recurrence rate of cherubic lesions. In this article, we present cherubism in a young girl that relapsed after 5 surgical operations before her appearance to our clinic.

Key Words: Cherubism, surgery, treatment, recurrence

Cherubism is a rare, benign disease characterized by bone degradation and most often symmetric involvement of the mandible and/or maxilla. In most cases, it occurs in the mandible and occurrence frequency is higher in males compared with females. With the maxillary involvement, the floor of the orbit and the anterior wall of the maxillary sinus are affected by the disease.[1]

It manifests usually in children between 12 and 36 months of age with the painless, progressive enlargement of the jaws and the cheeks, frequently associated with dental malformations.[2] The condition may progress until puberty and exhibits partial or complete spontaneous regression in adulthood.[3] Cherubism was initially characterized as familial, particularly as a form of craniofacial fibrous dysplasia; however, its etiology has remained unclear. Both hereditary and sporadic cases have now been reported, and recent genetic studies have shown cherubism to be a separate entity at the molecular level.[4] It has been suggested that the possible etiological pathway is related to an autosomal dominant pattern by *SH3BP2* gene mutations on chromosome 4p16.3.[5]

The management of the disease is mostly conservative because it shows regression by puberty. Surgical correction of bone malformations is performed through local curettage and jaw contouring. Intralesional steroid injections and systemic administration of calcitonin have been recommended to promote regression of the lesions. In either case, the age of the patient affects the successful outcome of the treatment.[3,6]

In this article, we report a cherubism case in a young girl who underwent a number of surgical procedures that were complicated by relapse. In this case, both the mandible and the maxilla were affected by the disease. Moreover, dental conditions such as missing or malformed permanent teeth and malocclusion were present.

CLINICAL REPORT

A 20-year-old female patient presented to the Department of Oral and Maxillofacial Radiology, Faculty of Dentistry, Istanbul University, with a complaint of asymmetric involvement of her jaws.

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FIGURE 1. A and B, Facial view and anterior open bite deformity of a patient. C, Plaster cast of the patient showing disharmony of the dental arches. D, Lateral view of the patient showing the interocclusal relationship and decreased occlusal vertical dimension in the molar area.

She had esthetic and functional concerns regarding her teeth. During the extraoral examination, it was observed that the patient's face had characteristic cherubic features including painless bilateral fullness of the cheeks and a bilateral expansion of the posterior mandible (Fig. 1A). Clinical investigation revealed extensive dentoalveolar deformities, skeletal anterior open bite accompanied by protraction of the premaxillary segment, and posterior crossbite (Figs. 1B, D). Diagnostic plaster casts were obtained, and centric relation record was detected by using facebow transfer (Fig. 1C). As a result, the patient had a speech deficiency and deformed alveolar bone crest, with missing teeth and disharmony of both dental arches.

An initial panoramic radiograph of the patient revealed variable, bilateral, expansive, and radiolucent lytic lesions with a ground-glass appearance involving both the mandible and the maxilla (Fig. 2A). Some of her permanent teeth were missing, and all of the present teeth were malformed. In addition, impacted and dilacerated teeth were detected. Dental volumetric computed tomographic images showed cortical bone expansion and abnormal bone formation in the coronal and axial sections (Figs. 3A, B). Focal perforations in both jaws were observed in three-dimensional reconstructed images (Figs. 3C, D). The patient reported that the lesions in the maxilla and the mandible were operated on 5 times before her appearance to our clinic. However, there was recidivism of the lesions after all of the operations. An incisional biopsy was performed, and characteristic lesions of cherubism were reported according to the histopathologic examination. Serum levels of the patient were in the reference range. In the anamnesis, it was registered that no family member of the patient had the disease.

Because of the high incidence of lesion recidivism, the patient has not been approved for invasive treatments. The patient consulted with prosthodontists for fixture prosthesis to determine the noninvasive methods. However, it was not possible because of excessive dentoalveolar defects and skeletal malocclusion. Hence, radiologic and clinical follow-up of the patient was approved. Throughout the 4-year follow-up period, the control radiographs indicated no regression and there was no progression of the lesions (Fig. 2B).

DISCUSSION

Several studies have reported that regression of the lesions and spontaneous regeneration of the bone may occur in patients with

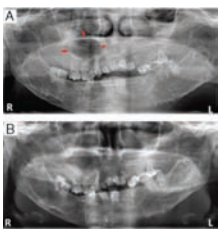

FIGURE 2. A, Initial panoramic radiograph of the patient showing widespread multilocular radiolucencies; red arrows show the bone cavity caused by surgical interventions. B, Panoramic radiograph after a 4-year follow-up; the lesions are similar to the initial radiograph.

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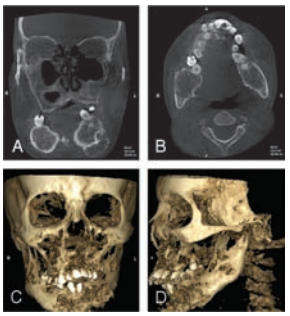

FIGURE 3. A and B, Dental volumetric computed tomographic scans show multiloculated lytic lesions affecting the body and rami of the mandible and maxilla on the coronal and axial sections. C and D, Three-dimensional reconstructed images show focal perforations in both jaws.

cherubism at young ages.[7,8] Throughout our patient's 4-year follow-up period, we observed neither radiographic regression nor progression of the lesions. Regression potential might be reduced after puberty, or surgical interventions might affect the mechanisms of bone repair in our case.

Early studies have reported cherubism to be a genetic disease with autosomal dominant inheritance and a variable degree of penetrance.[3–6] Therefore, familial occurrence has been one of the most common clinical investigations of cherubism.[9] Ueki et al[5] reported *SH3BP2* mutations that cause increased osteoblastic and osteoclastic activity in 66 patients. Henceforth, correct gene encoding is necessary for normal tooth eruption and bone formation. In our case, there was no history of cherubism in any of the patient's family members. This case may be an example of sporadic case of cherubism that was not inherited by parents but arose through a mutation.

The radiologic appearance of cherubic lesions is characteristic, with bilaterally symmetric, well-defined, multilocular radiolucencies in the mandible that extend from the region of the molar teeth toward the midline. However, Mani et al[10] reported unilateral involvement of the disease. The case presented herein showed similar characteristics with classic radiographic appearances of cherubism. In addition, the panoramic radiograph revealed diffuse-bordered radiolucent defects on the right side of the upper jaw that might have developed after recurrent surgical interventions.

There are many difficulties associated with the prosthetic rehabilitation of patients with cherubism. Khojasteh et al[11] reported a successful rate of dental implants with onlay bone augmentation during the 2-year follow-up of a patient with cherubism. However, in our case, the skeletal arch relationships and limited bone formation may not be suitable for implant placement. We observed that the correction of the skeletal malformations is the most important aspect for providing the patients' esthetic needs.

CONCLUSIONS

In this case, we concluded that a number of surgical procedures resulted in negative effects on the management of cherubism and the patient's quality of life. Therefore, specialists need to evaluate teenage patients with cherubism carefully when considering surgical management and, perhaps, consider intralesional steroid injections or systemic administration of calcitonin instead. In addition, a noninvasive prosthetic approach can be considered as the functional and esthetic rehabilitation of patients with cherubism.

REFERENCES

1. Daniels JS. Recurrent calcifying odontogenic cyst involving the maxillary sinus. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2004;98:660–664
2. Jain V, Sharma R. Radiographic, CT and MRI features of cherubism. *Pediatr Radiol* 2006;36:1099–1104
3. Etoz OA, Dolanmaz D, Gunhan O. Treatment of cherubism. *Eur J Dent* 2011;5:486–491
4. Novack DV, Faccio R. Jawing about TNF: new hope for cherubism. *Cell* 2007;128:15–17
5. Ueki Y, Tiziani V, Santanna C, et al. Mutations in the gene encoding c-Abl-binding protein SH3BP2 cause cherubism. *Nat Genet* 2001;28:125–126
6. Fernandes Gomes M, Ferraz de Brito Penna Forte L, Hiraoka CM, et al. Clinical and surgical management of an aggressive cherubism treated with autogenous bone graft and calcitonin. *ISRN Dent* 2011;2011:1–6
7. Timosca GC. Cherubism: regression of the lesions and spontaneous bone regeneration. *Rev Stomatol Chir Maxillofac* 1996;97:172–177
8. Hyckel P, Berndt A, Schleier P, et al. Cherubism—new hypotheses on pathogenesis and therapeutic consequences. *J Craniomaxillofac Surg* 2005;33:61–68
9. Mortellaro C, Bello C, Lucchino AG, et al. Diagnosis and treatment of familial cherubism characterized by early onset and rapid development. *J Craniofac Surg* 2009;20:116–120
10. Mani S, Natarajan B, Rajaram K, et al. Rare form of cherubism: case report with review of literature. *J Pharm Bioallied Sci* 2013;5:142–146
11. Khojasteh A, Sadr SJ, Saboury A, et al. Onlay bone augmentation and open sinus lifting with simultaneous implant placement in a cherubic patient. *J Craniofac Surg* 2014;25:193–196

Dehiscence of the Infraorbital Canal With the Maxillary Antral Empyema: A New Cause of Facial Pain

Jae Ki Kim, MD, Seong Kyeong Yang, MD, Dan Bi Shin, MD, and Jung Gwon Nam, MD, PhD

Abstract: The infraorbital nerve is the largest cutaneous branch of the maxillary divisions of the trigeminal nerve. It may produce a bony ridge on the antral roof but usually goes through within the maxillary bone as a discrete canal. Rarely, it could be partially or completely dehiscent, lying submucosally on the antral roof as in this case.

We describe a case of longstanding facial pain because of dehiscence of the infraorbital canal associated with the maxillary antral empyema. Endoscopic sinus surgery was successful in relieving the symptom.

Key Words: Facial pain, dehiscent infraorbital canal, maxillary antral empyema

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The nerve, now called the infraorbital nerve, travels anteriorly in the base of the orbit through the infraorbital canal and emerges from the infraorbital foramen onto the face reaching the skin. Once the nerve exits the infraorbital foramen, it divides into 4 branches, which only include the sensory nerve of the face between the lower eyelid and the upper lip.[1]

Facial pain in the distribution of the infraorbital nerve, which has no obvious causes to explain their symptoms, is sometimes associated with dehiscence of the infraorbital canal.[2] Although the antral wall of the infraorbital canal is particularly thin, the dehiscent infraorbital canal is rare. But it has been described that the dehiscent infraorbital canal might be a cause of facial pain due to the nerve being exposed to inflamed mucosa.[3]

Extensive studies of ostial size and patency within the antrum have clearly shown that negative-pressure changes occurred when the ostial diameter fell below a critical level. In literature, it was described that this negative pressure could make either facial pain or vacuum headache.[4]

We describe a case of dehiscence of the infraorbital canal with the maxillary antral empyema causing facial pain with a large-enough ostium of the maxillary sinus.

CASE REPORT

A 43-year-old woman presented with a history of longstanding left-sided facial pain in the distribution of the infraorbital nerve 5 months ago. The pain was dull and had to be endured throughout the day. The patient has complained of facial pain, although she had previously received endoscopic sinus surgery twice at a primary clinic. But she did not complain of other rhinologic symptoms such as nasal obstruction, rhinorrhea, and hyposmia. Endoscopic examination showed slightly polypoid mucosa in both maxillary antra, but any other abnormality was unremarkable. Computed tomography (CT) revealed soft-tissue density in both maxillary sinuses, both frontal sinuses, and left sphenoid sinus. On the coronal plane of the CT scan, we could find the route of the infraorbital nerve in left maxillary sinus (Fig. 1). We diagnosed that the recurred chronic rhinosinusitis with the facial pain occurred by inflammatory mucosa of the maxillary sinus. The patient underwent endoscopic sinus surgery under general anesthesia. We removed the inflammatory air cells and mucosa in both ethmoid sinuses, both frontal sinuses, and left sphenoid sinus. Both ostia of maxillary were large enough. Endoscopy showed that the infraorbital nerve was completely exposed in the maxillary antrum and the antral mucosa polypoid. The antral empyema of the left maxillary sinus, furthermore, was present in the end of the dehiscent infraorbital canal (Fig. 2A). It was felt in a surgical ground that the facial pain could be due to this empyema, which might irritate the infraorbital nerve. We conducted inferior meatal antrostomy to remove the mass because we could not reach it via middle meatal antrostomy. The mass was completely removed. After removal of the mass, repeated normal saline irrigation mixed with antibiotics was done. We could also detect the dehiscent infraorbital canal in the right maxillary sinus. There was a mild inflammation similar to left maxillary sinus, but no empyema was found, unlike left maxillary sinus (Fig. 2B). On the next day, the patient was discharged without any serious complications of the operation. One week after the operation, the patient's facial pain was gradually relieved. On the following month, endoscopy showed normal mucosa in the left maxillary sinus without any visible empyema (Fig. 3). She had nothing to complain in regard to the facial pain.

FIGURE 1. Coronal plane of CT. We could find the route of the infraorbital nerve in left maxillary sinus (arrow). Soft tissue density is seen in both maxillary sinuses.

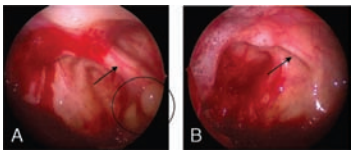

FIGURE 2. A, Endoscopic view (4 mm, 70 degrees) of the left maxillary sinus via middle meatal antrostomy shows the infraorbital nerve in the roof of the sinus (arrow) and empyema in the end of the dehiscent infraorbital nerve (circle). B, Endoscopic view (4 mm, 70 degrees) of the right maxillary sinus via middle meatal antrostomy shows the infraorbital nerve in the roof of the sinus(arrow).

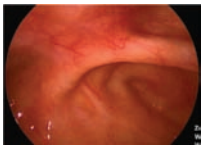

FIGURE 3. At 1 month after operation, endoscopy (4 mm, 70 degrees) showed normalized mucosa in left maxillary sinus and no recurrence of the maxillary antral empyema.

DISCUSSION

The infraorbital canal lies in the orbital part of the maxilla and allows the passage of the infraorbital nerve. The infraorbital nerve, a branch of the maxillary division of the trigeminal nerve, is responsible for sensory innervation to the part of the malar between lower eyelid and upper lip. It traverses the antral roof but is usually completely encased in a bony canal. The antral wall of the infraorbital canal is particularly thin with an average thickness of 0.2 mm, and the thickness of the antral wall depends on the age and the degree of pneumatization of the maxillary sinus. It is reported that the incidence of the complete dehiscent infraorbital canal is between 12% and 16% in dried skull specimens.[5,6]

When the infraorbital nerve canal is completely dehiscent, it may be easily irritated by mucosal inflammation or mass as shown in our case. The causes of the facial pain in the distribution of the trigeminal nerve are numerous and varied; hence, diagnoses may be made in the absence of positive clinical findings. However, this case shows that there may be an underlying anatomical cause that the maxillary antral empyema is irritated and partially compresses the dehiscent infraorbital nerve in the maxillary sinus.

The mechanism of action for the facial pain developed by the dehiscent infraorbital nerve is not so obvious. But that changes in atmospheric pressure can cause barotrauma in head and neck is well documented.[7] Some authors suggested that variation in the pressure within the antrum and poor ventilation caused by a small sinus ostium might also be a contributing factor.[3] Other authors implicated vasodilatation of antral mucosal blood vessels as a cause of the positional discomfort that might occur with the maxillary ostial occlusion.[8] Sessle et al[9] suggested that the trigeminal primary afferent fibers were terminated in the nasal mucosa as free nerve endings and activated by noxious stimuli. These noxious stimuli might include mechanical stimuli and chemical irritants. Sharma et al[2] proposed that maxillary antral cyst, which compressed the exposed infraorbital nerve, might be a cause of both recurrent facial pain and paresthesia when flying. They proposed that the decreased atmospheric pressure experienced during flying led to an expansion in the cyst size. This then compressed the exposed infraorbital nerve, leading to facial pain and paresthesia. They found that, once

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the cyst had been surgically removed, the patient ceased to experience further symptoms.[2] In our case, both maxillary ostia were large enough not to induce negative pressure. Also, infectious degree of both maxillary sinuses was similarly mild, but patient complained of only left-sided facial pain. We propose that dehiscence of the infaorbital canal with the maxillary antral empyema could be an irritating factor that in turn explains the facial pain is due to chemical irritants and mechanical stimuli such as partial compression of the dehiscent infraorbital nerve.

Treatments of facial pain induced by the dehiscent infraorbital nerve can vary. Initial attempt at surgical cure is performance of either inferior meatal or middle meatal antrostomy. Negative pressure caused by small ostium is cured by this surgical method.[4] Normal saline irrigation is also effective in inflamed antral mucosa. Repeated irrigation of the antrum can relieve the discomfort.[3] The infraorbital neuropexy using the subdermal fat graft or the temporalis fascial graft is also a possible treatment regimen.[4] This procedure was developed to provide additional protection for the dehiscent infraorbital nerve. A subdermal fat graft harvested from the abdominal or postauricular skin is placed between the mucosa of the antrum and the infraorbital nerve by a sublabial approach. In our case, we supposed that the facial pain was caused not only by the dehiscent infraorbital nerve but also by the empyema placed near the infraorbital nerve. Therefore, in this case, surgical removal of the maxillary antral empyema was considered as the treatment of choice. We found that the facial pain ceased after surgical removal of the maxillary antral empyema via endoscopic sinus surgery. We could not improve the pathologic confirmation because of the small size of the mass. Empyema was removed by microdebrider through the inferior meatal antrostomy. But considering the appearance and the condition of mass formation, we could not suspect other causes of facial pain except the dehiscent infraorbital canal associated with the maxillary antral empyema.

In our case, the patient had previously received endoscopic sinus surgery twice, but the facial pain was not resolved. We thought that previous operators could not identify the dehiscent infraorbital canal with the maxillary antral empyema. Although the infraorbital nerve is not routinely visualized during endoscopic sinus surgery, it can often be identified by passing a 30- or 70-degree telescope through the middle meatal antrostomy. It could not be indentified without a careful attention. Therefore, although not recognized before, it is important to remember that dehiscence of the infraorbital canal with the maxillary antral empyema can be a possible cause of facial pain.

CONCLUSIONS

A dehiscent infraorbital canal with the maxillary antral empyema may be a cause of facial pain because exposed infraorbital nerve can be affected by the maxillary antral empyema through chemical irritants and partial mechanical compression. It should be included as one of the differential etiologies of facial pain.

REFERENCES

1. Leo JT, Cassell MD, Bergman RA. Variation in human infraorbital nerve, canal and foramen. *Ann Anat* 1995;177:93–95
2. Sharma N, De M, Pracy P. Recurrent facial paraesthesia secondary to maxillary antral cyst and dehiscent infraorbital canal: case report. *J Laryngol Otol* 2007;121:e6
3. Whittet HB, Quiney RE. Dehiscence of the infraorbital nerve as a new cause of facial pain. *Br Med J (Clin Res Ed)* 1988;296:18–19
4. Whittet HB. Infraorbital nerve dehiscence: the anatomic cause of maxillary sinus "vacuum headache"? *Otolaryngol Head Neck Surg* 1992;107:21–28
5. Hollinshead WH. *Anatomy for regeon v.1: the head and neck*. New York: Harper & Row; 1968
6. Gray HGCM. *Anatomy of the human body*. Philadelphia, PA: Lea & Febiger; 1966
7. Mirza S, Richardson H. Otic barotrauma from air travel. *J Laryngol Otol* 2005;119:366–370
8. Falck B, Svanholm H, Aust R, et al. The relationship between body posture and pressure in occluded maxillary sinus of man. *Rhinology* 1989;27:161–167
9. Sessle BJ. Peripheral and central mechanisms of orofacial inflammatory pain. *Int Rev Neurobiol* 2011;97:179–206

Surgical Treatment of Severe Frontal Bone Fracture

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Abstract: Craniofacial trauma can lead to several complications. The combined fractures of anterior and posterior walls of the frontal bone are almost always followed by lesions in nasofrontal orifices and disruption of nasofrontal ostia or ducts, a significant factor for the development of early and late complications after sinus fractures. This article reports a case of trauma patient, who underwent neurological evaluation and at first showed good general condition. Computed tomography noted fracture of the anterior and posterior walls of the frontal sinus and small foci of pneumocephalus in the cerebral cortex. The patient was monitored periodically and 9 days after trauma showed increased areas of pneumocephalus in prefrontal cortex, cerebrospinal fluid draining, and large dura mater lesion, with signs of necrosis and inflammation (meningitis). The necrotic tissues were removed, and dura mater was repaired through the approximation with resorbable wire polyglactin 910 5-0, oxidized cellulose application, and bonding with human fibrin sealant (fibrinogen, thrombin, and calcium chloride). Sinusectomy, frontal sinus, and nasofrontal duct obliteration with pedicled pericranium flap were performed. Tomographically, a reanatomization was noted in frontal region, and a 12-month follow-up showed no complication. The use of fibrin glue to repair dura mater lacerations, as well as the pedicle pericranium flap for frontal sinus and nasofrontal duct obliteration, is an efficient method for treating fractures of the frontal bone.

Key Words: fractures, bone, cerebrospinal fluid, frontal sinus

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The craniofacial trauma can lead to several complications such as cerebrospinal fluid leakage, meningitis, mucocele, mucopyoceles, cerebral abscess, and encephalocele formation.[1] In general, anterior wall fractures are reconstructed, whereas the posterior wall fractures are treated by cranialization followed by dura mater repair and/or frontal sinus obliteration.[2–7]

Combined anterior and posterior wall fractures are almost always followed by injuries in nasofrontal orifices.[8–11] The rupture of nasofrontal ostiums or ducts is an important factor for the development of early and late complications after frontal sinus fractures.[12]

This article reports the importance of multidisciplinary treatment and monitoring of neurological patients with frontal sinus fracture, presenting meningitis and liquoric fistula, submitted to surgical treatment.

CASE REPORT

The patient was a 24-year-old man, a victim of motorcycle accident. Neurological evaluation showed the patient is in good general condition, conscious, oriented, eupneic, hydrated, ruddy, afebrile, and ambulating, and has a Glasgow Coma Scale (GCS) score of 15. Computed tomography (CT) of axial slices, fracture of anterior and posterior frontal sinus walls, and small foci of pneumocephalus in cerebral cortex were observed. Facing the clinical conditions, the neurosurgery team decided monitoring the patient periodically, with CT scans every 72 hours for observation until the surgical reconstruction in a second moment.

The staff of oral and maxillofacial surgery was triggered for evaluating the case and confirmed through the clinical and tomographic features, the fracture of the frontal bone, hemifracture Le Fort III on the left, and Le Fort II on the right side, besides symphysis fracture with plate fracture in mandibular body on the right side (Fig. 1).

The third CT scan, 9 days after trauma, showed increased areas of pneumocephalus in prefrontal cortex (Fig. 2), and clinically, consciousness level (GCS score = 10) decreased, with signs of abundant cerebrospinal fluid drainage in nasal cavity and oropharynx. Then, he was referred to the operating room; the craniotomy was performed by the neurosurgeon (Figs. 3A, B),

FIGURE 1. Skull CT scan, showing panfacial hemi–Le Fort III fracture on the left and Le Fort II on the right side, involving the anterior and posterior frontal bone walls.

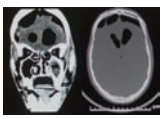

FIGURE 2. Coronal and axial CT scans showing large foci of pneumocephalus.

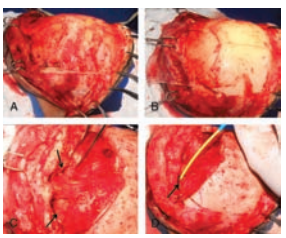

FIGURE 3. A, Surgical access and demarcation of pericranium flap. B, Frontal bone fracture was observed after lifting pericranium flap. C, A large dura mater lesion was observed after craniotomy (black arrows). D, Dura mater sealing was performed using human fibrinogen glue (black arrows).

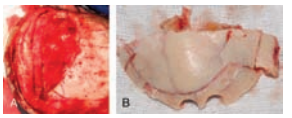

FIGURE 4. A, Frontal sinus and nasofrontal duct obliteration and pericranium flap previously confectioned. B, Fragment of the frontal sinus anterior wall and part of the frontal bone osteotomized to perform craniotomy, which was used to fixation and reanatomization of frontal region.

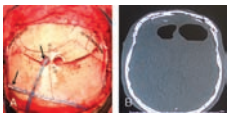

FIGURE 5. A, Fixation of frontal sinus anterior wall and part of frontal bone fragment. Arrows indicate the position of 2 suction drains (Portovac), either into the fixed bone, in contact with brain tissue, or externally, in contact with the access. B, Postoperative 1-day CT, showing fracture reduction and the presence of pneumocephalus, as expected for the first moments of postoperative.

which observed a large dura mater lesion, with signs of necrosis and inflammation (meningitis) (Fig. 3C, black arrows). The necrotic tissues were removed, and dura mater was repaired through the approximation with resorbable wire polyglactin 910 5-0, oxidized cellulose application, and bonding with human fibrin sealant (fibrinogen, thrombin, and calcium chloride) (Fig. 3D).

Sinusectomy, frontal sinus, and nasofrontal duct obliteration with pedicled pericranium flap were performed (Fig. 4A). The fragments fractured and removed for craniotomy (Fig. 4B) were repositioned and fixed with miniplates and titanium screws of 1.5-mm system (Fig. 5A). A Portovac suction drain was installed and maintained for 24 hours in order to drain possible hematomas or edema (Fig. 4A, black arrows). Postoperatively, the patient restored the level of consciousness within the first days (Fig. 5B). A 12-month follow-up showed no complication.

DISCUSSION

In order to select the best treatment for frontal sinus fractures, a detailed initial diagnosis[13] is necessary. According to Castro et al,[14] the main goals of preoperative examinations are to determine if there was involvement of anterior, posterior, or both osseous plates; significant displacement; nasofrontal duct obstruction; and cerebral vascular lesion with dura mater involvement, and the best way to observe the output of nasofrontal duct is through CT of thin slices in high-resolution.

Also, some signs or symptoms of more severe complications of these fractures may be subclinical and appear later.[15–18] This might explain the delay in the onset of symptoms in this case, because only the third CT, performed 9 days after trauma, showed increased pneumoencephalus, affecting large part of prefrontal cortex, and clinically the patient presented cognition reduction, not responding to questions clearly (GCS = 10), and apparent drainage of cerebrospinal fluid, what led to immediate surgical approach for dura mater repair and frontal sinus reconstruction.

Surgical repair of dura mater lesion consists of a primary closure using absorbable suture thread; however, in the lacerations with substance loss, the reconstruction needs to be performed with grafts from muscular fascia, pericranium flap, or by glue.[8–11] In the current case, it was noted after craniotomy that dura mater presented laceration and necrosis caused by meningitis, besides active drain of cerebrospinal fluid. Therefore, the neurosurgeon adopted for dura mater repair, the removal of necrotic tissue, oxidized cellulose application, and fibrin glue. The cerebrospinal fluid leak draining rapidly stopped, allowing a fast and efficient surgical repair, with no need for grafts.

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The next step of surgical procedure in these fractures is the frontal sinus and nasofrontal duct obliteration.[19] Statkievicz et al[8] reported that the pedicled pericranium flap is a good choice for frontal sinus and nasofrontal duct obliteration, because of its high vascularization, what reduces the risk of infections. Moreover, this material is easy to obtain and presents no risk of morbidity to the donor site as well as the necessity of a second surgical intervention. In this case, pericranium flap was recommended as a method for frontal sinus obliteration, performed through the subgaleal plane.[20,21]

CONCLUSIONS

The use of fibrin glue to repair dura mater lacerations, as well as the pedicle pericranium flap for frontal sinus and nasofrontal duct obliteration, is an efficient method for treating fractures of the frontal bone.

REFERENCES

1. Gumussoy M, Ugur O, Cukurova I, et al. Recurrent meningitis and frontal encephalocele as delayed complications of craniofacial trauma. *J Craniofac Surg* 2014;25:529–530
2. Wolfe SA, Johnson P. Frontal sinus injuries: primary care and management of late complications. *Plast Reconstr Surg* 1988;82:781–791
3. Heller EM, Jacobs JB, Holliday RA. Evaluation of the frontonasal duct in frontal sinus fractures. *Head Neck* 1989;11:46–50
4. Helmy ES, Koh ML, Bays RA. Management of frontal sinus fractures. Review of the literature and clinical update. *Oral Surg Oral Med Oral Pathol* 1990;69:137–148
5. Rohrich RJ, Hollier LH. Management of frontal sinus fractures. Changing concepts. *Clin Plast Surg* 1992;19:219–232
6. Ioannides C, Freihofer HP, Friens J. Fractures of the frontal sinus: a rationale of treatment. *Br J Plast Surg* 1993;46:208–214
7. Ioannides C, Freihofer HP. Fractures of the frontal sinus: classification and its implication for surgical treatment. *Am J Otolaryngol* 1999;20:273–280
8. Statkievicz C, Faverani LP, Ramalho-Ferreira G, et al. Reconstruction of extensive frontal fracture with titanium mesh. *J Craniofac Surg* 2014;25:712–714
9. de Melo WM, Coléte JZ, Mariano RC, et al. Anterior pericranial flap for frontal sinus duct obliteration: is it a valuable resource? *J Craniofac Surg* 2013;24:147–149
10. Morais de Melo W, Koogi Sonoda C, Garcia IR Jr. Vascular pericranial graft: a viable resource for frontal sinus obliteration. *J Craniofac Surg* 2013;24:5–7
11. Kim YJ, Kim HR, Jun YJ, et al. Usefulness of vascularized galeal frontalis myofascial flap as treatment for postoperative infection in frontal sinus fracture. *J Craniofac Surg* 2011;22:1968–1971
12. Stanley R Jr. Management of severe frontobasilar skull fractures. *Otolaryngol Clin North Am* 1991;24:139–150
13. Tiwari P, Higuera S, Thornton J, et al. The management of frontal sinus fractures. *J Oral Maxillofac Surg* 2005;63:1354–1360
14. Castro B, Walcott BP, Redjal N, et al. Cerebrospinal fluid fistula prevention and treatment following frontal sinus fractures: a review of initial management and outcomes. *Neurosurg Focus* 2012;32:E1
15. Wallis A, Donald PJ. Frontal sinus fractures: a review of 72 cases. *Laryngoscope* 1988;98:593–598
16. Shumrick KA, Smith CP. The use of cancellous bone for frontal sinus obliteration and reconstruction of frontal bony defects. *Arch Otolaryngol Head Neck Surg* 1994;120:1003–1009
17. Correa AJ, Duncavage JA, Fortune DS, et al. Osteoplastic flap for obliteration of the frontal sinus: five years' experience. *Otolaryngol Head Neck Surg* 1999;121:731–735
18. Mendians AE, Marks SC. Outcome of frontal sinus obliteration. *Laryngoscope* 1999;109:1495–1498
19. Parhiscar A, Har-El G. Frontal sinus obliteration with the pericranial flap. *Otolaryngol Head Neck Surg* 2001;124:304–307
20. Donath A, Sindwani R. Frontal sinus cranialization using the pericranial flap: an added layer of protection. *Laryngoscope* 2006;116:1585–1588
21. Rocchi G, Caroli E, Belli E, et al. Severe craniofacial fractures with frontobasal involvement and cerebrospinal fluid fistula: indications for surgical repair. *Surg Neurol* 2005;63:559–563

The Importance of Histopathologic Analysis of Pericoronal Follicles for the Early Identification of Ameloblastomas

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Abstract: The objective of this study was to discuss the importance of performing histopathological examination of pericoronal follicles as a routine procedure, so as to enable the early identification of odontogenic lesions. We describe two clinical cases with histopathological diagnoses of ameloblastomas who did not show clinical or radiographic signs of disease before microscopic examination.

Key Words: odontogenic lesions, dental tissues, early diagnosis

CLINICAL REPORT

Patient 1

A 37-year-old female patient sought the clinic for the removal of an impacted third molar. Radiography revealed an impacted mandibular right third molar (tooth 48), in horizontal position. However, no abnormalities were detected during radiographic and physical examination. During the surgery, the presence of a follicle of unusual dimensions was observed. The report of the histopathologic analysis was ameloblastoma. The patient has been regularly followed up for over 10 years, with no clinical or radiographic signs of relapse.

Patient 2

A 17-year-old male patient sought the clinic with a complaint of increased volume in the mentonian region. During the anamnesis, the patient reported a previous surgical intervention on this specific region 5 years ago and was not able to confirm if the soft tissues associated with the referred tooth were histopathologically analyzed. The panoramic radiograph showed a radiolucent, multilocular area in the mentonian region, extending from the right first molar to the left second premolar, preserving the mandibular basal bone, expanding cortical bones, and showing resorption of the

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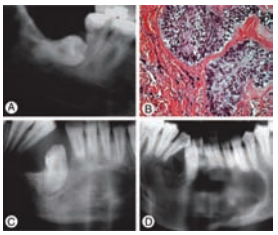

FIGURE 1. Panoramic radiograph showing an impacted mandibular third molar without radiographic abnormalities (A); hematoxylin-eosin stain section showing fibrous connective tissue with epithelial islands, columnar cells at the basal layer, in palisade arrangement, and loosely arranged central cells, resembling the stellate reticulum of the enamel organ (B, original magnifications ×200 and ×400). Panoramic radiograph showing a radiolucent image around tooth 43 at 5 years ago (C). Panoramic radiograph showing an extent radiolucent image in mentonian area (D).

roots of teeth. An incisional biopsy was performed, and the final diagnosis of solid ameloblastoma was established.

DISCUSSION

Histologically, pericoronal follicles present fibrous connective tissue, with a variable presence of epithelial remnants of the dental lamina and reduced enamel epithelium of the enamel organ. Depending on external stimuli, the structures comprising pericoronal follicles can show a proliferative potential.[3,4] Baumgart et al[4] and Meleti and van der Waal[3] have suggested that common odontogenic cysts may originate from reduced enamel epithelium, whereas odontogenic tumors originate from epithelial remnants of the dental lamina.

Alterations in epithelial remnants of pericoronal follicles characteristic of ameloblastoma have been described by Meleti and van der Waal[3] and Tegginamani and Prasad,[9] who analyzed patients with dentigerous cysts and pericoronal follicles. These authors found that the proliferation of remnants of odontogenic epithelium may present characteristics compatible with those of ameloblastomas, which the authors classified as "ameloblastomatous features." According to the authors, pericoronal follicles showing those features could be classified as "focal ameloblastomas." In our patient 1, the findings corroborate those of previous studies by showing a pericoronal follicle with no clinical or radiographic abnormalities, but with histopathologic characteristics of ameloblastoma.

This reinforces the importance of studying pericoronal follicles for the diagnosis of odontogenic cysts and tumors. Using histopathologic examination as a routine procedure in clinical practice would allow dental surgeons to identify histopathologic abnormalities earlier and thus diagnose the lesion at an early stage, avoiding the need for more invasive therapies, as is usually required for these tumors. In addition, in patient 2, we could speculate that the material extracted from the crown of the impacted tooth, which was probably discarded, may have already contained early signs indicative of the development of an aggressive lesion.

Our report corroborates the finding that most ameloblastomas originate from remnants of dental lamina. Therefore, tissues surrounding the crowns of impacted teeth, regardless of their clinical or radiographic appearance, should be histologically assessed. This strategy would enable the early identification of odontogenic cysts and tumors and the implementation of less invasive treatment approaches. The cases here described underscore the importance of establishing the microscopic diagnosis of any tissue removed from the oral cavity, so as to identify histopathologic findings of benign odontogenic tumors even when radiographic signs of disease are absent.

REFERENCES

1. Damante JH, Fleury RN. A contribution to the diagnosis of the small dentigerous cyst or the paradental cyst. *Pesqui Odontol Bras* 2001;1:238–246
2. Oliveira MG, Lauxen IS, Chaves ACM, et al. Odontogenic epithelium: immunolabeling of Ki-67, EGFR and survivin in pericoronal follicles, dentigerous cysts and keratocystic odontogenic tumors. *Head Neck Pathol* 2011;5:1–7
3. Meleti M, van der Waal I. Clinicopathological evaluation of 164 dental follicles and dentigerous cysts with emphasis on the presence of odontogenic epithelium in the connective tissue. The hypothesis of "focal ameloblastoma". *Med Oral Patol Oral Cir Bucal* 2013;18:60–64
4. Baumgart CD, Lauxen IS, Sant'Ana Filho M, et al. Epidermal growth factor receptor distribution in pericoronal follicles: relationship with the origin of odontogenic cysts and tumors. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2007;103:240–245
5. Cabbar F, Güler N, Comunoglu N, et al. Determination of potential cellular proliferation in the odontogenic epithelia of the dental follicle of the asymptomatic impacted third molars. *J Oral Maxillofac Surg* 2008;66:2004–2011
6. Villalba L, Stolbizer F, Blasco F, et al. Pericoronal Follicles of asymptomatic impacted teeth: a radiographic, histomorphologic, and immunohistochemical study. *Int J Dent* 2012;2012:1–6
7. Barnes L, Eveson JW, Reichart P, Sidransky D. *World Health Organization Classification of Tumours. Pathology and Genetics Head and Neck Tumours.* Lyon, France: IARC Press, 2005
8. More C, Tailor M, Patel HJ, et al. Radiographic analysis of ameloblastoma: a retrospective study. *Ind J Dent Res* 2012;23:698
9. Tegginamani AS, Prasad R. Histopathologic evaluation of follicular tissues associated with impacted lower third molars. *J Oral Maxillofac Pathol* 2013;17:41–44

Pure Endoscopic Endonasal Removal of Unusual Anterior Skull Base Aneursymal Bone Cyst Extending to the Frontal Lobe

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Abstract: Aneurysmal bone cysts (ABCs) are benign, nonneoplastic, hemorrhagic, and expansile osseous lesions that present most frequently at age younger than 20 years. Aneurysmal bone cysts typically involve long bones of extremities, thorax, pelvis, or spinal column. Skull base involvement is very rare. The authors report the case of a 23-year-old woman with ABC of the skull base and total removal of lesion with pure endoscopic endonasal approach. The patient had presented with nasal obstruction for 6 months. Physical

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and neurological examination findings were normal except for bilateral anosmia. Cranial magnetic resonance imaging (MRI) revealed a tumor occupying ethmoid sinuses anterior skull base that extended into bilateral frontal lobes. The patient underwent pure endoscopic endonasal surgery, and the tumor was resected gross-totally. Histologic examination revealed ABC. Consequently, ABC should be considered in differential diagnosis of skull base pathologies. Endoscopic endonasal surgical approach is a safe, minimally invasive, and effective way in the treatment of these tumors.

Key Words: Aneurysmal bone cyst, anterior skull base, endoscopic endonasal, frontal lobe

Aneurysmal bone cysts are members of benign fibrosseous lesion groups such as ossifying fibroma, fibrous dysplasia, and benign osteoblastoma.[1–4] Aneurysmal bone cysts were first described by Jaffe and Lichtenstien as a nonneoplastic, hemorrhagic, multiseptate cystic and expansile benign osseous lesions in 1942.[5–9] They are most commonly found in long bones of extremities, thorax, pelvis, and spinal column; and skull base involvement is very rare.[5,10] Most patients having ABCs are younger than 20 years.[5,6,11,12] According to Lichtenstien, ABCs could be lesions that developed secondary to circulation impairment as seen in venous thrombosis or arteriovenous malformation.[13,14]

Clinical findings of ABCs depend on affected region. In cranial involvement, headache, nasal obstruction, and cranial nerve paralysis are remarkable for ABCs.[12,13,15] Radiography of aneurysmal bone cysts generally shows an expansile cystic lesion with a honeycomb or a soap-bubble appearance.[1,16] Treatment options of ABCs include surgical resection, curettage, cryotherapy, sclerotherapy, and radiotherapy.[5,7,17–22] Many transcranial surgical approach with different succes rates, cosmetic problems, and reconstruction problems have been used for surgical resection of skull base ABCs until today.[1,5,11] Surgical resection with gross-total excision has perfect outcome and considered as best approach for treatment of ABCs.[1,11,23,24]

The authors present gross-total resection of a rare ABC that occupies anterior skull base and extends to bilateral frontal lobes with pure endoscopic endonasal surgical approach.

CLINICAL REPORT

A 23-year-old woman presented with impairment of the sense of smell. Neurological examination was remarkable for bilateral anosmia. Cranial magnetic resonance imaging and paranasal computed tomography revealed a multiseptated, trabeculated tumor, which occupied ethmoid sinuses and expanded through nasal cavity, anterior cranial fossa, and frontal lobes, with a diffuse heterogenous enhancement after gadolinium administration (Figs. 1A–C). Digital subtraction angiography was performed to reveal lesion vascularity and relation with cerebral vascular structures.

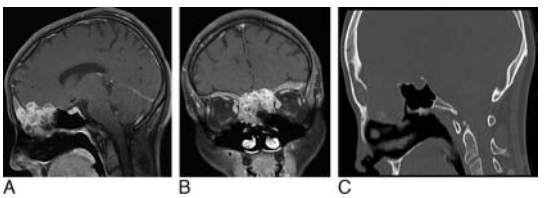

FIGURE 1. T1-weighted sagittal MRI (A) and T1-weighted coronal MRI (B) show a multiseptated, trabeculated mass lesion, which occupied ethmoid sinuses, expanded through nasal cavity and frontal lobes, with a diffuse heterogenous contrast enhancement. Sagittal reconstruction paranasal CT (C) shows the antreior skull base destruction and thin bone lamellas into the lesion.

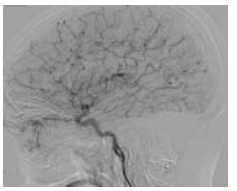

FIGURE 2. Sagittal digital substruction angiogram shows the intense tumor blush at late capillary stage.

Vascular supply of tumor arises from branches of bilateral external carotid artery; left branches dominated. In addition, intense tumor blush was demonstrated at late capillary stage (Fig. 2). No endovascular embolization was performed.

The patient underwent a binostril endoscopic endonasal transethmoidal transcribriform approach in supine position. Bilateral ethmoid sinuses were occupied by yellow-white, hemorrhagic tumor with multiple bony compartments. The tumor extended to anterior cranial fossa and eroded the dura. The tumor was resected gross-totally including the intracranial part. Both of the olfactory nerves were seen and preserved. After resection, free graft of fascia lata from the right thigh and fibrine tissue adhesives were used for skull base reconstruction. At the end, a foley catheter was placed for immobilization of reconstruction materials. External lumbar drainage catheter was replaced. No nasal packing was used.

Postoperative cranial computed tomography and cranial magnetic resonance imaging showed the gross total removal of the tumor (Fig. 3). Nasal foley catheter was removed at postoperative second day, and the external lumbar drainage catheter was removed at postoperative third day. The postoperative course was uneventful, and the patient was discharged at postoperative fifth day. Olfactory function was intact after surgery. She did not receive postoperative radiotherapy. No recurrence was observed after 10 months.

Histopathological examination showed cavernous spaces filled with blood. The spaces were separated by collagenous tissue containing fibroblasts, focal collections of osteoclast, hemosiderin-laden macrophages, and reactive bone formation (Fig. 4). Histological evaluation confirmed that the lesion was an ABC.

DISCUSSION

Aneurysmal bone cyst is a nonneoplastic lesion that is seen rarely. Aneurysmal bone cyst presents most frequently during the first 2 decades. Pathogenesis of ABC include several stages; the first stage is composed of osteolysis and periosteal widening.[5] Progressive bone destruction constitute second stage that the lesions grow rapidly, after that lesions get more stabile stage.[5] Typically, ABCs are most commonly found in long bones of extremities, thorax, pelvis, and spinal column. Skull base and cranial bone involvement is very rare.[5,10] In this report, the authors present a patient's clinical presentation and treatment with aneurysmal bone cyst involving skull base and extending intracranially.

Treatment options of ABCs include surgical resection, curettage, cryotherapy, sclerotherapy, and radiotherapy.[5,7,17–22]

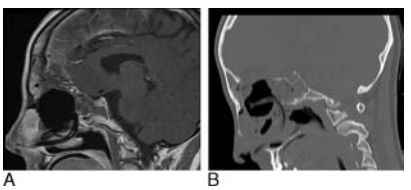

FIGURE 3. Postoperative T1-weighted sagittal MR (A) and sagittal reconstraction paranasal CT (B) show the gross total removal of lesion.

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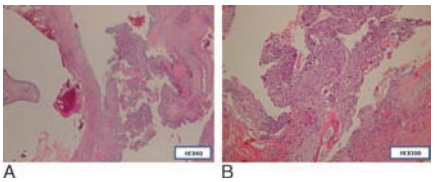

FIGURE 4. A, Photomicrograph showing the blood-containing cystic spaces that contain blood in the lumen. The cyst walls consist of fibrous septum that do not contain cells and osteoid formation in cyst walls (hematoxylin and eosin stain; original magnification ×40). B, Osteoid formation and adjacent cellular region in cyst walls and osteoclast, fibroblast, and hemosiderin-laden macrophages constitude cellular region. Osteoblastic and osteoclastic accumulation are noted arround immature bone trabeculae.

Percutaneous sclerotherapy is a minimally invasive therapy that is combined with surgery and endovascular therapy options.[10,20] In the aspect of neurological impairment and hemorrhage, if the intracranial lesions have high risk with surgery, sclerotherapy can be used alone.[6,10,25] Curettage succeeds in halting progression of disease or even causes regression in cranial ABCs, but there is a high rate of recurrence.[10,26] Total surgical removal is the best available treatment option.[10,27–29] Transcranial and endoscopic endonasal skull base approaches can be used. Difficulty of total resection, risk of recurrence, cosmetic problems, difficulty of reconstruction after resection that depends on commonly used transcranial skull base approach cannot be ignored.[1,5,11]

In recent years, endoscopic endonasal skull base approaches advanced to a very important position owing to technological development and increasing experience. Nowadays, anterior, middle, and posterior skull base lesions can be treated with minimally invasive endoscopic endonasal approach. Advantages of this approach include the following: geting through lesions in a physiological way, direct observation of lesions, allowing for total resection of lesions with domination of the whole skull base that is also the origin of lesions, does not cause cosmetic problems, patients are comfortable postoperatively, and reduction of hospital stay. The most important disadvantange of this approach is postoperative cerebrospinal fluid leak. However, with increasing experience and improved methods of endoscopic approach, skull base reconstruction has ceased to be a serious problem based on the very successful results with endoscopic endonasal approach. Bear in mind that, especially as we presented in this report, there is a need for sufficient experience and certain period of training for extented endoscopic endonasal approaches. The endoscopic endonasal approach that we used in this case is less invasive, more efficient, and safer than transcranial approaches. In addition, this approach has less rate of complication at both the preoperative and postoperative period; therefore, it offers better quality of life at postoperative period.

Consequently, ABC involving skull base is very rare, but these lesions should be kept in mind when making differential diagnosis of skull base pathologies. Recently, the endoscopic endonasal approach, which is a minimally invasive method, is highly effective and safe as a treatment choice for skull base pathologies.

REFERENCES

1. Nadkarnı T, Goel A, Desai K, et al. Massive aneurysmal bone cyst of the anterior cranial fossa floor. Case report. *Neurol Med Chir (Tokyo)* 2001;41:615–619
2. Fu YS, Perzin KH. Non-epithelial tumors of the nasal cavity,paranasal sinuses and nasopharynx:a clinicopathologic study. *Cancer* 1974;33:1289–1305
3. Langdon D, Rapidis AD, Patel MF. Ossifying fibroma—one disease or six? An analysis of 39 fibro-osseous lesios of the jaws. *Br J Oral Surg* 1976;14:1–11
4. Saito K, Fukuta K, Takahashi M, et al. Benign fibroosseous lesions involving the skull base, paranasal sinuses and nasal cavity. *J Neurosurg* 1998;88:1116–1119
5. Salmasi V, Blitz AM, Ishii M, et al. Expanded endonasal endoscopic approach for resection of a large skull base aneurysmal bone cyst in a pediatric patient with extensive cranial fibrous dysplasia. *Childs Nerv Syst* 2011;27:649–656
6. Manjila S, Zender C, Weaver J, et al. Aneurysmal bone cyst within fibrous dysplasia of the anterior skull base: continued intracranial extension after endoscopic resections requiring craniofacial approach with free tissue transfer reconstruction. *Childs Nerv Syst* 2013;29:1183–1192
7. Vergel De Dios AM, Bond JR, Shives TC, et al. Aneurysmal bone cyst. A clinicopathologic study of 238 cases. *Cancer* 1992;69:2921–2931
8. Jaffe HL, Lichtenstein L. Solitary unicameral bone cyst: with emphasis on the Roentgen picture, the pathologic appearance and pathogenesis. *Arch Surg* 1942;44:1004–1025
9. Martinez V, Sissons HA. Aneurysmal bone cyst. A review of 123 cases including primary lesions and those secondary to other bone pathology. *Cancer* 1988;61:2291–2304
10. Bozbuğa M, Turan Süslü H. Aneurysmal bone cyst of the sphenoid bone extending into the ethmoid sinus, nasal cavity and orbita in a child. *Turkish Neurosurgery* 2009;19 No:2:172–176
11. Sheikh BY, Kanaan I, AlWatban J, et al. Aneurysmal bone cyst ınvolving the skull base: report of three cases. *Skull Base Surgery* 19999: Number 2:145–148
12. Terkawi AS, Al-Qahtani KH, Baksh E, et al. Fibrous dysplasia and aneurysmal bone cyst of the skull base presenting with blindness: a report of a rare locally aggressive example. *Head Neck Oncol* 2011;3:15
13. Lui YW, Dasari SB, Young RJ. Sphenoid masses in children: radiologic differential diagnosis with pathologic correlation. *AJNR Am J Neuroradiol* 2011;32:617–626
14. Lichtenstein L. Aneurysmal bone cyst: a pathological entity commonly mistaken for giant cell tumor and occasionally for hemangioma and osteogenic sarcoma. *Cancer* 1950;3:279–289
15. Delorit GJ, Summers GW. Aneurysmal bone cyst of the sphenoid sinus. *Trans Am Acad Ophthalmol Otolaryngol* 1975;80:438–443
16. Luccarelli G, Fornari M, Savoiardo M. Angiography and computerized tomography in the diagnosis of aneurysmal bone cyst of the skull: case report. *J Neurosurg* 1980;53:113–116
17. Biesecker JL, Marcove RC, Huvos AG, et al. Aneurysmal bone cysts. A clinicopathologic study of 66 cases. *Cancer* 1970;26:615–625
18. Boriani S, De Iure F, Campanacci L, et al. Aneurysmal bone cyst of the mobile spine: report on 41 cases. *Spine* 2001;26:27–35
19. Campanacci M, Capanna R, Picci P. Unicameral and aneurysmal bone cysts. *Clin Orthop Relat Res* 1986;204:25–36
20. Dubois J, Chigot V, Grimard G, et al. Sclerotherapy in aneurysmal bone cysts in children: a review of 17 cases. *Pediatr Radiol* 2003;33:365–372
21. Ruiter DJ, van Rijssel TG, van der Velde EA. Aneurysmal bone cysts: a clinicopathological study of 105 cases. *Cancer* 1976;39:2231–2239
22. Tillman BP, Dahlin DC, Lipscomb PR, et al. Aneurysmal bone cyst: an analysis of ninety-five cases. *Mayo Clin Proc* 1968;43:478–495
23. Keuskamp PA, Horoupian DS, Fein JM. Aneurysmal bone cyst of the temporal bone presenting as a spontaneous intracerebral hemorrhage: case report. *Neurosurgery* 1980;7:166–170
24. Constantini FE, Iraci G, Benedetti A, et al. Aneurysmal bone cyst as an Intracranial space-occupying lesion: case report. *J Neurosurgery* 1966;25:205–207
25. Kumar P, Singh S, Phadke RV, et al. Role of radiotherapy in a recurrent aneurysmal bone cyst of the temporal bone:case report. *Neurosurgery* 2006;58:E584
26. Purohit A, Chopra S, Sinha VD, et al. Aneurysmal bone cyst of the temporal bone: case report. *Neurol India* 2002;50:511–513
27. de Minteguiaga C, Portier F, Guichard JP, et al. Aneurysmal bone cyst in sphenoid bone:treatment with minimally invasive surgery. *Ann Otol Rhinol Laryngol* 2001;110:331–334
28. Kimmelman CP, Potsic WP, Schut L. Aneurysmal bone cyst of the sphenoid in a child. *Ann Otol Rhinol Laryngol* 1982;91:339–341
29. Tamimi AF, Nimri C, Huseini M, et al. Aneurysmal bone cyst of the sphenoid bone as an intracranial and orbital space-occupying lesion. *Pediatr Neurosurg* 2005;41:280–282

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Comparative Study Between Partial Inferior Turbinotomy and Microdebrider-Assisted Inferior Turbinoplasty

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Purpose: The purpose of our study was to compare the inferior turbinotomy and the microdebrider-assisted inferior turbinoplasty in patients with hypertrophy of the inferior turbinate.

Material and Methods: We carried out a retrospective review of 205 patients, 96 women and 109 men, with a mean age of 48 years, operated on for hypertrophy of the inferior turbinate between May 2005 and May 2012. Forty-seven patients were excluded from our study because in these patients, nasal obstruction was caused by a specific pathologic condition (allergy, tumors or polyps, recurrent rhinosinusitis, etc). The remaining 158 patients were randomly assigned to undergo partial inferior turbinoplasty through the use of microdebrider (group A, n = 79) or partial inferior turbinotomy (group B, n = 79). Surgical outcome was evaluated according to 4 distinct parameters: nasal endoscopic findings, nasal subjective symptoms, anterior rhinomanometry, and nasal mucociliary transport time. These evaluations were made before surgery and 1 week and 3 months after surgery. The follow-up was a minimum of 24 months and a maximum of 60 months, with a mean follow-up of 42 months.

Results: Turbinate edema and secretions decreased significantly ($P < 0.05$) in groups A and B 3 months after surgery. In group A, crusting was not observed after surgery. In group B, crusting had increased significantly ($P < 0.005$) 1 week after surgery and then decreased significantly at the third month after surgery. Subjective nasal symptoms including nasal obstruction, sneezing, snoring, itchy nose, hyposmia, headache, and dryness were significantly improved in both groups from the third month after surgery ($P < 0.05$). Rhinomanometric measurements demonstrated a significant nasal flow increase at 3 months ($P < 0.05$). The mean nasal mucociliary transport time slightly increased in both groups 1 week after surgery, and then restabilized to preoperative values at the third-month follow-up in both groups (difference not significant).

Conclusions: Microdebrider-assisted inferior turbinoplasty and partial inferior turbinotomy are very effective surgical techniques for solving hypertrophy of the inferior turbinates and therefore related problems of nasal obstruction. Microdebrider-assisted inferior turbinoplasty compared to partial inferior turbinotomy ensures a greater preservation of the nasal mucosa to prevent nasal bleeding.

Key Words: Turbinate hypertrophy, partial inferior turbinotomy, microdebrider, turbinoplasty, turbinectomy, nasal obstruction

Hypertrophy of the turbinate is the second leading cause of nasal obstruction after septal deviation.[1] Passali et al[2] have stated that 20% of the population has a chronic nasal obstruction caused by inferior turbinate hypertrophy. Causes of turbinate hypertrophy can be allergic or pseudoallergic nasal hyperreactivity, nasal decongestant abuse, septal deviation, and nasal trauma.[3]

The effects on nasal respiratory function are clinically important because, although not life endangering, symptoms have a significant impact on the quality of life of patients.

The first-line treatment of this disease is obviously pharmacological. Frequently, standard medical treatment with steroids, topical decongestants, and antihistamines is inadequate; and a surgical reduction of the inferior turbinate must be performed.

Over the years, various surgical techniques have been developed to reduce hypertrophy of the inferior turbinates, including total turbinectomy, partial inferior turbinotomy, turbinoplasty, submucosal resection, laser-assisted turbinoplasty, cryosurgery, electrocautery with monopolar and/or bipolar techniques, vidian neurectomy, radiofrequency volumetric tissue reduction, or the use of different types of lasers and "powered" instrumentation.[4–7]

Although these surgical techniques can provide better results than medical treatment alone, complications may include nasal and postnasal discharge, headache, facial discomfort, empty nose syndrome, dryness, bleeding, postoperative pain, anosmia, foul smell, synechia, or atrophy of the inferior turbinates, as well as nasal hyporeactivity, owing to damage of the nerve receptors complex covering the nasal mucosa.[8–12]

Currently, a technique that is giving us excellent outcomes is the microdebrider-assisted inferior turbinoplasty (MAIT).

The microdebrider has been used since the 1990 s in endoscopic rhinosinusal surgery and has been introduced in turbinate surgery to offer a submucosal approach to preserving the integrity of nasal mucosa[13–15]However, some authors consider submucosal techniques as dangerous for mucosal function because they can cause nasal denervation and mucosal metaplastic alterations.[3,9]In our experience, after an adequate learning curve, we have completely resolved this complication.

Another technique capable of efficiently solving nasal obstruction without mucosal dysfunction and denervation is partial inferior turbinotomy (PIT).[9,16,17]This technique represents an evolution of the total inferior turbinectomy and, although it has lower risks compared to the latter, it gives better results.

This study will compare the results of partial inferior turbinoplasty through debrider, and partial inferior turbinotomy by cold knife, evaluating both postoperative symptoms and patients' satisfaction.

MATERIALS AND METHODS

Two hundred five patients (96 women and 109 men; age range, 15–78; with a mean age of 47 years) were observed at the Maxillofacial Surgery Department of the University Federico II of Naples between May 2005 and May 2012. These were retrospectively analyzed to compare 2 surgical techniques.

All patients had nasal obstruction symptoms caused by the hypertrophy of the inferior turbinates, which had not responded to

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treatment with steroids, topical decongestants, and antihistamines over the previous 3 months.

Fourty-seven patients were not included in this study because they exhibited other diseases beyond a hypertrophy of the inferior turbinates.

The remaining 158 patients were randomly assigned to undergo partial inferior turbinoplasty through the use of microdebrider (n = 79) or partial inferior turbinotomy (n = 79).

Microdebrider-Assisted Inferior Turbinoplasty (MAIT)

Microdebrider-assisted turbinoplasty was performed under general anesthesia. Some authors have reported performing local anesthesia for the same approach. This procedure was performed under 30-degree endoscopic guidance.

An infiltration of the inferior turbinate with 1% xylocaine at 1:20.000 epinephrine was made.

A StortzDrillcutx 40711070 was used for microdebrider-assisted turbinoplasty. After creating an anteroinferior submucosal pocket on the inferior turbinate with a conventional 15 blade, the microdebrider unit was set at 7000-rpm oscillating mode. The inferior turbinate size was reduced with an inferior turbinate 2-mm blade especially from the anterior head, taking great care to stay in the submucosal plane.

Nasal packing was performed using Merocel (Medtronic, Mystic, CY, USA). The patients with no complications were discharged after 24 hours. All patients received postoperative antibiotic therapy (cephalosporin for 5 days and tapered Deltacortene, 5 mg, for 7–9 days).

Partial Inferior Turbinotomy (PIT)

Partial inferior turbinotomy was performed under general anesthesia. Any redundant mucosa of the turbinate was excised with turbinate scissors to remove primarily the hypertrophic tissue of the inferior turbinate, preserving its head. The resection included the mucosa, the submucosal tissue, and, in case of bone hypertrophy, a small resection of the inferior turbinate bone. The extent of the resection depended on the degree of hypertrophy.

Nasal packing was performed using Merocel (Medtronic, Mystic). Patients with no complications were discharged after 24 hours.

Postoperatively, all patients received antibiotic therapy (cephalosporin for 5 days and tapered Deltacortene, 5 mg, for 7–9 days).

Evaluation

We evaluated all patients before the procedures weekly in the first month, and then monthly for 6 months and yearly for a minimum of 24 months and a maximum of 60 months, with a mean follow-up of 42 months.

Each patient was studied preoperatively by performing a computed tomographic scan without contrast media of the nose and paranasal sinuses through an active anterior rhinomanometry examination to evaluate the nasal flow.

Turbinate edema, secretions, and crusts were evaluated in the treatment of these patients using a 5-point scale. (zero, absent; 1, mild; 2, moderate; 3, severe; and 4, very severe).[9]

A standard visual analog scale ranging from zero (no symptoms) to 10 (the most severe symptoms) was used to assess subjective symptoms including nasal obstruction, postnasal discharge, headache, facial discomfort, empty nose syndrome, dryness, bleeding, postoperative pain, anosmia, foul smell, synechia, or atrophy of the inferior turbinates.

Anterior active rhinomanometry was performed according to the International Committee for the Standardization of Rhinomanometry guidelines. The flow was evaluated at a transnasal pressure of 150 Pa.[18] This test was performed with a nasal spray containing xylometazoline before and 10 minutes after decongestion.[19]

The nasal mucociliary transport time was evaluated with the saccharin test. Saccharin transit time was measured after depositing 15 mg of sodium saccharin behind the inferior turbinate. The patients were instructed to sit with their head inclined forward at a 10-degree angle, swallowing every 30 seconds and avoiding blowing their nose. The test was stopped when the saccharin was tasted.[20,21]

Statistical analysis was performed with the SPSS for Windows and the Student t test statistical software package. $P < 0.05$ was considered statistically significant.

RESULTS

All the 158 patients of the 2 groups (A and B) completed the follow-up sessions at 1 week and 3 months after the surgery. No uncontrolled bleeding was observed. Compared to preoperative levels, edema reduction at a week after surgery was not statistically significant, whereas at 3 months after the surgery, it was statistically significant in both groups. The nasal secretions at a week after the surgery remained stable in group A and increased in group B (difference not statistically significant) 3 months after surgery. However, there was a statistically significant reduction in both groups. One week after the surgery, only the group that had undergone the turbinotomy had a formation of crusts (statistically significant compared to the preoperative). These crusts at 3 months after the surgery, however, had healed (Table 1).

A week after the surgery, there was no statistically significant reduction of nasal obstruction in either group. This reduction was, however, statistically significant 3 months after the surgery in both groups.

The typical symptoms related to nasal obstruction (sneezing, itchy nose, snoring, hyposmia, dryness, headache) were significantly reduced from 3 months after the surgery in both surgical groups. All the subjective nasal symptoms were lower in group A than in group B, but these differences were of no statistical significance (Table 2).

Only in group B did we observe a significantly decreased nasal flow measured from the left and right rhinomanometric 7 days after the surgery. At 3 months after the surgery, the rhinomanometric result significantly improved in both groups with no significant difference between group A and group B (Table 3).

The mean nasal mucociliar transport time slightly increased in both groups after 1 week of surgery and then restabilized to preoperative values at 3 months' follow-up in both groups (difference not significant; Table 3)

TABLE 1. Nasal Findings (Mean ± SD)

	Preoperative	1 Week	3 Month	P^+
Turbinate edema				
Group A	2.64 ± 0.50	2.20 ± 0.78	0.65 ± 0.62	<0.0001
Group B	2.70 ± 0.52	1.80 ± 0.62	0.56 ± 0.48	<0.0001
Secretions				
Group A	1.90 ± 0.76	1.90 ± 0.72	0.50 ± 0.50	<0.0001
Group B	1.75 ± 0.72	2.24 ± 0.46	0.60 ± 0.50	<0.0001
Crusts				
Group A	—	—	—	—
Group B	—	2.20 ± 0.54	—	—

Five-point scale (zero, absent; 1, mild; 2, moderate; 3, severe; 4, very severe) was used.

$^+$ Comparing preoperative with 3-month follow-up values.

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TABLE 2. Nasal Subjective Symptom (Mean ± SD)

	Preoperative	1 Week	3 Month	P^+
Nasal obstruction				
Group A	7.92 ± 1.61	7.24 ± 1.68	1.15 ± 1.17	<0.0001
Group B	8.40 ± 1.43	7.46 ± 1.62	1.41 ± 1.05	<0.0001
Sneezing				
Group A	5.72 ± 1.02	5.56 ± 1.09	1.67 ± 1.06	<0.0001
Group B	5.61 ± 1.03	5.42 ± 1.07	1.81 ± 0.96	<0.0001
Snoring				
Group A	6.54 ± 1.06	6.09 ± 1.01	1.18 ± 0.67	<0.0001
Group B	6.50 ± 1.18	6.41 ± 0.92	1.26 ± 0.65	<0.0001
Itchy nose				
Group A	3.70 ± 1.15	3.05 ± 1.16	1.16 ± 0.61	<0.0001
Group B	3.85 ± 1.19	3.32 ± 1.02	1.21 ± 0.49	<0.0001
Hyposmia				
Group A	6.91 ± 1.13	5.42 ± 1.28	1.21 ± 1.12	<0.0001
Group B	6.92 ± 0.74	5.68 ± 0.77	1.42 ± 1.25	<0.0001
Headache				
Group A	3.80 ± 1.34	3.35 ± 1.36	1.45 ± 0.61	<0.0001
Group B	4.20 ± 1.32	3.95 ± 1.21	1.80 ± 0.39	<0.0001
Dryness				
Group A	4.14 ± 1.08	3.70 ± 0.94	1.65 ± 0.61	<0.0001
Group B	4.35 ± 1.06	3.82 ± 0.91	1.96 ± 0.52	<0.0001

Visual analog scale ranging from zero (no symptoms) to 10 (the most severe symptom) was used to determine the degree of the subjective symptoms.
$^+$ Comparing preoperative with 3-month follow-up values.

DISCUSSION

Since the early 1900 s, the inferior turbinates have been subjected to surgical manipulation to solve the problem of nasal obstruction and thereby to improve breathing. Given the important role in the physiology of the nasal turbinate, both for the dynamic ventilator and for the important functions of the covering mucosa,[22] over the years, many surgical techniques have been proposed to reduce hypertrophy of the inferior turbinates, including total turbinectomy, partial inferior turbinotomy, turbinoplasty, submucosal resection, laser-assisted turbinoplasty, cryosurgery, electrocautery with monopolar and/or bipolar techniques, vidian neurectomy, radiofrequency volumetric tissue reduction, or the use of different types of lasers and "powered" instrumentation.[4–7]

Hot et al[23] analyzed all these surgical procedures, concluding that many techniques can damage the function of the mucosa of the turbinate without resolving nasal obstruction. They conclude the review preferring the "infra-tubinal turbinoplasty" technique.

Therefore, the aim of turbinate surgery should be to reduce the volume of the inferior turbinate while retaining the nasal mucosa to comply with the function of heating and humidification of inhaled air of the turbinates.[24]

TABLE 3. Results of NMTT and AAR (Mean ± SD)

	Preoperative	1 Week	3 Month	P^+
NMTT				
Group A	14.50 ± 2.06	14.90 ± 2.21	14.62 ± 2.08	>0.05
Group B	14.15 ± 2.04	14.45 ± 1.56	14.23 ± 2.04	>0.05
AAR, Pa/cm^3				
Group A	1.31 ± 0.32	0.92 ± 0.25	0.20 ± 0.07	<0.05
Group B	1.30 ± 0.31	0.22 ± 0.08	0.21 ± 0.06	<0.05

$^+$ Comparing preoperative with 3-month follow-up values.
NMTT, nasal mucociliary transport time; AAR, anterior active rhinomanometry.

A study of Passali et al[2] described the inferior partial turbinotomy as the technique that solves most or all the problems of nasal obstruction. This technique, however, albeit minimally, impairs the nasal mucosa and requires great skills on the part of the surgeon.

Sapci et al[25] in their2005 study reported that partial inferior turbinotomy compared to other techniques is more effective in preserving mucociliary function. Salzano et al,[9] in comparing the turbinotomy with the "hot procedures" (radiofrequency, high frequency, and eletrocautery), report that partial inferior turbinotomy is the most effective in improving nasal obstruction and related nasal symptoms specially as far as preserving the normal mucosal reactivity is concerned.

The microdebrider, initially used since the 1990 s in endoscopic rhinosinusal surgery, was introduced into turbinate surgery as a submucosa corridor with the advantage of not altering the function of the nasal mucosa.[13–15]

The first authors to use the microdebrider to reduce the hypertrophy of the inferior turbinates were Davis and Nishioka[26] in 1996. Lee and Chen[27] reported that MAIT is a minimally invasive technique, safe and effective for the treatment of hypertrophic rhinitis and hypertrophy of the inferior turbinates. In addition, Gupta et al[28] and Cingi et al[29]showed the effectiveness of this technique for the recovery of respiration and the resolution of nasal obstruction. Neri et al[3] in 2013 confirmed what has been previously described and explained how just by respecting nasal turbinate mucosa during MAIT surgery it is possible to achieve a long-term resolution of nasal obstruction and its symptoms associated, respecting mucociliary function.

Owing to the limited data on the efficacy of these 2 alternative surgical modalities for turbinate surgery, our study compared their effects with regard to nasal obstruction and related symptoms, nasal endoscopic findings, nasal mucociliary transport time, and active anterior rhinomanometry in a patient population affected by hypertrophy of the inferior turbinates.

In our study, both groups showed a nonsignificant decrease of nasal secretions after 1 week of intervention (group A, 0.51 ± 0.49; and group B, 0.53 ± 0.5 vs preoperative values of group A, 2.64 ± 0.51 and group B, 2.7 ± 0.52 of secretions, respectively). This difference becomes significant at 3 months after the surgery. Both groups showed a nonsignificant decrease of turbinate edema 1 week after the surgery: 0.65 ± 0.62 (group A) and 0.56 ± 0.48 (group 2) versus preoperative values of 1.9 ± 0.76 (group A) and 1.75 ± 0.72 (group B) of turbinate edema, respectively. This difference becomes significant at 3 months after the surgery.

Only in group B, however, was there a formation of crusts a week after surgery (2.2 ± 0.54), which had resolved at 3 months after the surgery.

In both groups (MAIT and turbinotomy), there was relief of nasal obstruction and symptoms related to it. The differences in sneezing, snoring, itchy nose, hyposmia, headache, and dryness is always statistically significant in group A and in group B compared with preoperative values. The values at 3 months of the intervention of subjective nasal symptoms of group A were slower than those of group B, but this difference is not statistically significant.

In this study, the normalization of nasal mucociliary clearance 3 months after the surgery is a sign of how both techniques reversibly damage the nasal mucosa and do not cause loss of function. In fact, at 3 months after surgery the NMMT shows us very similar to preoperative values in both groups (14.62 ± 2.08 and 14.23 ± 2.04 vs 14.5 ± 2.06 and 14.15 ± 2.04, respectively).

At active anterior rhinomanometry, the mean total nasal resistance and the total inspiration flow strongly improved 3 months after surgery in both groups (0.2 ± 0.07 and 0.21 ± 0.06 vs 1.31 ± 0.32 and 1.3 ± 0.31, respectively; $P < 0.001$).

At active anterior rhinomanometry, the mean total nasal resistance and the total flow at deep inhalation improved 3 months after

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surgery in both groups. This examination is crucial for assessing nasal obstruction.

The recovery of vasomotor reactivity documented in our sample with this examination is further evidence of how the MAIT and the PIT are 2 optimal techniques compared to hot procedures. Moreover, hot procedures generate damage to the nerves and submucosal fibrosis, which causes hyporeactivity to stimuli altering normal physiology.[3,9,30]

In our experience, neither techniques presented the denervation described by some authors[3,9] because after an adequate learning curve, this problem can be solved respecting inferior turbinate anatomy.

CONCLUSIONS

To our knowledge, this is the first study that compares the MAIT technique to turbinotomy with a very large sample of 158 patients.

Given the results obtained, we can state that these 2 surgical techniques on the inferior turbinate are both very effective and solve the pathologic condition while respecting the nasal mucosa and thus the function of the turbinate. In our opinion, MAIT is more suitable to the individual anatomical variations of each patient, providing fast healing and preserving the humidification and mucociliary transport functions. In addition, for an inexperienced surgeon dealing with this type of surgery, MAIT represents a less risky bleeding procedure because it allows an extensive visual control of the inferior turbinate.

REFERENCES

1. Tasman AJ. Die untere Nasenmuschel. Dysregulation und chirurgischeVerkleinerung. *Laryngorhinootologie* 2002;81:822–833
2. Passali D, Bellussi L, Damiani V, et al. Allergic rhinitis in Italy: epidemiology and definition of most commonly used diagnostic and therapeutic modalities. *Acta Otorhinolaryngol Ital* 2003;23:257–264
3. Neri G, Mastronardi V, Traini T, et al. Respecting nasal mucosa during turbinate surgery: end of the dogma? *Rhinology* 2013;51:368–375
4. Mol MK, Huizing EH. Treatment of inferior turbinate pathology: a review and critical evaluation of the different techniques. *Rhinology* 2000;38:157–166
5. Batra PS, Seiden AM, Smith TL. Surgical management of adult inferior turbinate hypertrophy: a systematic review of the evidence. *Laryngoscope* 2009;119:1819–1827
6. Passali D, Lauriello M, Anselmi M, et al. Treatment of hypertrophy of inferior turbinate: long-term results in 382 patients randomly assigned to therapy. *Ann Otol Rhinol Laryngol* 1999;108:569–575
7. Jackson LE, Koch RJ. Controversies in the management of inferior turbinate hypertrophy: a comprehensive review. *Plast Reconstr Surg* 1999;103:300–312
8. Goode RL. Diagnosis and treatment of turbinate dysfunction: a self-instructional package Washington DC: American Academy of Otolaryngology; 1977:36-52
9. Salzano FA, Mora R, Dellepiane M, et al. Radiofrequency, high-frequency, and electrocautery treatments vs partial inferior turbinotomy: microscopic and macroscopic effects on nasal mucosa. *Arch Otolaryngol Head Neck Surg* 2009;135:752–758
10. Mathai J. Inferior turbinectomy for nasal obstruction review of 75 cases. *Indian J Otolaryngol Head Neck Surg* 2004;56:23–26
11. Salzano FA, Mora R, Penco S, et al. Nasal tactile sensitivity in allergic rhinitis. *Acta Otolaryngol* 2011;131:640–644
12. Salzano FA, Guastini L, Mora R, et al. Nasaltactilesensitivity in elderly. *Acta Otolaryngol* 2010;130:1389–1393
13. Yanez C. New technique for turbinate reduction in chronic hypertrophic rhinitis, intraturbinate stroma removal using the microdebrider. *Operat Tech Otolaryngol Head Neck Surg* 1998;9:135–137
14. Van delden MR, Cook PR, Davis WE. Endoscopic partial inferior turbinoplasty. *Otolaryngol Head Neck Surg* 1999;121:406–409
15. Ikeda K, Oshima T, Suzuki M, et al. Functional inferior turbinosurgery for the treatment of resistance chronic rhinitis. *Acta Otolaryngol* 2006;126:739–745
16. Mori S. Submucous turbinectomy decreases not only nasal stiffness but also sneezing and rhinorrea in patients with perennial allergic rhinitis. *Clin Exp Allergy* 1999;29:1542–1548
17. Chevretton EB, Hopkins C, Black IM. Degloving of the inferior turbinates: pilot study to assess the effectiveness of a new technique in turbinate reduction. *J Laryngol Otol* 2003;117:866–870
18. Salzano FA, Manola M, Tricarico D, et al. Mucociliary clearance after aerobic exertion in athlete. *Acta Otorhinolaryngol Ital* 2000;20:171–176
19. Jones AS, Wight RG, Kabil Y, et al. Predicting the outcome of submucosal diathermy to the inferior turbinate. *Clin Otolaryngol* 1989;14:41–44
20. Clement PA. Committee report on standardization of rhinomanometry. *Rhinology* 1984;22:151–155
21. Cavaliere M, Mottola G, Iemma M. Comparison of the effectiveness and safety of radiofrequency turbinoplasty and traditional surgical technique in treatment of inferior turbinate hypertrophy. *Otolaryngol Head and Neck Surg* 2005;133:972–978
22. Passali D, ed. *Around the nose*. Firenze, Italy, Conti Tipocolor 1988: 1–244.
23. Hol MK, Huizing EH. Treatment of inferior turbinate pathology: a review and critical evaluation of the different techniques. *Rhinology* 2000;38:157–166
24. Chen YL, Liu CM, Huang HM. Comparison of microdebrider-assisted inferior turbinoplasty and submucosal resection for children with hypertrophic inferior turbinates. *Int J Pediatr Otorhinolaryngol* 2007;71:921–927
25. Sapci T, Sahin B, Karavus A, et al. Comparison of the effects of radiofrequency tissue ablation, CO_2 laser ablation, and partial turbinectomy applications on nasal mucociliary functions. *Laryngoscope* 2003;113:514–519
26. Davis WE, Nishioka GJ. Endoscopic partial inferior turbinectomy using a power microcutting instrument. *Ear Nose Throat J* 1996;75:49–50
27. Lee CF, Chen TA. Power microdebrider-assisted modification of endoscopic inferior turbinoplasty: a preliminary report. *Chang Gung Med J* 2004;27:359–365
28. Gupta A, Mercurio E, Bielamowicz S. Endoscopic inferior turbinate reduction: an outcomes analysis. *Laryngoscope* 2001;111:1957–1959
29. Cingi C, Ure B, Cakli H, et al. Microdebrider-assisted versus radiofrequency-assisted inferior turbinoplasty: a prospective study with objective and subjective outcome measures. *Acta Otorynolaryngol Ital* 2010;30:138–143
30. Leong SC, Farmer SEJ, Eccles R. Coblation for inferior turbinate reduction: a long-term follow-up with subjective and objective assessment. *Rhinology* 2010;48–1:108–112

Congenital Laryngomucocele: A Rare Cause of Airway Obstruction in a Newborn

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Abstract: Congenital laryngoceles are defined as cystic dilatation of laryngeal saccules and are an extremely rare cause of newborn respiratory distress. A laryngomucocele occurs when the neck of the laryngocele gets obstructed and fills with the mucoid secretions of the saccule. It may cause stridor, respiratory distress, and severe airway obstruction in the narrow airway of a newborn and necessitates urgent surgical intervention. There is only 1 case of congenital laryngomucocele reported in an autopsy examination in the English literature, and here we report the first living congenital laryngomucocele case and discuss the clinical approach.

Key Words: Laryngocele, laryngomucocele, airway, obstruction, newborn

Laryngocele is an air-filled cystic dilatation of laryngeal saccule that extends upward within the false vocal fold and communicates with the laryngeal lumen. When the cavity is filled with mucus or pus, it is called laryngomucocele and laryngopyocele, respectively.[1,2] Its etiology is still unclear, but it is caused by congenital and acquired causes. The congenital large saccule, weakness of periventricular tissues, and acquired factors such as coughing, straining, or blowing wind instruments that lead to an increase in the intralaryngeal pressure are blamed as predisposing factors. According to its relationship to thyrohyoid membrane, it is classified as ''internal'' and ''combined or mixed'' types. Although it is a benign lesion, it has the potential to enlarge and may cause serious airway obstruction and need urgent intervention. Laryngocele may be seen at any age, but it is most commonly presented at the fifth and sixth decades and is extremely rare in newborn periods. According to our knowledge, only 1 case of congenital laryngomucocele in a newborn has been reported with a postmortem examination in the literature.[3,4]

Here we report a newborn treated with the diagnosis of congenital combined laryngomucocele and discuss the clinical aspects, differential diagnosis, and treatment of this rare entity.

CASE

A female baby was born from a 22-year-old mother with a gestational age of 42 weeks with no prenatal diagnosis. The patient suffered from severe respiratory distress and stridor soon after birth and intubated in another hospital. The patient was referred to our department for unsuccessful several weaning attempts. On her physical examination, there were no dysmorphic features of nose, jaw, and tongue or facial deformities. A firm smooth swelling 2 cm in diameter on the right side of the neck was noted (Fig. 1). Neck ultrasonography showed a lobulated cystic lesion with a diameter of 20 mm close to the right thyroid lobe. Computed tomography of the neck revealed thin-walled fluid-filled ovoid cystic masses of which one had the dimensions of 15×15 mm starting from the right aryepiglottic fold and widening in the caudal part. The second cystic component was 13×10 mm in the anterior part of the first lesion. These cystic lesions were narrowing the laryngeal air column at the paraglottic level (Fig. 2). According to these findings, this lesion was regarded as a combined laryngomucocele. Direct laryngoscopy was performed under general anesthesia and a swelling filling the right aryepiglottic fold and false vocal cord was noticed. The external component of the lesion was totally excised by lateral cervical approach, and endolaryngeal marsupialization and aspiration of mucus was performed to the internal part. Morphologically, the cysts were lined by stratified squamous cell layer and were surrounded by fibrous stroma consistent with laryngocele. Two months later, she was admitted to the emergency department with respiratory distress and stridor again and endolaryngeal cyst excision to the internal part was performed. Follow-up was uneventful for a 1-year period.

FIGURE 1. Preoperative view of combined laryngomucocele.

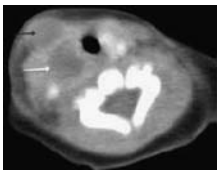

FIGURE 2. CT scan of the neck shows 2 attached cystic lesions (internal [15×15 mm] and external [15×15 mm] part of combined laryngomucocele).

DISCUSSION

A laryngomucocele requires a preexisting laryngocele and occurs when the neck of the laryngocele becomes obstructed and accumulates with the mucoid secretions of the epithelial lining of the laryngocele. Laryngoceles are uncommon among adults, usually appearing at the fifth and sixth decades of life with a 5-to-1 male predominance and extremely rare in newborns. According to our knowledge, only one congenital laryngomucocele case in a postmortem study in a newborn has been reported in the literature.[4]

Although the physiopathology of laryngoceles has not been fully understood and different theories have been proposed, congenital and acquired factors are accepted as predisposing factors. Embryologically, ventricle of the larynx and the saccule develops at the end of the second intrauterine month as a secondary outpouching from the laryngeal lumen.[5] Congenital large saccule, weakness of the periventricular connective tissues, and the thyroepiglottic and aryepiglottic muscles are accepted as predisposing factors in congenital laryngocele.[6] Factors that cause an increase in laryngeal pressure such as chronic coughing, straining, or playing a wind instrument or mechanical obstruction of the ventricle caused by intraventricular pressure such as laryngeal carcinoma, postoperative fibrosis, or amyloidosis promote dilatation of the saccule and predispose the occurrence of the laryngocele.

Laryngoceles are classified as internal and combined type according to their relationship with the thyrohyoid membrane. The former classification of internal, external, and combined or mixed types is being abandoned because purely external laryngoceles cannot exist. Internal laryngoceles are confined within the false vocal fold, laterally limited by the thyrohyoid cartilage and medially limited by the laryngeal wall. Combined laryngoceles extend upward, and when they get enlarged, their sac protrudes through the thyrohyoid membrane and present as an anterior neck mass.[7,8] The clinical features of this rare pathology depend on the size and type of laryngocele and are highly variable. Although they are usually asymptomatic and incidentally found in adults, hoarseness, sore throat, dysphonia, dysphagia, neck swelling, and stridor or respiratory difficulties may be seen as presenting symptoms. They may cause severe respiratory distress and stridor and become a life-threatening event due to mechanical obstruction of the narrower and smaller airway of a neonate. Airway obstruction cannot always

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be diagnosed or predicted prenatally. The main symptom after birth is stridor. In our case, the patient had an anterior neck mass representing the external component and respiratory distress with expiratory stridor representing the internal component causing airway obstruction. Biphasic or expiratory stridor represents obstructions under the level of glottis. Although laryngomalacia is the most common cause of infant stridor and respiratory distress vocal cord palsy, subglottic stenosis or subglottic hemangioma and laryngeal cysts should be considered in the differential diagnosis.[9,10] Laryngeal cysts are also fluid-filled dilatations of the laryngeal ventricle but do not communicate with the laryngeal lumen. Cyanotic spells, changes in the voice, onset of symptoms after birth, and changes in the symptoms by feeding, activity, or position should be noted for accurate diagnosis. Clinical symptoms may suddenly worsen if the mucus in the laryngomucocele get infected and laryngopyocele occurs. After establishing a safe airway, a detailed assessment for the diagnosis and appropriate management is mandatory.[11,12]

The diagnosis of a laryngomucocele mainly depends on physical examination findings and laryngeal examination by laryngoscopy. Inspiratory stridor and neck swelling are useful physical examination findings, but further imaging modalities such as ultrasonography, computed tomography (CT), or magnetic resonance imaging (MRI) are needed to assess the origin and connection of the saccule and exclude pathologies in the differential diagnosis. CT or MRI is also helpful for determining the cysts if filled with air or mucus and showing a combined laryngocele as well if only one component is clinically suspected.[13]

Although various treatment modalities have been utilized, there is no consensus regarding the management of laryngoceles. Total cyst excision was traditionally recommended for both internal and combined types using external approach, but microlaryngoscopy (endolaryngeal), robotic surgery, and carbon dioxide laser therapy gained popularity in the last 2 decades. In our patient, we performed an external lateral neck approach without any resection to the thyroid cartilage for the external part, and endolaryngeal marsupialization and aspiration of mucus was performed to the internal part. Due to the recurrence of internal part 2 months after the operation, we performed an endolaryngeal cyst excision to the internal part.[8,14]

As a conclusion, laryngomucocele may cause severe respiratory distress and may be a life-threatening condition in neonates. After establishing a safe airway, a detailed assessment for the diagnosis and appropriate management is mandatory.

REFERENCES

1. Holinger LD, Barnes DR, Smid LJ, et al. Laryngocele and saccular cysts. *Ann Otol Rhinol Laryngol* 1978;87:675–685
2. Weissler MC, Fried MP, Kelly JH. Laryngopyocele as a cause of airway obstruction. *Laryngoscope* 1985;95:1348–1351
3. Özcan C, Vayisoglu Y, Güner N, et al. External laryngopyocele: a rare cause of upper airway obstruction. *J Craniofac Surg* 2010;21:2022–2024
4. Cunha MS, Janeiro P, Fernandes R, et al. Congenital laryngomucocoele: a rare cause for CHAOS. *BMJ Case Rep* 2009;2009:1–3
5. Maharaj D, Fernandes CM, Pinto AP. Laryngopyocele (a report of two cases). *J Laryngol Otol* 1987;101:838–842
6. Chu L, Gussack GS, Orr JB, et al. Neonatal laryngoceles: a cause for airway obstruction. *Arch Otolaryngol Head Neck Surg* 1994;120:454–458. doi: 10.1001/archotol.1994.01880280082016
7. Thomé R, Thomé DC, De La Cortina RA. Lateral thyrotomy approach on the paraglottic space for laryngocele resection. *Laryngoscope* 2000;110:447–450
8. Karol Zelenik, Lucia Stanikova, Katarina Smatanova, et al. Treatment of laryngoceles: what is the progress over the last two decades? *BioMed Res Int* 2014;2014:1–6
9. Lyons M, Vlastarakos PV, Nikolopoulos TP. Congenital and acquired developmental problems of the upper airway in newborns and infants. *Early Hum Dev* 2012;88:951–955
10. Holinger LD. Etiology of stridor in the neonate, infant and child. *Ann Otol Rhinol Laryngol* 1980;89:397–400
11. Vasileiadis I, Kapetanakis S, Petousis A, et al. Internal laryngopyocele as a cause of acute airway obstruction: an extremely rare case and review of the literature. *Acta Otorhinolaryngol Ital* 2012;32:58–62
12. Abramson AL, Zielinski B. Congenital laryngeal saccular cyst of the newborn. *Laryngoscope* 1984;94:1580–1582
13. Nazaroğlu H, Ozateş M, Uyar A, et al. Laryngopyocele: signs on computed tomography. *Eur J Radiol* 2000;33:63–65
14. Ettema SL, Carothers DG, Hoffman HT. Laryngocele resection by combined external and endoscopic laser approach. *Ann Otol Rhinol Laryngol* 2003;112:361–364

Interdisciplinary Treatment for an Adult Patient With Anterior Open Bite, Severe Periodontitis, and Intellectual Disability

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Abstract: This case report describes the beneficial effects of the interdisciplinary treatment of an adult patient with severe skeletal periodontal problems. A 30-year-old female patient presented with anterior open bite, gummy smile, and facial asymmetry. The patient had chronic generalized severe periodontitis with pathologic maxillary anterior teeth migration and mild intellectual disability. Treatment included 6 months of periodontal treatment, followed by presurgical orthodontic treatment, a Le Fort I osteotomy with anterior segmental osteotomy, a bilateral sagittal split ramus osteotomy, and postsurgical orthodontic treatment. After treatment completion, the patient exhibited functional and aesthetic improvements. Her periodontal condition improved and was maintained after the treatment. Here, we demonstrate a successful treatment outcome in a complicated case following a systematic interdisciplinary approach performed with the correct diagnosis and treatment planning.

Key Words: Anterior open bite, chronic severe periodontitis, intellectual disability, Le Fort I osteotomy with anterior segmental osteotomy, bilateral sagittal split ramus osteotomy

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With increasing numbers of adult orthodontic patients, orthodontic treatment of patients with periodontal disease is becoming more common. Common characteristics of dentition with periodontal disease include proclination of maxillary anterior teeth, diastema, irregular interdental spacing, rotation, extrusion, tipping, and drifting.[1] The main factors known to influence tooth position are the periodontal tissues, occlusal factors, soft tissue pressures, and oral habits.[2] The coexistence of periodontal disease, skeletal problems, and poor habits can rapidly worsen a patient's condition. Appropriate intervention can lead to a better quality of life, including functional and aesthetic improvements.

Intellectual disability is a disability characterized by significant limitations in both intellectual functioning and in adaptive behavior, which covers many everyday social and practical skills. This disability originates before the age of 18.[3] People with intellectual disabilities can and do learn new skills, but they learn them more slowly. It can be categorized as mild, moderate, severe, or profound. Many of the characteristics of mild intellectual disability correspond to those of learning disabilities. The intellectual development will be slow; however, mild intellectual disability patients have the potential to learn within the regular practical skills.[3]

The present case report describes the favorable outcome of an interdisciplinary treatment approach involving orthodontics, orthognathic surgery, periodontics, and prosthodontics in a patient with dentofacial disharmony, severe periodontitis, and intellectual disability.

DIAGNOSIS AND ETIOLOGY

A 30-year-old woman visited the orthodontic department at Yonsei University Dental Hospital in Seoul, Korea. Her chief complaint was the maxillary anterior teeth protrusion and facial asymmetry. On the pretreatment questionnaire, she reported a strong desire to improve her facial appearance. She had mild intellectual disabilities and was accompanied by her mother. She and her mother reported that the teeth seemed to be protruding further forward over time. The patient had nocturnal mouth breathing habit.

Pretreatment facial photographs showed a mandibular deviation toward the left, a small nasolabial angle, and a convex profile. She had mentalis hyperactivity at lip seal and her lips were incompetent and the incisor-stomion distance was 15.0 mm at rest. She showed excessive display of gingivae when smiling (Fig. 1).

Intraorally, the patient had a Class II molar relationship bilaterally, particularly more compromised on the left side. Overjet was 16.0 mm and overbite was −2.0 mm. The mandibular dental midline had 3.0 mm deviation toward the left. Clinical features, including maxillary anterior teeth flaring and the spacing around the canines,

FIGURE 1. Facial and intraoral photographs taken before periodontal treatment.

FIGURE 2. Pretreatment cephalometric and panoramic radiographs.

indicated the presence of pathological migration. She also exhibited dental calculus deposition, especially on the maxillary anterior teeth, and inflammation that led to gingival swelling with spontaneous gingival bleeding. The maxillary arch form was rather constricted relative to the mandibular arch form. The mandibular left first and second premolars had been restored by splinted crowns. In the maxilla and mandible, a total of 8 moderate dental caries were observed (Fig. 1).

Lateral cephalometric analysis showed a skeletal Class II relationship: SNA, 81.7 degrees; SNB, 66.8 degrees; ANB angle, 14.9 degrees; Wits, 17.5 mm; and APDI, 62.2 degrees. There was a steep mandibular plane angle (61.0 degrees) and a large gonial angle (137.1 degrees). The maxillary incisors were inclined labially at an angle of 118.8 degrees towards the SN plane. The upper and lower lips protruded with respect to the aesthetic E-line. A posteroanterior cephalometric analysis indicated the chin was deviated by 4.0 mm toward the left (Fig. 2 and Table 1).

Panoramic and periapical radiographs showed generalized horizontal bone loss in both arches and vertical bone defects between the mandibular left second premolar and first molar. Examination showed the presence of a major gingival inflammatory process, which after closer study was diagnosed as chronic generalized severe periodontitis, as it was found that there were clinical attachment loss (CAL) up to 7.0 mm deep around the mandibular left second premolar and first molar. There were also CAL of up to 5.0 mm at the maxillary premolars, canines, lateral incisors, central incisors, and left molars. CAL of up to 4.0 mm was observed at the mandibular right molars and premolars, lateral incisors, and central incisors (Figs. 2, 3).

TABLE 1. Lateral Cephalometric Measurements

Measurements	Norm	Pretreatment	Posttreatment
Skeletal			
SNA (degrees)	81.6 ± 3.1	81.7	79.5
SNB (degrees)	79.1 ± 3	66.8	68.3
ANB (degrees)	2.4 ± 1.8	14.9	11.2
Wits (mm)	−2.8 ± 2.5	17.5	4.1
APDI (degrees)	84 ± 4	62.2	71.1
ODI (degrees)	71 ± 5	70.8	69.2
Go-Me (mm)	76 ± 4	74.4	81.4
Gonial angle (degrees)	118.6 ± 5.8	128.2	129.8
SN to GoGn (degrees)	34 ± 5	61	56.7
Ramal height (mm)	51.6 ± 4.2	39	37.8
Dental factors			
U1-SN (degrees)	106 ± 5	118.8	94.4
L1-GoGn (degrees)	94 ± 5	88	90.2
Soft tissues			
Upper lip E-line (mm)	−1 ± 2	9.2	1.4
Lower Lip E-line (mm)	1 ± 2	4.8	2.7

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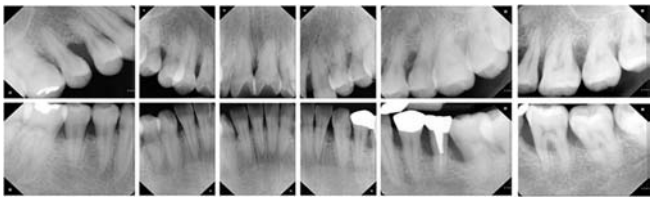

FIGURE 3. Pretreatment intraoral radiographs.

TREATMENT OBJECTIVES

Because of asymmetric long face with skeletal II profile and pathologic migration of the maxillary anterior teeth exacerbated gingival inflammation, the patient expressed concern about her maxillary anterior teeth protrusion and facial asymmetry. The following treatment objectives were planned: (1) to correct the pathologic periodontal condition and facilitate good oral hygiene; (2) to correct facial asymmetry and coordinate the facial, maxillary, and mandibular dental midlines; (3) to correct the skeletal Class II anteroposterior jaw relationship; (4) to correct the excessive display of gingivae; and (5) to relieve the proclined incisors and achieve an ideal overjet and overbite relationship.

TREATMENT ALTERNATIVES

Based on the treatment objectives, the following treatment options were considered: (1) camouflage orthodontic treatment including retractions of anterior teeth by extraction of upper first premolars and intrusions of upper dentitions with temporary skeletal anchorage devices, (2) distraction osteogenesis to increase the ramus length, and (3) two-jaw orthognathic surgery. All options included periodontal treatment. As this patient had severe skeletal and dental problems beyond the limitations of camouflage treatment, it would have been difficult to obtain satisfactory improvement without surgery. Furthermore, the patient had reduced periodontal support and intellectual disability, and thus intrusions were an inappropriate treatment if she could not maintain good oral hygiene during orthodontic treatment. Distraction osteogenesis could induce a relatively physiological response through increasing the ramus length; however, this procedure is invasive and technically difficult, and can result in imprecise movement, patient discomfort, and long treatment time. Moreover, this patient required corrections in the maxilla, which would be limited by treatment with only distraction osteogenesis. Due to all of these considerations, we ultimately planned Le Fort I surgery with anterior segmental osteotomy and superior repositioning of maxilla, bilateral sagittal split ramus osteotomy for mandibular advancement, and advancement genioplasty.

TREATMENT PROGRESS

Before starting orthodontic treatment, the patient was referred to a periodontist, where she received the initial phase of the periodontal treatment including scaling, root planning, and instructions in oral hygiene to resolve gingival bleeding and inflammation. This was followed by the corrective phase of the periodontal treatment. This phase consisted of modified Widman flap from the maxillary right canine to the left canine and another modified Widman flap from the mandibular left first premolar to the second molar. Over the next 5 months, while waiting for adequate tissue healing to occur, the patient received periodic supportive periodontal treatment at 3-month intervals and a repeated oral hygiene program including brushing, rinsing, and professional cleaning at regular follow-up visits (Fig. 4).

Preoperative orthodontic treatment included segmental leveling and aligning of upper teeth and arch coordination. For anterior segmental osteotomy, the roots around the osteotomy lines were arranged divergently to minimize root damage during surgery. The patient's poor oral hygiene complicated the progression of orthodontic treatment. To reduce plaque and gingival inflammation, the patient followed a repeated oral hygiene program at every visit. After 13 months of preoperative orthodontic treatment, the surgical arch wire was located (Fig. 5). The orthognathic surgery involved a segmental Le Fort I procedure. After extraction of the maxillary first premolars, the maxilla was segmented between the maxillary canines and the second premolars to facilitate differential movement between the anterior and posterior maxilla. The surgery was performed with 8.5 mm of anterior impaction and 5 mm of posterior impaction. The autorotation of the mandible serves to correct the anterior open bite and decrease lower facial height. Bilateral sagittal split ramus osteotomy was performed to advance the mandible and rotate it to a Class I canine position with ideal overbite and overjet. In addition, genioplasty was performed to advance the chin for an additional 8 mm. After surgery, the remaining space on the upper premolar regions was closed. After 19 months of surgical orthodontic treatment, the fixed orthodontic appliances were removed. Fixed lingual retainers and removable circumferential retainers were used in both arches for retention. During the retention period, the patient's periodontal status was reevaluated. The patient was referred to a periodontist for sustained periodontal management and to a prosthodontist for caries treatment and the final prosthesis of the lower left premolars.

FIGURE 4. Facial and intraoral photographs after periodontal treatment.

TREATMENT RESULTS

After the periodontal treatment, the patient presented improved oral condition and reduced spacing (Fig. 4). Post-treatment photographs

FIGURE 5. Facial and intraoral photographs with surgical arch wire.

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FIGURE 6. Facial and intraoral photographs at 1 year after debond.

(at 1 year after debond) showed an improved convex profile and decreased lip protrusion. The overjet was reduced and there were marked improvements in lip incompetency, gummy smile, and occlusion (Fig. 6). Panoramic and periapical radiographs showed that the alveolar bone level was not worsened (Figs. 7, 8). Superimposition of the lateral cephalometric images showed superior repositioning of the maxilla. The mandible was advanced by autorotation and bilateral sagittal split ramus osteotomy. The chin was advanced by genioplasty with reduction of anterior facial height (Fig. 9).

DISCUSSION

Several factors contributed to the patient's pretreatment appearance. First, she had nocturnal mouth breathing habit (respiratory disorder). Nasal obstruction and habitual mouth breathing is suspected in the etiology of various malocclusion such as anterior open bite and posterior cross bite. Functional changes after the mouth was established as the airway were thought to induce alterations in dentofacial growth in human,[4] though the relationship between mouth breathing and altered dentofacial growth is controversial.[5] Second, the large overjet resulted in the lower lip being located behind the upper anterior teeth. The overjet was continuously worsened by the pressure of the lower lip on the backside of the upper anterior teeth and the weakened upper lip pressure. Third, due to poor oral hygiene, the patient suffered from chronic severe periodontitis, and pathologic tooth migration occurred due to weakened periodontal support. Her lip incompetency was aggravated by gingival inflammation. Fourth, the patient had mild intellectual disability. A previous study reported that people with intellectual disability are more likely to have poor oral hygiene and periodontal disease and possibly more likely to have untreated caries than people without intellectual disability.[6]

FIGURE 7. Cephalometric and panoramic radiographs at 1 year after debond.

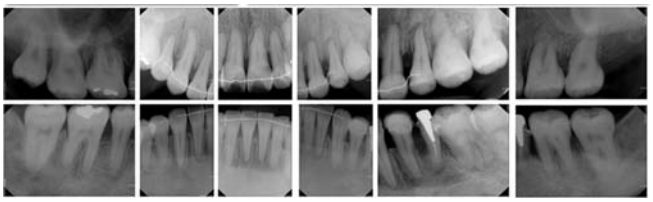

FIGURE 8. Intraoral radiographs at 1 year after debond.

FIGURE 9. Superimposition of cephalometric tracings taken before treatment (blue line) and at 1 year after debond (red line).

Pathologic tooth migration is defined as a change in tooth position that occurs due to disruption of the forces that maintain teeth in a normal relationship. In this case, after periodontal treatment, spontaneous correction of the maxillary anterior teeth was observed. Wound contraction or shrinkage during wound healing might play a role in leading to spontaneous repositioning of teeth.[7,8] However, it was not convincing that the maxillary teeth had accomplished "the newly established equilibrium forces" because several factors contributed to the patient's pretreatment condition. To solve this problem, interdisciplinary treatment with surgico-orthodontic treatment was required.

The orthodontic treatment of patients with periodontal disease requires several considerations. Before the start of orthodontic treatment, any pathological problems should be removed, including resolution of periodontal problems.[9] In a state of inflammation, orthodontic tooth movements are not completely controlled, which can result in rapid irreversible destruction of support tissue. It is suggested that orthodontic treatment be started 2 to 6 months after periodontal treatment to allow time for periodontal tissue remodeling, restoration of health, and evaluation of patient's compliance.[10] Before treatment, the patient should be capable of adequately maintaining oral hygiene, and active orthodontic treatment should be delayed until satisfactory plaque control is achieved to prevent alveolar bone loss during orthodontic treatment. In periodontally compromised dentitions, the loss of alveolar bone results in apical movement of the center of resistance of the involved teeth.[11] In such cases, procedures should be performed with light force, longer interval of force activation, greater moment, and a short treatment period.[12] Furthermore, a longer retention period and long-term maintenance are needed after orthodontic treatment.[10] In this case, skeletal problems were resolved with the help of two-jaw surgery. Attempting camouflage treatment rather than surgery would have likely resulted in unsatisfactory outcomes, longer treatment time, and relatively worse periodontal status. Anterior segmental osteotomy can be useful for achieving a large amount of anterior teeth retraction without anchor loss, as well as rapid changes of facial profile, with shortened treatment duration.[13] This procedure can also reduce the side effects associated with large amounts of tooth movement.

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After the systemic interdisciplinary treatment, multi-dimensional problems were solved. However, the patient's intellectual disability contributed to creating the original state of poor oral hygiene. Unless her intellectual disability is not resolved, her oral hygiene maintenance would be difficult. Therefore, to maintain good oral hygiene and healthy periodontal supports, repeated oral hygiene program including brushing, rinsing, and professional cleaning by a caregiver will be sustained at regular follow-up visit because the best way to improve oral health for people with intellectual disability is effective prevention.[6]

REFERENCES

1. Gkantidis N, Christou P, Topouzelis N. The orthodontic-periodontic interrelationship in integrated treatment challenges: a systematic review. *J Oral Rehabil* 2010;37:377–390
2. Brunsvold MA. Pathologic tooth migration. *J Periodontol* 2005;76:859–866
3. Robert S, Schalock B, Wil HEB, et al. Intellectual Disability: Definition, Classification, and Systems of Supports. *Washington, DC: American Association on Intellectual and Developmental Disabilities* 2010
4. Fujimoto S, Yamaguchi K, Gunjigake K. Clinical estimation of mouth breathing. *Am J Orthod Dentofacial Orthop* 2009;136:630e631–637; discussion 630–631.
5. Krakauer LH, Guilherme A. Relationship between mouth breathing and postural alterations of children: a descriptive analysis. *Int J Orofacial Myology* 2000;26:13–23
6. Anders PL, Davis EL. Oral health of patients with intellectual disabilities: a systematic review. *Spec Care Dentist* 2010;30:110–117
7. Sato S, Ujiie H, Ito K. Spontaneous correction of pathologic tooth migration and reduced infrabony pockets following nonsurgical periodontal therapy: a case report. *Int J Periodontics Restorative Dent* 2004;24:456–461
8. Kumar V, Subbappa A, Thomas CM. Reactive repositioning of pathologically migrated teeth following periodontal therapy. *Quintessence Int* 2009;40:355–358
9. Sanders NL. Evidence-based care in orthodontics and periodontics: a review of the literature. *J Am Dent Assoc* 1999;130:521–527
10. Zachrisson BU. Clinical implications of recent orthodontic-periodontic research findings. *Semin Orthod* 1996;2:4–12
11. Ong MM, Wang HL. Periodontic and orthodontic treatment in adults. *Am J Orthod Dentofacial Orthop* 2002;122:420–428
12. Geramy A. Alveolar bone resorption and the center of resistance modification (3-D analysis by means of the finite element method). *Am J Orthod Dentofacial Orthop* 2000;117:399–405
13. Park JU, Hwang YS. Evaluation of the soft and hard tissue changes after anterior segmental osteotomy on the maxilla and mandible. *J Oral Maxillofac Surg* 2008;66:98–103

Endoscopic Modified Medial Maxillectomy for Treatment of Inverted Papilloma Originating From the Maxillary Sinus

Selim S. Erbek, MD, Alper Koycu, MD, and Fuat Buyuklu, MD

Abstract: Endoscopic approaches have become an alternative to external approaches in the treatment of sinonasal inverted papillomas (IPs) in recent years. The aim of this study was to analyze the outcomes of endoscopic modified medial maxillectomy preserving the nasolacrimal duct and the inferior turbinate in selected IP cases. Medical charts of patients diagnosed with IP originating from the maxillary sinus between July 2008 and August 2013 were reviewed. Eight patients who had undergone endoscopic modified medial maxillectomy were included in the study. Attachment of IP was located on the medial wall of the maxillary sinus in all cases. The nasolacrimal duct was preserved in all of the patients. The inferior turbinate was completely preserved in 5 patients, and the anterior part of the inferior turbinate was preserved in 3 patients. The mean follow-up period of the patients was 30.8 months (12–60 mo). None of the patients had recurrence or major complications. The postoperative complaints were minor hemorrhagic discharge and crusting for the first few weeks. Endoscopic modified medial maxillectomy preserving the nasolacrimal duct and the inferior turbinate provides good surgical and functional outcomes in selected IP cases.

Key Words: Inverted papilloma, endoscopic modified medial maxillectomy

Inverted papilloma (IP) of the paranasal sinuses is a histologically benign but locally destructive tumor.[1] It usually originates from the lateral wall of the nasal cavity and the maxillary sinus, followed by the ethmoid cells, the sphenoid sinus, and the frontal sinuses.[2] Incidence of IP is 0.5 to 1.5 per 100,000 cases a year, with a male predominance.[3] Histologic appearance is characterized by an invading epithelium into the stroma with intact basement membrane that separates the epithelial component from the underlying connective tissue stroma.[2] This tumor is associated with high recurrence and low malignant transformation rates.

Patients with IP have traditionally been operated on with an external approach in the last century. Recently, endoscopic approaches have become an alternative in the treatment of sinonasal IP. Furthermore, studies showed that endoscopic techniques have lower recurrence rates than that of open approaches.[4] Endoscopic medial maxillectomy has been used for the treatment of IP originating from the maxillary sinus. However, the procedure has numerous complications, such as epiphora, incrustation, and inability to feel the nasal airflow, due to resection of the nasolacrimal duct and the inferior turbinate. In the last years, a few new techniques, preserving the nasolacrimal duct and the inferior turbinate to reduce the undesired effects of medial maxillectomy, have been defined and termed as *endoscopic modified medial maxillectomy* (*EMMM*).[5–8] In this report, we describe a new method that can be termed as *EMMM* preserving the nasolacrimal duct and the inferior turbinate.

MATERIALS AND METHODS

Subjects

Baskent University Institutional Review Board approved this study. The medical charts of patients who were diagnosed with IP between July 2008 and August 2013 at the Department of

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TABLE 1. Krouse Classification for IP

T1	The tumor is totally limited to the nasal cavity. No malignancy.
T2	The tumor involves the osteomeatal complex, the ethmoid cells, and/or the medial portion of the maxillary sinus, with or without involvement of the nasal cavity. No malignancy.
T3	The tumor involves the lateral, inferior, superior, anterior, or posterior walls of the maxillary sinus; the sphenoid sinus; and/or the frontal sinus, with or without involvement of the medial portion of the maxillary sinus, the ethmoid cells, or the nasal cavity. No malignancy.
T4	Tumors with any extranasal/extrasinus extension. Malignant tumor.

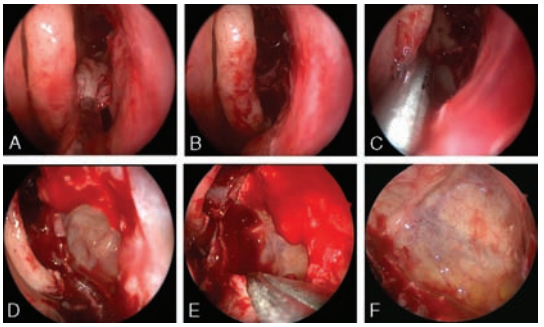

FIGURE 1. Surgical steps of the surgery (A–F).

Otolaryngology, Baskent University Hospital, Ankara, Turkey, were retrospectively reviewed. Eight patients who underwent EMMM were included in the study. The patients were preoperatively evaluated by nasal endoscopy, computed tomography, and magnetic resonance imaging. Staging was done according to the Krouse[9] classification (Table 1). Surgery was primary in 6 patients. Two of the patients were revision cases who had been operated on in another hospital before. Exclusion criteria were the use of external approaches, removal of the nasolacrimal duct and/or the inferior turbinate during surgery, and the presence of malignancies.

Surgical Technique

Endoscopic modified medial maxillectomy was performed under general anesthesia, using 0-, 45-, and 70-degree rigid endoscopes by the 2 senior authors (S.S.E. and F.B.). Cotton swabs soaked with cocaine 4% were placed in both nasal cavities; thus, nasal decongestion was achieved before surgery. As a first step, the exophytic part of the IP was debulked with forceps to provide a sufficient amount of specimen for histopathologic evaluation and to reveal the attachment of the tumor (Fig. 1A). Then, we removed the anterior ethmoid cells and shaved the lateral surface of the middle turbinate to achieve a better visualization (Figs. 1B, C). If an involvement was suspected, we removed the anterior part of the middle turbinate. A wide maxillary antrostomy was performed, and then the medial maxillary wall was removed from the posterior wall of the maxillary sinus posteriorly to the nasolacrimal duct anteriorly (Fig. 1D). Meanwhile, we removed the posterior part of the inferior turbinate if it is involved by the tumor. If the tumor did not involve the inferior turbinate, we elevated the soft tissue of the inferior turbinate behind the level of the nasolacrimal duct and removed the bony turbinate to visualize the medial wall of the maxillary sinus better. Normal mucosa around the attachment point of the tumor was removed, and the underlying bone was drilled with a diamond burr (Fig. 1E). Finally, all maxillary sinus walls were controlled for a tumor remnant by angled endoscopes (Fig. 1F). Posterior ethmoidectomy, sphenoid sinusotomy, and/or frontal sinusotomy was performed according to the tumor extension. The surgical field was supported by gel foams, and the nasal cavity was packed with a glove finger.

The patients were discharged the day after surgery. Oral antibiotics were prescribed for 7 days, and nasal irrigation was prescribed for 1 month postoperatively.

RESULTS

Table 2 shows the demographics of the patients. All of the patients were men. The mean age at diagnosis was 61.3 years (range, 33–84 y). All IPs were located on the medial wall of the maxillary sinus. Extent of the lesions can be seen in Table 2. The nasolacrimal duct was preserved in all cases. The inferior turbinate was completely preserved in 5 patients, and the anterior part of the inferior turbinate was preserved in 3 patients. The middle turbinate was partially resected in 4 patients who had middle turbinate involvement. Histopathologic examination revealed IP in 7 patients and oncocytic type of IP in 1 patient. There was no finding of malignancy in any of the specimens. The mean follow-up period was 30.8 months (range, 12–60 mo). None of the patients had recurrence or major complications (Fig. 2). The postoperative complaints were minor hemorrhagic discharge and crusting for the first few weeks.

DISCUSSION

External surgical approaches for the management of IP have been restricted to high-stage (T4) tumors, and endoscopic approaches have been used in primary and in some of the revision cases.[10] Most surgeons prefer to perform endoscopic medial maxillectomy for the

TABLE 2. Patients' Demographics

Age	Sex	Side/Stage	Primary Versus Secondary	Follow-up, mo	Extent of the Lesion
57	Male	Right/T2	Primary	16	Nasal cavity
68	Male	Left/T2	Secondary	22	Nasal cavity and anterior ethmoid cells
33	Male	Right/T2	Primary	18	Nasal cavity and anterior ethmoid cells
68	Male	Right/T3	Secondary	12	Anterior and posterior ethmoid cells, sphenoid sinus, frontal recess
58	Male	Left/T3	Primary	58	Anterior and posterior ethmoid cells, sphenoid sinus
72	Male	Left/T3	Primary	47	Anterior and posterior ethmoid cells, sphenoid sinus
48	Male	Left/T2	Primary	60	Nasal cavity
64	Male	Left/T2	Primary	14	Nasal cavity

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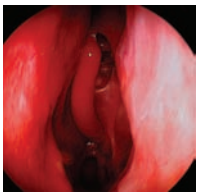

FIGURE 2. Endoscopic view of the surgical site at 12 months postoperatively. The nasolacrimal duct and the anterior part of the inferior turbinate were preserved, and the patient is disease-free.

treatment of IP. In this surgery, the lateral nasal wall is completely removed together with the nasolacrimal duct and the inferior turbinate. Thus, most patients complain about nasal crusting, atrophic rhinitis, and epiphora.

Several endoscopic modified maxillectomy techniques for the treatment of IP originating from the maxillary sinus have been defined in the literature.[5–7] However, these techniques are particularly suggested for tumors involving the anterior or the anteromedial wall of the maxillary sinus. The techniques briefly include separation of the inferior turbinate from the lateral wall and identifying the nasolacrimal duct at the beginning of the surgery and replacing them at the end of the surgery. However, there are no data regarding the functions of replaced inferior turbinate postoperatively.[6] Invasive procedures such as dacryocystorhinostomy and/or resection of the piriform aperture could be a part of such a surgery.[11]

Our surgical approach is based on the philosophy of attachment-oriented surgery for IP, as stated in the literature. Landsberg et al[12] suggested that attachment site diameters are small even in case of extensive tumor, and the identification of the attachment at the beginning of the surgery would provide better outcomes and lower morbidity. Pagella et al[13] similarly advocated that pedicle-oriented surgery would provide good surgical results and preservation of the uninvolved structures such as the inferior turbinate and the nasolacrimal duct. These authors noted that the attachment was always limited to a single site, and a diffuse attachment site along the involved sinus was never seen in their patients. In the beginning of the surgery, debulking of the tumor and identifying the attachment of the tumor allowed us to perform minimally invasive surgery and to remove the tumor precisely. Thus, the patients whose tumors originated from the medial wall of the maxillary sinus underwent EMMM. Limited extensions to the anterior maxillary sinus wall or below the level of attachment of the inferior turbinate could be removed using curved instruments in our cases.

As described in this article, if the soft tissue of the inferior turbinate is separated and the bony attachment of the inferior turbinate around the attachment of the tumor is removed carefully, it is possible to obtain safe surgical margins. None of the tumors affected the inferior turbinate anterior to the nasolacrimal duct in our patients. If the posterior part of the inferior turbinate was involved, we cut the inferior turbinate just behind the nasolacrimal duct and removed the diseased part of this structure. Rutherford and Brown[8] reported that, if there is no tumor involvement, the inferior turbinate and the nasolacrimal duct can be protected without further manipulations, in selected cases. These authors described 4 patients: the attachment of the tumor was on the posterior maxillary sinus wall in 2 patients, on the lateral maxillary sinus wall in 1 patient, and on the inferior orbital wall in 1 patient. Noninvasive approaches seem to be adequate for different locations in the maxillary sinus, if the attachment is well defined at the beginning of the surgery.

In the current study, no patients complained about atrophic rhinitis and epiphora. Only transient nasal crusting and minor hemorrhagic discharge were experienced by the patients for a few weeks postoperatively. None of the patients presented with a residual disease or recurrence during the follow-up (mean, 30.8 mo). Because most of the recurrences occur in the first postoperative year,[14,15] we conclude that our technique is successful to control the disease.

There are limitations of the current study. First, a relatively small number of patients were included. Second, operated tumors were located on one single area of the maxillary sinus. In cases that the tumors cannot be reached even by the curved instruments and angled endoscopes, different approaches are required, and the inferior turbinate and/or the nasolacrimal duct may be sacrificed to achieve a successful tumor control.

CONCLUSIONS

Endoscopic modified medial maxillectomy, which is described in this study, provides a minimally invasive surgical technique to avoid surgical complications for selected patients with IP. The critical point for our approach is to identify the attachment point of IP during the operation and to perform EMMM if it originates from the medial maxillary sinus wall.

REFERENCES

1. Philpott CM, Dharamsi A, Witheford M, et al. Endoscopic management of inverted papillomas: long-term results—the St. Paul's Sinus Centre experience. *Rhinology* 2010;48:358–363
2. Lund V, Stammberger H, Nicolai P, et al. European position paper on endoscopic management of tumors of the nose, paranasal sinuses and skull base. *Rhinol Suppl* 2010;1:1–143
3. Buchwald C, Franzmann MB, Tos M. Sinonasal papillomas: a report of 82 cases in Copenhagen County, including a longitudinal epidemiological and clinical study. *Laryngoscope* 1995;105:72–79
4. Busquets JM, Hwang PH. Endoscopic resection of sinonasal inverted papilloma: a meta-analysis. *Otolaryngol Head Neck Surg* 2006;134:476–482
5. Wang C, Han D, Zhang L. Modified endoscopic maxillary medial sinusotomy for sinonasal inverted papilloma with attachment to the anterior medial wall of maxillary sinus. *ORL J Otorhinolaryngol Relat Spec* 2012;74:97–101
6. Gras-Cabrerizo JR, Massegur-Solench H, Pujol-Olmo A, et al. Endoscopic medial maxillectomy with preservation of inferior turbinate: how do we do it? *Eur Arch Otorhinolaryngol* 2011;268:389–392
7. Nakayama T, Asaka D, Okushi T, et al. Endoscopic medial maxillectomy with preservation of inferior turbinate and nasolacrimal duct. *Am J Rhinol Allergy* 2012;26:405–408
8. Rutherford KD, Brown SM. Endoscopic resection of maxillary sinus inverted papillomas with inferior turbinate preservation. *Otolaryngol Head Neck Surg* 2010;142:760–762
9. Krouse JH. Development of a staging system for inverted papilloma. *Laryngoscope* 2000;110:965–968
10. Lian F, Juan H. Different endoscopic strategies in the management of recurrent sinonasal inverted papilloma. *J Craniofac Surg* 2012;23:e44–e48
11. Weber RK, Werner JA, Hildenbrand T. Endonasal endoscopic medial maxillectomy with preservation of the inferior turbinate. *Am J Rhinol Allergy* 2010;24:132–135
12. Landsberg R, Cavel O, Segev Y, et al. Attachment-oriented endoscopic surgical strategy for sinonasal inverted papilloma. *Am J Rhinol* 2008;22:629–634
13. Pagella F, Pusateri A, Giourgos G, et al. Evolution in the treatment of sinonasal inverted papilloma: pedicle-oriented endoscopic surgery. *Am J Rhinol Allergy* 2014;28:75–81
14. Wolfe SG, Schlosser RJ, Bolger WE, et al. Endoscopic and endoscope-assisted resections of inverted papillomas. *Otolaryngol Head Neck Surg* 2004;131:174–179
15. Lawson W, Patel ZM. The evolution of management for inverted papilloma: an analysis of 200 cases. *Otolaryngol Head Neck Surg* 2009;140:330–335

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Endoscopic Endonasal Versus Transfacial Approach for Blowout Fractures of the Medial Orbital Wall

Mario Pagnoni, MD, Filippo Giovannetti, MD, Giulia Amodeo, MD, Paolo Priore, MD, and Giorgio Iannetti, MD

Abstract: In the last decades, the introduction of computed tomography has allowed an increase in the number of diagnosed fractures of the medial orbital wall. To repair medial wall fractures, many surgical techniques have been proposed (1), each one with its advantages and disadvantages. In this study, we compared endoscopic endonasal and transcutaneous reduction approaches in terms of surgery time and clinical outcome. Between 2001 and 2005, 81 patients with orbital wall fractures were treated at our department. Among these 81 patients, 24 (29.63%) were affected by a medial orbital fracture. Patients with fracture to both floor and medial walls underwent floor reduction by a transcutaneous subpalpebral approach (n = 9, 11.1%), whereas patients with isolated medial wall fracture underwent medial wall reduction by a transcutaneous subpalpebral approach using alloplastic implants (n = 8, 9.88%) or were treated by endoscopic approach (n = 5, 6.17%). After surgery, oculomotor function improved in all 22 patients. None of the patients had complications. Computed tomography revealed a well-consolidated site of fracture in both endoscopic endonasal and transcutaneous approaches. The average operating time for endoscopic endonasal and transfacial approach was 50 and 45 minutes, respectively.

In this paper, the author proposed a results comparison between the endoscopic approach and the transcutaneous one.

Key Words: Medial orbital wall fractures, endoscopic approach, transcutaneous approach

Blowout fractures of the medial orbital wall are observed more frequently now than in the past, as a result of the introduction of computed tomography and the increased incidence of high-energy impact injuries. Medial wall fractures are often followed by posttraumatic enophthalmos caused by orbital volume increase or other complications, such as diplopia. The treatment should be started as early as possible because a delay can cause enophthalmos that leads to clinical and diagnostic failure.[1]

Medial wall fractures can be repaired by surgical intervention via various access routes, for example, transcutaneous approach, transconjunctival approach, and endoscopic endonasal approach.[2–6] Each approach has its advantages and disadvantages. The choice of approach depends on the anatomic location of the fracture and on its extent. While making the choice, it is also important to consider the extent of medial wall exposure, the size of implant or bone graft insertion, and the possibility of surgical damage of important periorbital or intraorbital structures or of postoperative scar deformities .[3,7]

The purpose of this study was to compare endoscopic endonasal and transcutaneous reduction approaches in terms of surgery time and clinical outcome.

MATERIALS AND METHODS

In the period from 2001 to 2005, 81 patients with orbital wall fractures were treated at the Oral and Maxillofacial Surgery Department in the General Hospital Umberto I, University of Rome La Sapienza. The study was conducted in compliance with the principles of the Declaration of Helsinki. Each patient gave its own written informed consent. Among these 81 patients, 24 were affected by a medial orbital fracture (Fig. 1): 9 cases of fracture to both floor and medial walls and 15 cases of isolated medial wall fractures. The patients included 62 men and 19 women, aged between 11 and 80 years (average, 34 years). The causes of fracture were traffic accident in 28 cases, physical assault in 24, accidental trauma in 19, sports injury in 4, work accident in 4, and neurologic accident in 2. The period between injury and surgery varied between 0 and 51 days (average, 6 days).

Each patient underwent an eye examination. An ophthalmologist may diagnose the medial wall fracture. Through examination of eye movements by duction and version test, it is possible to detect eye movement deficit caused by the fracture. Two tests were performed. The red light test analyzes the capacity to see 1 or 2 different colors. In front of the patient, a target light is placed, whereas in front of the eye with better visual acuity, a red filter density is placed, which promotes diplopia. The light is moved in different positions of gaze, and the patient should indicate the relative position of the 2 lights. In case of binocular vision, the patient will see only 1 light, whereas in case of diplopia, the patient will see 2 lights of different colors. In the Hess Lancaster screen test, a red glass is placed in front of the right eye and a green glass in front of the left eye. A bright colorful green stripe is projected on the screen, and the patient has to superimpose its stripe to the one projected. The strip light is projected on the screen at precise points corresponding to the action of the muscles in the different positions of gaze. The distance between the 2 stripes represents the subjective ocular deviation. The position of the corneal apex was measured by exophthalmometry, which allows to quantify the degree of enophthalmos measuring the above position. All patients were diagnosed with preoperative computed tomography in axial and coronal projections, which confirmed the prolapse of the orbital contents. Before treatment, 9 of the 22 patients had no diplopia, 7 had diplopia on downward gaze, 2 had diplopia on upward gaze, 2 had total diplopia, and on 2 patients, diplopia could not be assessed.

The choice of surgical procedure depended on the nature of the fracture: patients with fracture in both floor and medial walls underwent floor reduction by transcutaneous subpalpebral approach; of the 15 patients with isolated medial wall fracture, 2 had no treatment, 8 underwent medial wall reduction by a transcutaneous subpalpebral approach using alloplastic implants to support the fractured walls, and 5 patients were treated by endoscopic approach.

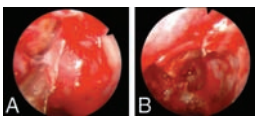

FIGURE 1. Medial orbital wall fracture endoscopic image.

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Patient follow-up varied between 6 months and 1 year. Clinical outcome was evaluated by computed tomography and on the basis of postoperative residual diplopia.

Surgical Technique

All operations were performed under general anaesthesia.

Endoscopic Endonasal Approach

A solution of lidocaine (2%) and adrenaline 1:200,000 was injected into the middle turbinate, uncinate process, and septum. A horizontal incision was made over the uncinate process, and the uncinate process was excised to gain access to the ethmoid bone. This is a standard technique for an endonasal ethmoidal operation. Then, the ethmoidal bulla was opened.

The medial orbital wall fractured bone chips on the orbital periosteum were preserved. The fractured partition walls of the ethmoid air cells were removed to prevent posttraumatic and/or postoperative paranasal cysts; however, when preserved, the fractured bones of the medial orbital wall can help in regenerating a firmer medial wall. We have performed the fixation by Merocel packing placed between the reduced medial wall and the middle turbinate for a mean period of 7 days (SD, 4.59 days).

Transfacial Approach

The incision was made over the medial canthal area, and the periosteum was dissected together with the medial canthal tendon; the herniated orbital contents were restored into the orbital cavity. The defect of the medial wall was reconstructed with a bovine pericardium graft.

Transconjunctival Approach

A transconjunctival incision was made to access the bone fracture, which was reduced and fixed by using alloplastic prosthesis.

RESULTS

After surgery, oculomotor function improved in all 22 patients with medial orbital wall fractures. None of the patients had complications. Diplopia was completely solved in 16 patients, but it persisted on downward gaze in 2 patients and on upward gaze in 1 patient. Computed tomography revealed a well-consolidated site of fracture in both endoscopic endonasal and transcutaneous approaches (Figs. 2 and 3).

Only one patient, who underwent an open approach, reported permanent diplopia in the first 5 days after surgery. The other 11 patients referred no postsurgery diplopia.

Computed tomography performed in each patient at 7 days and then at 3 months after surgical intervention, showed good stabilization of the medial orbital wall with no medial rectus muscle entrapment.

In only 2 patients treated by open technique, the cutaneous scar remained visible at 6 months after surgical treatment.

The average operating time was 50 minutes in the endoscopic endonasal group and 45 minutes in the transfacial reduction group (SD, 5.20 minutes).

DISCUSSION

In this paper, endoscopic endonasal and transcutaneous approaches for the repair of medial wall fracture are compared in terms of surgery time and clinical outcome.[8] External approach was first presented by Maisel et al[9] in 1975. In 1978, Pearl and Vistnes[1] used autogenous, homologous, and alloplastic material to repair the defect in orbital wall fractures, and in 1982, Bartkowski and Krzystkowa[10] presented a transfacial approach by transconjunctival or subciliary incision on the basis of 90 patients with floor and medial wall fracture.

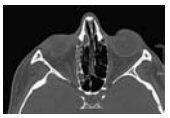

FIGURE 2. Axial TC post.

FIGURE 3. Coronal TC post.

The transcutaneous approach is performed by a subciliary incision, a subpalpebral incision, a superior and/or inferior eyelid incision, a medial canthal incision, and a bicoronal incision. An inferior or a medial conjunctival incision performs the transconjunctival approach.

The transcutaneous approach has some disadvantages, such as external scarring and, in the medial canthal incision, the risk of alloplastic implant deformity and infection. Bicoronal incision leaves no facial scars but is considered an invasive approach. The transconjunctival approach has the advantage of leaving no external scars, but it could provide an inadequately narrow surgical field for the reduction.[4,5,11,12]

Endoscopic endonasal surgery was developed in Germany and Japan.[13–16] It has been applied for several pathologies, such as paranasal sinusitis, cerebrospinal rhinorrhoea, and decompression of optic neuropathy. The use of the endoscope requires an experienced surgeon. This surgery projects a magnified operative field on the television monitor, so it offers a clear view of the operation site. Michel[17] first described endonasal endoscopic reduction of the medial orbital wall in 1993. Since then, several authors have described the endoscopic endonasal route access for reduction of medial orbital fracture.[18–20] The advantages of this approach are the following: it is a minimally invasive approach with short operating time; and it leaves no external scarring, thus providing better aesthetic results. In endoscopic endonasal surgery, the herniated orbital contents are just minimally pressed and relocated to the original position. Damage of the optic nerve, a severe complication of medial orbital wall reduction by transcutaneous approach, is rarely observed in endoscopic endonasal surgery because the instruments placed along the fractured fragments do not invade the optical canal.[21,22] The success of symptom improvement (eg, diplopia, eye movement and enophthalmos) is comparable with more invasive approaches.

In light of these observations, endoscopic endonasal surgery can be considered a good alternative for the repair of medial orbital wall fractures.

REFERENCES

1. Pearl RM, Vistnes LM. Orbital blowout fractures: an approach to management. *Ann Plast Surg* 1978;1:267–270
2. Lee CS, Yoon JS, Lee SY. Combined transconjunctival and transcaruncular approach for repair of large medial orbital wall fractures. *Arch Ophthalmol* 2009;127:291–296
3. Han K, Choi JH, Choi TH, et al. Comparison of endoscopic endonasal reduction and transcaruncular reduction for the treatment of medial orbital wall fractures. *Ann Plast Surg* 2009;62:258–264
4. Kim DW, Choi SR, Park SH, et al. Versatile use of extended transconjunctival approach for orbital reconstruction. *Ann Plast Surg* 2009;62:374–380
5. Kim HS, Kim SE, Evans GR, et al. The usability of the upper eyelid crease approach for correction of medial orbital wall blowout fracture. *Plast Reconstr Surg* 2012;130:898–905

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6. Cheong EC, Chen CT, Chen YR. Broad application of the endoscope for orbital floor reconstruction: long-term follow-up results. *Plast Reconstr Surg* 2010;125:969–978
7. Belli E, Matteini C, Mazzone N. Evolution in diagnosis and repairing of orbital medial wall fractures. *J Craniofac Surg* 2009;20:191–193
8. Jin HR, Yeon JY, Shin SO, et al. Endoscopic versus external repair of orbital blowout fractures. *Otolaryngol Head Neck Surg* 2007;136:38–44
9. Maisel RH, Acomb TE, Cantrell RW. Medial orbital blow-out fracture: a case report. *Laryngoscope* 1975;85:1211–1215
10. Bartkowski SB, Krzystkowa KM. Blow-out fracture of the orbit. Diagnostic and therapeutic considerations, and results in 90 patients treated. *J Maxillofac Surg* 1982;10:155–164
11. Novelli G, Ferrari L, Sozzi D, et al. Transconjunctival approach in orbital traumatology: a review of 56 cases. *J Craniomaxillofac Surg* 2011;39:266–270
12. Scolozzi P. Reconstruction of severe medial orbital wall fractures using titanium mesh plates placed using transcaruncular-transconjunctival approach: a successful combination of 2 techniques. *J Oral Maxillofac Surg* 2011;69:1415–1420
13. Chen CT, Chen YR. Endoscopically assisted repair of orbital floor fractures. *Plast Reconstr Surg* 2001;108:2011–2018
14. Chen CT, Chen YR, Tung TC, et al. Endoscopically assisted reconstruction of orbital medial wall fractures. *Plast Reconstr Surg* 1999;103:714–720quiz 721
15. Hinohira Y, Takahashi H, Komori M, et al. Endoscopic endonasal management of medial orbital blowout fractures. *Facial Plast Surg* 2009;25:17–22
16. Jin HR, Shin SO, Choo MJ, et al. Endonasal endoscopic reduction of blowout fractures of the medial orbital wall. *J Oral Maxillofac Surg* 2000;58:847–851
17. Michel O. Isolated medial orbital wall fractures: results of minimally invasive endoscopically controlled endonasal surgical technique. *Laryngorhinootologie* 1993;72:450–454
18. Lee HM, Han SK, Chae SW, et al. Endoscopic endonasal reconstruction of blowout fractures of the medial orbital walls. *Plast Reconstr Surg* 2002;109:872–876
19. Lee TH, Lee HM, Lee JM, et al. Endoscopic reduction of orbital medial wall fracture using rotational repositioning of the fractured: lamina papyracea fragment. *J Craniofac Surg* 2014;25:460–462
20. Park CH, Choi DJ, Lee JH, et al. Endoscopic reduction of medial orbital wall fractures using the rolled silastic sheet technique. *J Trauma* 2009;66:1421–1424
21. Rhee JS, Lynch J, Loehrl TA. Intranasal endoscopy-assisted repair of medial orbital wall fractures. *Arch Facial Plast Surg* 2000;2:269–273
22. Sanno T, Tahara S, Nomura T, et al. Endoscopic endonasal reduction for blowout fracture of the medial orbital wall. *Plast Reconstr Surg* 2003;112:1228–1237discussion 1238

Spatial Changes of the Chin in the Vertical and Sagittal Planes After Superior Repositioning of the Maxilla

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Purpose: The mandible autorotates after maxillary superior repositioning. The aim of this study was to address the changes in chin position in the vertical and sagittal planes after maxillary superior repositioning.

Materials and Methods: This cross-sectional study assessed participants who had class I occlusion with vertical maxillary excess and underwent maxillary superior repositioning. Two lateral cephalograms were taken in central occlusion and natural head position. The amount of maxillary superior repositioning was documented for every participant according to lateral cephalometric indices. The distance between the most prominent point of the chin and a perpendicular line from *N* to the Frankfort line was used to determine the sagittal changes of the chin before and after surgery. The distance from the *N* point to *Me* was used to assess the vertical changes of the chin before and after operation. The Pearson correlation test was used to determine the correlation between the amount of maxillary superior repositioning and the vertical and horizontal changes of the chin. The linear regression model was applied to predict the changes of the chin (dependent factor) according to the vertical change of the maxilla (predictive factor). The occlusal plane angle change, mandibular length, and mandibular plane angle were considered as variable factors.

Results: Twenty participants were studied. Analysis of the data demonstrated a significant correlation between the maxillary superior repositioning (predictive factor) and the horizontal and vertical changes of the chin. For every 1 mm of vertical change in the maxilla, the chin could be expected to move 0.21 mm horizontally. For a standard deviation increase of 1 in the maxillary position, the chin advanced by 0.753 of the standard deviation ($\beta = 0.753$). For every 1-mm change of the maxilla vertically, it could be estimated that the chin moved 0.71 mm vertically when the amount of maxillary impaction was 8 mm or less. For an increase in standard deviation of 1 in the position of the maxilla, the chin moved superiorly by 0.711 of a standard deviation ($\beta = 0.711$). In maxillary superior repositioning greater than 8 mm, for every 1 mm of superior repositioning, the chin moved 0.44 mm superiorly. There was a positive correlation between the occlusal plane change, mandibular length, mandibular plane angle as well as the vertical and horizontal changes of the pogonion ($P = 0.001$).

Conclusions: The chin position after maxillary superior repositioning can be predicted according to the amount of maxillary vertical changes. The vertical change of the chin is more predictable than the horizontal change.

Key Words: Maxillary superior repositioning, chin, pogonion, osteotomy

Vertical maxillary excess (VME) is present in a large proportion of dentofacial deformities; a number of people with VME seek treatment of a gummy smile appearance.[1,2] It is well known that excessive vertical growth of the maxilla causes the mandible to rotate downward and backward (clockwise rotation). Mandibular

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rotation[3,4] guides the mandible to dental class II position in patients with class I occlusion. Treatment of these patients mandates repositioning the maxilla superiorly to obtain ideal tooth exposure. One of the consequences is autorotation of the mandible and displacement of bony chin position and the pogonion.[5] In orthognathic surgery that includes vertical repositioning of the maxilla and autorotation of the mandible, the mandible rotates from the patient's original occlusal position to a point that is located more superiorly.

It is crucial to predict the amount of mandibular autorotation in maxillary superior repositioning and pogonion displacement in vertical and sagittal planes because it causes significant changes in the lower one third of the face. Pogonion changes have a positive correlation with the amount of maxillary superior repositioning. A few studies have focused on the autorotation of the chin.[6,7] However, the amount of change in the sagittal and vertical planes after maxillary superior repositioning is a matter of controversy.

Change of chin position after maxillary superior repositioning due to mandibular autorotation is well known. However, the amount of this change has been controversial. In this study, we tried to answer the following questions: "Is there a linear correlation between maxillary superior repositioning and vertical and horizontal changes of the pogonion?" and "Can we predict chin changes according to the amount of maxillary superior repositioning?"

MATERIALS AND METHODS

This cross-sectional study sought to evaluate changes of the chin position after maxillary superior repositioning from September 1, 2011 to October 31, 2012, and was approved by the research committee of the Medical Ethics Group of Shiraz University of Medical Sciences. Eligible participants for inclusion had class I occlusion with vertical maxillary excess that needed maxillary superior repositioning without anterior-posterior movement or rotation of the maxilla. Participants were excluded from the study if they had a history of genioplasty, mandibular osteotomy, genial augmentation, temporomandibular joint deformity, occlusal adjustment after the surgery, or previous mandibular fractures.

Cephalometric Technique

The participants were positioned in the cephalostat (Orthoceph; Siemens AG, Munich, Germany), and then the head holder was adjusted until the ear rods could be positioned into the ears without moving the patient. All radiographs were taken in the natural head position and in a centric occlusion and with a metric ruler in front of the midfacial vertical line. No occipital supplement was used. According to cephalometric standards, the film distance to the x-ray tube was fixed at 150 cm and the film distance to the midsagittal plane of the patient's head was fixed at at 18 cm. Two lateral cephalograms were taken immediately before the surgery and 1 week after the surgery.

Tracing Technique

All the cephalograms were traced by a single operator on Garware matte acetate tracing paper of 0.003″ thickness with 3-H microlead pencil. Cephalometric landmarks were located, identified, and marked.

Preoperative and postoperative cephalometric analyses were used to determine the amount of the maxillary superior repositioning The analysis measured the linear distances (in millimeters) from the maxillary landmarks (U1 and U6) to vertical reference lines (Fig. 1). The vertical reference was established perpendicular to SN_7, with its origin at the sella.[8] The changes of *A* point in the anterior-posterior direction were measured to the SN perpendicular line in the *N* point.

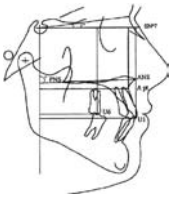

FIGURE 1. The linear distances (in millimeters) from the maxillary landmarks (U1 and U6) to vertical reference lines. The vertical reference was established perpendicular to SN_7, with its origin at the sella.

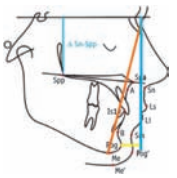

FIGURE 2. Measurement of the horizontal and vertical changes of the chin after maxillary superior repositioning.

The distance between the most prominent point of the chin and a perpendicular line from *N* to the Frankfort line was used to determine the sagittal changes of the chin before and after the surgery. The distance from the *N* point to the *Me* point was applied to assess the vertical changes of the chin before and after the operation. All participants underwent conventional Le Fort I osteotomy with rigid fixation using 4 plates and 16 screws (Fig. 2). The occlusal plane was formed by the half distance point between the incisal edges of the maxillary and mandibular incisors and the mesial cuspid tips of the mandibular bilateral permanent first molars. The mandibular length was measured from the condylion (the most superior point on the head of the condylar head) to the pogonion. The mandibular plane angle was determined as an angle between *GO-Me* line and *SN*. Occlusal plane change, mandibular plane angle, and the mandibular length were considered as variable factors.

Statistical Analysis

The statistical analyses were performed using the statistical package SPSS. The Pearson correlation test was used to determine the correlation between the amount of maxillary superior repositioning and the vertical and horizontal changes of the chin. The linear regression model was applied to predict changes of the chin (dependent factor) according to the vertical change of the maxilla and occlusal plane angle changes (predictive factor).

RESULTS

Twenty participants (12 females and 8 males with the mean [SD] age of 24.5 [3.2] y) who underwent maxillary superior repositioning were studied. The mean (SD) maxillary superior repositioning was 9.05 (3.30) mm, the mean (SD) horizontal change of the chin was 6.15 (2.10) mm, and the mean (SD) vertical change of the chin was 7.1 (2.51) mm. The mean (SD) anterior-posterior changes of the maxilla at the *A* point was 1.25 (0.98) mm. The mean (SD) of mandibular length was 113.4 (3.69) mm, and the mean (SD) of mandibular plane angle 26.7 (2.07). The mean (SD) of the occlusal plane change was 5.25 (2.46).

Results showed a positive correlation between the mandibular length and the vertical and horizontal changes ($P < 0.001$). There

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TABLE 1. Correlation Between Variables and the Horizontal Changes in the Pogonion

Variables		Horizontal Change, mean (SD)	Pearson Correlation Test
Occlusal plane change	5.25 (2.46)	6.15 (2.10)	$P < 0.001$
Mandibular length	113.4 (3.69)	6.15 (2.10)	$P < 0.001$
Mandibular plane angle	26.7 (2.07)	6.15 (2.10)	$P < 0.001$

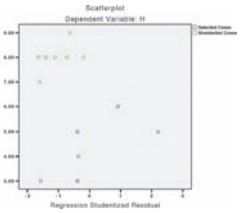

FIGURE 3. The horizontal change of the chin was correlated with the amount of maxillary superior repositioning when the amount of maxillary superior repositioning was 8 mm or less.

was a positive correlation between the mandibular plane angle and the vertical and horizontal changes ($P < 0.001$) (Tables 1 and 2).

Data analysis demonstrated a significant correlation between the maxillary superior repositioning (predictive factor) and the horizontal and vertical changes of the chin ($P < 0.001$) (Table 1).

The linear regression model showed that approximately 56.7% of the horizontal change of the chin could be accounted for by the amount of maxillary superior repositioning when the amount of impaction was 8 mm or less ($R^2 = 0.567$) (Fig. 3). For every 1 mm of vertical change in the maxilla, the chin could be expected to move 0.21 mm horizontally when the maxilla was impacted 8 mm or less. For a standard deviation increase of 1 in the maxillary position, the chin advanced by 0.753 of the standard deviation ($\beta = 0.753$). When maxillary impaction was more than 8 mm, the regression model did not follow a linear correlation between the amount of maxillary superior repositioning and horizontal change in the pogonion.

The assessment of the data using a linear regression model demonstrated that approximately 50.6% of the vertical change of the chin could be predicted by knowing the amount of the maxillary superior repositioning when the amount of maxillary impaction was 8 mm or less ($R^2 = 0.506$) (Fig. 4). For every 1-mm change of the maxilla vertically, the chin was estimated to move 0.75 mm vertically. For a standard deviation increase of 1 in the position of the maxilla, the chin moved superiorly by 0.927 of the standard deviation ($\beta = 0.711$). When the amount of maxillary impaction was more than 8 mm, the regression model showed that 17.9% of the vertical change of the chin was predicted by the amount of maxillary impaction ($R^2 = 0.179$). For every 1 mm of superior repositioning of the maxilla, the chin moved 0.44 mm superiorly. For a standard deviation increase of 1 in maxillary impaction, the chin moved superiorly by 0.423 of the standard deviation ($\beta = 0.423$).

The analysis of the data showed a positive correlation between the horizontal and vertical changes of pogonion and the occlusal plane angle changes ($P = 0.001$) (Table 3). A linear regression model demonstrated that 84.1% of vertical change of the chin could be predicted by the amount of occlusal plane change ($R^2 = 0.841$). A total of 78.4% of the horizontal change in the pogonion could be accounted by the amount of occlusal plane change ($R^2 = 0.784$).

DISCUSSION

In the VME cases, an increase in the lower one third of the face is a common problem. Nevertheless, increasing maxillary height causes a clockwise rotation in the mandible and the chin moves posteriorly and inferiorly. Maxillary superior repositioning is the choice treatment for patients with VME. After the mentioned surgery, the pogonion will rotate anteriorly and superiorly[3,4] (Figs. 5–7). The exact ratio between the amount of superior repositioning and the amount of anterior and superior autorotation of pogonion is a controversial issue. Understanding how the chin position will change after maxillary superior repositioning helps surgeons predict and suggest other adjunctive treatments such as advancement genioplasty.[9]

This study pointed out the high predictability of the chin position based on the amount of maxillary superior repositioning. The predictability of the vertical change was greater than that of the horizontal change. One of the main problems in the evaluation of the chin position after the maxillary superior repositioning is the change of the pogonion point. When the chin position changes after the maxillary surgery, the most prominent point will change. Consequently, determining the pogonion as a reference point for the chin position is unreliable. In the current study, we considered *Me* as a reference point for the vertical changes. However, the most prominent point was used to determine the horizontal change. Our study did not show predictability or a linear correlation between the horizontal change and the maxillary impaction when the amount of maxillary superior repositioning was more than 8 mm. The possible reason for this finding may be rotation and translation of the condyle after maxillary superior repositioning. The condyle does not rotate simply in the glenoid fossa.[10] The same reason can explain the different superior movement of the pogonion when the maxillary impaction was more than 8 mm and the lack of a linear correlation between them. The current study also demonstrated the effect of the mandibular length on the amount of changes in the pogonion. Participants with a long mandibular length may have a great change in the pogonion after maxillary superior repositioning. Increase in the mandibular length causes the larger arch length during mandibular autorotation. Subsequently, positional change of the pogonion in a mandible with a long length is more than that of a short mandible.

Few cases of absolute maxillary impaction without anterior or posterior maxillary movement could be imagined. In many instances, the maxilla can be moved somewhat posteriorly or anteriorly; thus, simultaneous mandibular surgery is not required to achieve the desired result. In some cases, however, simultaneous mandibular advancement or setback is required to achieve the

TABLE 2. Correlation Between Variables and the Horizontal Changes in the Pogonion

Variables		Horizontal Change, mean (SD)	Pearson Correlation Test
Occlusal plane change	5.25 (2.46)	6.15 (2.10)	$P < 0.001$
Mandibular length	113.4 (3.69)	6.15 (2.10)	$P < 0.001$
Mandibular plane angle	26.7 (2.07)	6.15 (2.10)	$P < 0.001$

TABLE 3. Correlation of the Horizontal and Vertical Changes of the Chin With the Amount of Maxillary Superior Reposition

Dependent Factors	Maxillary Superior Reposition, mean (SD)	Pearson Correlation
Horizontal change of the chin, mm	6.15 (2.10)	$P < 0.001$
Vertical change of the chin, mm	7.1 (2.51)	$P < 0.001$

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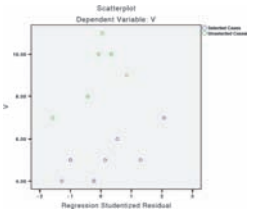

FIGURE 4. The vertical change of the chin was correlated with the amount of maxillary superior repositioning when the amount of maxillary superior repositioning was 8 mm or less.

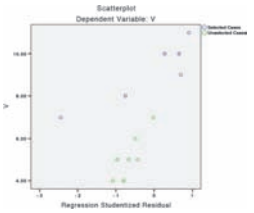

FIGURE 5. The vertical change of the chin was correlated with the amount of maxillary superior repositioning when the amount of maxillary superior repositioning was more than 8 mm.

FIGURE 6. A patient with a case of VME before orthognathic surgery.

desired occlusal and esthetic results.[11] Finding the center of rotation in the mandible after the maxillary impaction can guide the surgeon to better understand the chin position after maxillary superior repositioning. The center of the mandibular condyle was usually used as the rotation center of mandibular autorotation to predict mandibular autorotation paths and final mandibular positions presurgically.[6,12] Wang et al[6] showed that, after maxillary Le Fort I impaction osteotomy, the rotation centers of mandibular autorotation may not be located at the radiographic condylar center of the mandible with large individual variations in position. A study concluded that using the center of the condyle as the center of mandibular rotation in the planning of superior maxillary movement with a Le Fort I osteotomy may cause considerable error in the horizontal position of the maxilla in most cases.[13] It was shown that the mandible rotates around the same point during maxillary impaction surgery as that during initial jaw opening. This point, called the center of mandibular autorotation, could then be used to predict mandibular position and to decide whether only maxillary impaction would be needed to correct the occlusion and the facial profile.[14] It seems that the mandibular plane may play an important role in chin advancement in the sagittal plane. In participants with high mandibular plane angle, the advancement of the chin after maxillary superior repositioning could be expected. However, the chance of chin advancement in low mandibular plane angle after maxillary impaction is minimal.[14] The same results were achieved in our study. As the amount of mandibular plane angle increased, the amount of horizontal and vertical changes in the pogonion raised.

FIGURE 7. Remarkable change of the chin after maxillary superior repositioning.

Steinhäuser et al[7] studied the soft tissue profile after the maxillary impaction. Results demonstrated that the chin advanced, on average, by 79%, whereas the lower face was shortened by as much as 70% in the pogonion point. However, the second evaluation showed that the type and the extent of maxillary impaction led to significant changes in these parameters. Parallel maxillary impaction resulted in 100%, posterior impaction resulted in 80%, and posterior impaction with anterior subsidence resulted in 50% advancement of the mandible in the pogonion point in relation to the distance moved during impaction. Jamilian et al[15] studied the relationship between pogonion advancement and posterior maxillary impaction. They revealed significant correlations between maxillary impaction and pogonion advancement in both the formulas and clinical evaluation. The amounts of anterior facial height reduction and pogonion advancement were almost the same, and the anterior facial height was reduced 1.5 times more than the amount of maxillary impaction. It seems that the mandibular autorotation is a predictable phenomenon after maxillary impaction.[16]

CONCLUSIONS

The chin position after maxillary superior repositioning could be predicted according to the amount of maxillary vertical changes. The vertical change of the chin is more predictable than the horizontal change. The amount of positional change of the chin may be affected by the amount of the mandibular length.

REFERENCES

1. Juggins KJ, Nixon F, Cunningham SJ. Patient- and clinician-perceived need for orthognathic surgery. *Am J Orthod Dentofacial Orthop* 2005;128:697–702
2. Panossian AJ, Block MS. Evaluation of the smile: facial and dental considerations. *J Oral Maxillofac Surg* 2010;68:547–554
3. Sperry TP, Steinberg MJ, Gans BJ. Mandibular movement during autorotation as a result of maxillary impaction surgery. *Am J Orthod* 1982;81:116–123
4. Rekow ED, Speidel TM, Koenig RA. Location of the mandibular center of autorotation in maxillary impaction surgery. *Am J Orthod Dentofacial Orthop* 1993;103:530–536
5. Bryan DC. An investigation into the accuracy and validity of three points used in the assessment of autorotation in orthognathic surgery. *Br J Oral Maxillofac Surg* 1994;32:363–372
6. Wang YC, Ko EW, Huang CS, et al. The inter-relationship between mandibular autorotation and maxillary LeFort I impaction osteotomies. *J Craniofac Surg* 2006;17:898–904
7. Steinhäuser S, Richter U, Richter F, et al. Profile changes following maxillary impaction and autorotation of the mandible. *J Orofac Orthop* 2008;69:31–41
8. Jacobson R, Sarver DM. The predictability of maxillary repositioning in LeFort I orthognathic surgery. *Am J Orthod Dentofacial Orthop* 2002;122:142–154
9. Mortazavi H, Tabrizi R, Mohajerani H, et al. Evaluation of stability in advancement genioplasty: an 18-month follow-up. *Shiraz Univ Dent J* 2010;10:61–64

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10. Naeije M, Huddleston Slater JJ, Lobbezoo F. Variation in movement traces of the kinematic center of the temporomandibular joint. *J Orofac Pain* 1999;13:121–127
11. Epker BN, Fish LC. Surgical superior repositioning of the maxilla: what to do with the mandible? *Am J Orthod* 1980;78:164–191
12. Fish LC, Epker BN. Surgical-orthodontic cephalometric predictiontracing. *J Clin Orthod* 1980;14:36–52
13. Nattestad A, Vedtofte P. Mandibular autorotation in orthognathic surgery: a new method of locating the centre of mandibular rotation and determining its consequence in orthognathic surgery. *J Craniomaxillofac Surg* 1992;20:163–170
14. Nadjmi N, Mommaerts MY, Abeloos JV, et al. Prediction of mandibular autorotation. *J Oral Maxillofac Surg* 1998;56:1241–1247
15. Jamilian A, Showkatbakhsh A, Gholami D, et al. The relation between pogonion advancement and posterior maxillary impaction. *J Craniofac Surg* 2009;20:841–843
16. Wessberg GA, Washburn MC, LaBanc JP, et al. Autorotation of the mandible: effect of surgical superior repositioning of the maxilla on mandibular resting posture. *Am J Orthod* 1982;81:465–472

Recommendations for Therapeutic Decisions of Angiosarcoma of the Scalp and Face

Kun Hwang, MD, PhD, Mu Yeol Kim, BS, and Seung Hyun Lee, BS

Abstract: The aim of this study was to elucidate the therapeutic decision process of angiosarcoma of the scalp and face including treatment modalities, tumor size, tumor grade, and resection margins.

In a PubMed search, 170 abstracts were read and 32 full text articles were reviewed. Among them, 19 articles were analyzed.

Overall, survival did not differ significantly between the surgery group (23.6 ± 11.0 months) and the no surgery group (22.2 ± 8.0 months) ($P = 0.386$ [t test]). Among the patients who had undergone an operation, 64.4% had residual cancer cells at their surgical margin. Survival did not differ significantly according to the positive or negative resection margin ($P > 0.05$ [t tests]). Overall survival of the radiation therapy and chemotherapy group (37.0 ± 0.0 months) was significantly longer than that of the radiation therapy group (22.7 ± 7.6 months) or the chemotherapy group (15.1 ± 4.6 months). Overall survival, local recurrence-free survival, and distant metastasis-free survival were significantly longer in the T1 group (the tumor size being the same or smaller than 5 cm) than the T2 group (tumor larger than 5 cm) ($P < 0.05$ [t test]). The 2-year survival rates and the 5-year survival rates were significantly longer in the T1 group than in the T2 group ($P < 0.000$ [t test]). The overall survival of low grade tumor group (44.8 ± 10.4 months) was more than 2 times longer than the high-grade tumor group (22.3 ± 6.8 months) ($P = 0.000$ [t test]).

Surgeons should remember that they do not have to try to remove all the cancer cells in the operation theater. A combination of radiation and chemotherapy can bring better results than any single regimen. Lastly, early diagnosis and early treatment are essential.

Key Words: Hemangiosarcoma, scalp, face, therapy, survival

Angiosarcoma is a malignant tumor of vascular endothelial cells that arises in the head and neck. It is a rare, difficult-to-treat, and lethal tumor.[1]

Because of a tendency for diffuse, clinically undetectable local spreading, cutaneous angiosarcoma is difficult to treat with surgery alone.[2] In many series, potential treatment options for patients with angiosarcoma of the scalp and face have included surgery, radiation, and chemotherapy.[3]

Despite several retrospective analyses of angiosarcoma, there has been no systematic review for the treatment and prognostic factors of angiosarcomas of the scalp and face.

The aim of this study was to elucidate the therapeutic decision process of angiosarcoma of the scalp and face including treatment modalities, tumor size, tumor grade, and resection margins.

MATERIALS AND METHODS

In a PubMed search, 170 papers were searched using the terms *angiosarcoma*, *scalp*, and *controlled or prospective or restrospective or analysis or survival or follow-up or recurrence or prognosis.*

Studies that did not allow an evaluation of the prognosis of the angiosarcomas were excluded.

The abstracts were read and 32 full-text articles were reviewed. Among them, 19 articles were analyzed (Fig. 1).[1–19] No restrictions on language and publication forms were imposed. However, the searched full-text papers were mostly in English (one in Chinese). All articles were read by 2 independent reviewers who extracted

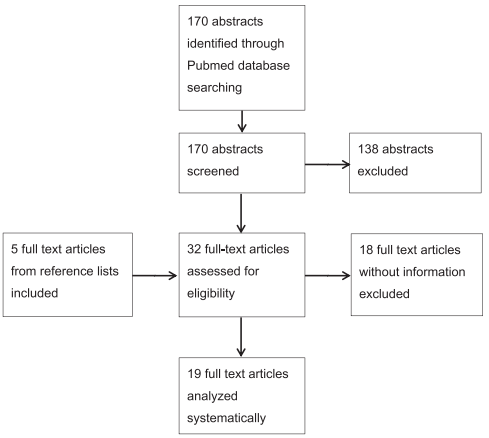

FIGURE 1. Flowchart of the selection process.

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TABLE 1. Previous Reports on the Results of Each Treatment for Angiosarcoma

Procedure	Authors	Year	Site	*n*	Overall Survival (Months)
Surgery	Rosai et al[4]	1976	Scalp	2	3.8
	Hodgkinson et al[5]	1979	Scalp and face	3	14.0
	Maddox et al[6*]	1981	Scalp and face	8	24.0
	Panje et al[7]	1986	Scalp and face	3	47.3
	Holden et al[8]	1987	Scalp and face	7	34.4
	Mark et al[9]	1993	Scalp and face	2	17.5
	Lydiatt et al[10]	1994	Scalp and face	3	34.0
	Buschmann et al[11]	2008	Scalp	16	17.1
	Subtotal			44	23.5 ± 10.3
Surgery + RT	Rosai et al[4]	1976	Scalp and face	1	16.0
	Hodgkinson et al[5]	1979	Scalp and face	2	33.5
	Maddox et al[6]	1981	Scalp and face	2	34.0
	Holden et al[8]	1987	Scalp and face	3	44.0
	Mark et al[9]	1993	Scalp and face	3	37.0
	Buschmann et al[11]	2008	Scalp	1	25.0
	Kohler et al[12]	2008	Scalp and face	13	15.8
	Subtotal			25	25.0 ± 11.0
Surgery + CT	Mark et al[9]	1993	Scalp	1	53.0
	Kohler et al[12]	2008	Scalp and face	2	6.9
	Subtotal			3	22.3 ± 21.7
Surgery + RT + CT	Mark et al[9]	1993	Scalp and face	2	33.5
	Buschmann et al[11]	2008	Scalp	2	14.0
	Kohler et al[12]	2008	Scalp and face	2	9.4
	Subtotal			6	19.0 ± 10.5
Surgery group	Sum			78	23.6 ± 11.0
RT	Rosai et al[4]	1976	Scalp	1	12.0
	Hodgkinson et al[5]	1979	Scalp and face	3	13.3
	Maddox et al[6]	1981	Scalp and face	3	15.0
	Panje et al[7]	1986	Scalp and face	2	7.5
	Holden et al[8]	1987	Scalp and face	45	28.6
	Mark et al[9]	1993	Scalp and face	2	10.0
	Hata et al[13]	2014	Scalp	17	14.0
	Subtotal			73	22.7 ± 7.6
CT	Maddox et al[6]	1981	Scalp and face	3	7.5
	Lydiatt et al[10*]	1994	Scalp and face	9	18.0
	Bacchi et al[14]	2010	Scalp	1	12.0
	Subtotal			13	15.1 ± 4.5
RT + CT	Mark et al[9]	1993	Scalp	4	37.0
	Subtotal			4	37.0 ± 0.0
No surgery group	Sum			90	22.2 ± 8.0
	Total			168	22.9 ± 9.8

*Median survival time, otherwise mean survival.
RT, radiotherapy; CT, chemotherapy; *n*, number of patients.

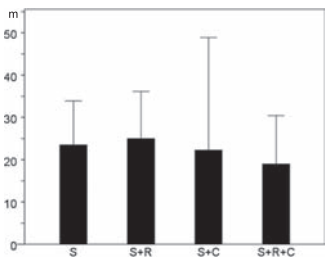

FIGURE 2. Overall survival in the surgery group. m, months; S, surgery; R, radiotherapy; C, chemotherapy.

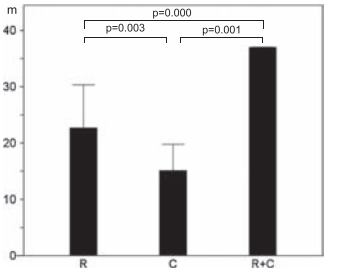

FIGURE 3. Overall survival in the no surgery group.

data from the articles. IBM SPSS Statistics 20 (IBM, Armonk, NY) was used for the statistical analysis.

We analyzed the survival rates according to procedures, tumor size (T1, same or smaller than 5 cm; T2, larger than 5 cm), tumor grade (low grade or high grade), and surgical resection margins (positive or negative).

Patient Groups in Procedures

1. Surgery group (S, surgery only; S + R, surgery and radiation therapy; S + C, surgery and chemotherapy; S + R + C, surgery, radiation, and chemotherapy)
2. No surgery group (R, radiation therapy; C, chemotherapy; R + C, radiation and chemotherapy)

RESULTS

Overall Survival in the Surgery Group and the No Surgery Group

Overall survival varied according to the treatment procedures (ranged from 15.1 to 37.0 months). Overall survival was 22.9 ± 9.8

TABLE 2. Survival by Tumor Size

				T1		T2		
		Year	N	*n*	Months	*n*	Months	*P*
OS	Rosai et al[4]	1976	4	4	8.9			
	Maddox and Evans[6*]	1981	16	10	27	6	8	
	Lydiatt et al[10]	1994	12			12	25	
	Sasaki et al[15*]	2002	30	14	20	16	7	0.007
	Pawlik et al[1*]	2003	29	18	48.7	11	11.1	<0.001
	Ohguri et al[16*]	2004	20	8	28.9	12	26.2	0.910
	Miki et al[17]	2013	17	2	33.7	15	23.5	0.329
	Total		128	56	31.4 ± 13.1	72	17.3 ± 8.3	0.000
LRFS	Maddox and Evans[6]	1981	16	10	19	6	1	
	Ohguri et al[16*]	2004	20	8	10.6	12	12.7	0.720
	Total		36	18	15.3 ± 4.3	18	8.8 ± 5.7	0.000
DMFS	Maddox and Evans[6]	1981	16	10	34	6	2	
	Ohguri et al[16*]	2004	20	8	19.9	12	17.1	0.790
	Total		36	36	27.7 ± 7.2	18	12.1 ± 7.3	0.000
		Year	N	*n*	%	*n*	%	
2YSR	Mark et al[9]	1993	11	11	90.9			
	Ogawa et al[18]	2012	48	22	25.6	26	19.2	0.446
	Total		59	33	47.4 ± 31.3	26	19.2 ± 0.0	0.000
5YSR	Lydiatt et al[10]	1994	18			18	33	
	Guadagnolo et al[19]	2010	60	49	42	11	12	0.002
	Total		78	49	42.0 ± 0.0	29	15.3 ± 3.5	0.000

*Median survival time, otherwise mean survival.
OS, overall survival; 2YSR, 2-year survival rate; N, total number of patients; *n*: number of patients.

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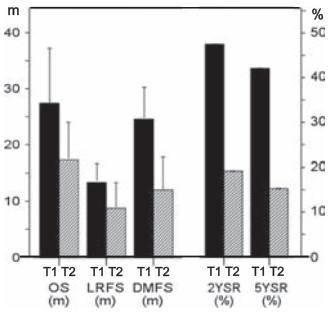

FIGURE 4. Survival by tumor size. OS, overall survival; 2YSR, 2-year survival rate.

months, and there were no significant differences between the surgery group (23.6 ± 11.0 months) and the no surgery group (22.2 ± 8.0 months) ($P = 0.386$ [t test]; Table 1).

Overall Survival in the Surgery Group

Overall survival did not vary significantly among the surgery groups (S, 23.5 ± 10.4 months; S + R, 25.0 ± 11.2 months; S + C: 22.3 ± 26.6 months; S + R + C, 19.0 ± 11.5 months; $P = 0.709$ [analysis of variance]) (Fig. 2).

Overall Survival in the no Surgery Group

Overall survival of the R + C group (37.0 ± 0.0 months) was the longest followed by the R group (22.7 ± 7.6 months). The C group was the shortest (15.1 ± 4.6 months). There were significant differences among the 3 subgroups ($P = 0.000$ [analysis of variance]) (Fig. 3).

Survival by Tumor Size

Overall survival was significantly longer in the T1 group (31.4 ± 13.1 months) than the survival of the T2 group (17.3 ± 8.3 months; $P = 0.000$ [t test]). Local recurrence-free survival (LRFS) was significantly longer in the T1 group (15.3 ± 4.3 months) than the LRFS in the T2 group (8.8 ± 5.7 months; $P = 0.000$, [t test]). Distant metastasis-free survival (DMFS) was also significantly longer in the T1 group (27.7 ± 7.2 months) than the DMFS in the T2 group (12.1 ± 7.3 months; $P = 0.000$ [t test]).

The 2-year survival rate was significantly longer in the T1 group ($47.4 \pm 31.3\%$) than the 2YSF in the T2 group ($19.2 \pm 0.0\%$; $P = 0.000$ [t test]). The 5-year survival rate (5YSR) was significantly longer in the T1 group ($42.0 \pm 0.0\%$) than the 5YSR in the T2 group ($15.3 \pm 3.5\%$; $P = 0.000$ [t test]; Table 2, Fig. 4).

Overall Survival by Tumor Grade

Overall survival was significantly longer in the low-grade group (44.8 ± 10.4 months) than survival in the high-grade group (22.3 ± 6.8 months; $P = 0.000$ [t test]; Table 3).

TABLE 3. Overall Survival by Tumor Grade

			Low		High		
	Year	N	n	Months	n	Months	P
Mark et al[9]	1993	9			9	24.8 (6–34)	
Pawlik et al[1]*	2003	25	6	48.7	19	28.4	0.590
Kohler et al[12]	2008	17	1	21.1	16	13.6 (5.0–83.1)	
	Total	51	7 (13.7%)	44.8 ± 10.4	44 (86.3%)	22.3 ± 6.8	0.000

Data in parentheses below overall survival are survival range.
*Median survival time, otherwise mean survival.

TABLE 4. Survival by Surgical Resection Margin

				Positive			Negative			
	Year	N		n			n			P
Pawlik et al[1]	2003	28	OS	22	26	m	6	NR		0.064
Buschmann et al[11]	2008	19	OS	13	15.8 (5–36)	m	6	20.2 (16–25)	m	0.125
Guadagnolo et al[19]	2010	43	5YSR	23	60	%	20	47	%	0.800
	Total	90		58 (64.4%)			32 (35.6%)			

Data in parentheses below overall survival are survival range.
NR, not reached.

Survival by Surgical Resection Margin

In the 90 patients who underwent surgical resection, only 32 patients (35.6%) had negative pathologic margins at the completion of their surgeries. The remaining patients (58 patients [64.4%]) still had residual cancer cells at their surgical margins.

Survival did not differ significantly according to the positive or negative resection margin ($P > 0.05$ [t tests]). In a study by Pawlik et al,[1] the overall median survival was 26 months in patients with positive margin; $P = 0.064$ [t test]). Buschmann reported that the mean survival of the negative margin was 20.2 months (range, 16–25 months), whereas the mean survival of the positive margins was 15.8 months (range, 5–36 months; ($P = 0.125$ [t test]).[11] Interestingly, Guadagnolo et al[19] wrote that the 5YSR of the patients with positive margin (60%) was higher than that of the patients with negative margin (47%) ($P = 0.800$ [t test]; (Table 4).

DISCUSSION

Since we could not use individual data in analyzing each paper, we calculated the mean and standard deviations through using mean values and the number of patients of each paper. In cases where certain subgroups consisted of only one paper, we could not yield a standard deviation.

Overall survival did not differ significantly between the surgery group (23.6 ± 11.0 months) and the no surgery group (22.2 ± 8.0 months) ($P = 0.386$ [t test]). Among the patients who underwent surgery, 64.4% had residual cancer cells at their surgical margins. Survival did not differ significantly according to positive or negative resection margins ($P > 0.05$ [t tests]). From these result, we could say that the surgeons do not have to try to remove all the cancer cells unless it causes severe ulcers or bleedings.

Overall survival of the R + C group (37.0 ± 0.0 months) was significantly longer than the survival of the R group (22.7 ± 7.6 months) or C group (15.1 ± 4.6 months). In inoperable cases, we should remember that combinations of radiation and chemotherapy can bring better results than radiation alone or chemotherapy alone.

Overall survival, LRFS, and DMFS were significantly longer in the T1 group than for those variables in the T2 group ($P < 0.05$ [t test]). The 2-year survival rate and 5YSR were significantly longer in the T1 group than for those variables in the T2 group ($P < 0.000$ [t test]). Overall survival of the low-grade tumor group (44.8 ± 10.4 months) was more than 2 times longer than the survival of the high-grade tumor (22.3 ± 6.8 months; $P = 0.000$ [t test]). At this point, we have to emphasize that early diagnosis and early treatment affect prognosis significantly.

In summary, surgeons should remember that they do not have to try to remove all the cancer cells in the operation theater. Combinations of radiation and chemotherapy can bring better results than

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any single regimen. Lastly, early diagnosis and early treatment are essential.

REFERENCES

1. Pawlik TM, Paulino AF, McGinn CJ, et al. Cutaneous angiosarcoma of the scalp: a multidisciplinary approach. *Cancer* 2003;98:1716–1726
2. Morrison WH, Byers RM, Garden AS, et al. Cutaneous angiosarcoma of the head and neck. A therapeutic dilemma. *Cancer* 1995;76:319–327
3. Lim SY, Pyon JK, Mun GH, et al. Surgical treatment of angiosarcoma of the scalp with superficial parotidectomy. *Ann Plast Surg* 2010;64:180–182
4. Rosai J, Sumner HW, Major MC, et al. Angiosarcoma of the skin: a clinicopathologic and fine structural study. *Hum Pathol* 1976;7:83–109
5. Hodgkinson DJ, Soule EH, Woods JE. Cutaneous angiosarcoma of the head and neck. *Cancer* 1979;44:1106–1113
6. Maddox JC, Evans HL. Angiosarcoma of skin and soft tissue: a study of forty-four cases. *Cancer* 1981;48:1907–1921
7. Panje WR, Moran WJ, Bostwick DG, et al. Angiosarcoma of the head and neck: review of 11 cases. *Laryngoscope* 1986;96:1381–1384
8. Holden CA, Spittle MF, Jones EW. Angiosarcoma of the face and scalp, prognosis and treatment. *Cancer* 1987;59:1046–1057
9. Mark RJ, Tran LM, Sercarz J, et al. Angiosarcoma of the head and neck: the UCLA experience 1955 through 1990. *Arch Otolaryngol Head Neck Surg* 1993;119:973–978
10. Lydiatt WM, Shaha AR, Shah JP. Angiosarcoma of the head and neck. *Am J Surg* 1994;168:451–454
11. Buschmann A, Lehnhardt M, Toman N, et al. Surgical treatment of angiosarcoma of the scalp: less is more. *Ann Plast Surg* 2008;61:399–403
12. Köhler HF, Neves RI, Brechtbühl ER, et al. Cutaneous angiosarcoma of the head and neck: report of 23 cases from a single institution. *Otolaryngol Head Neck Surg* 2008;139:519–524
13. Hata M, Wada H, Ogino I, et al. Radiation therapy for angiosarcoma of the scalp: treatment outcomes of total scalp irradiation with X-rays and electrons. *Strahlenther Onkol* 2014;190:899–904
14. Bacchi CE, Silva TR, Zambrano E, et al. Epithelioid angiosarcoma of the skin: a study of 18 cases with emphasis on its clinicopathologic spectrum and unusual morphologic features. *Am J Surg Pathol* 2010;34:1334–1343
15. Sasaki R, Soejima T, Kishi K, et al. Angiosarcoma treated with radiotherapy: impact of tumor type and size on outcome. *Int J Radiat Oncol Biol Phys* 2002;52:1032–1040
16. Ohguri T, Imada H, Nomoto S, et al. Angiosarcoma of the scalp treated with curative radiotherapy plus recombinant interleukin-2 immunotherapy. *Int J Radiat Oncol Biol Phys* 2005;61:1446–1453
17. Miki Y, Tada T, Kamo R, et al. Single institutional experience of the treatment of angiosarcoma of the face and scalp. *Br J Radiol* 2013;86:20130439
18. Ogawa K, Takahashi K, Asato Y, et al. Treatment and prognosis of angiosarcoma of the scalp and face: a retrospective analysis of 48 patients. *Br J Radiol* 2012;85:e1127–e1133
19. Guadagnolo BA, Zagars GK, Araujo D, et al. Outcomes after definitive treatment for cutaneous angiosarcoma of the face and scalp. *Head Neck* 2011;33:661–667

Extraoral Surgical Approach of Ectopic Mandibular Third Molar to the Lower Border of Mandible

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Abstract: The surgical removal of impacted lower third molar is an ordinary intervention. The treatment of choice in this pathology is an intraoral or, seldom, extraoral surgical approach. Various surgical procedures have been described to remove ectopic mandibular teeth. The more common technique is an intraoral approach (so named ''conservative''), even when the tooth is located in an ectopic area. However, the ''intraoral approach'' is often related with the difficulty of view, the bleeding of the surgical site, and with the possible lesions of inferior alveolar or lingual nerve. From the other side, a nonconservative surgical approach like ''extraoral pathway'' may be associated with no aesthetic cutaneous postoperative scar tissue. The aim of those 2 study cases is to highlight the management of infected ectopic third molars located close to the lower border of the mandibular body, underlining the anatomical land markers of the submandibular area. The authors have applied the extraoral pathway using an incision of small size. From our analysis, the treatment of those typical can be managed by using a ''mini-skin-incision'' (so termed as mini-submandibular approach) respecting the cosmetic expectations of the patients.

Key Words: Impacted lower third molar, extraoral surgery, submandibular access

Impacted (unerupted and partially erupted) lower third molars show a high incidence in 20% to 30% of the population with gender prevalence in females.[1–4] Often an incidental orthopantomography x-ray (OPT) notices this condition rather than the evidence of clinical symptoms, and a large part of unerupted and asymptomatic third molar teeth may be left in situ (so as to be defined ''no-surgery-treatment cases''). As a rule, those teeth are often associated with different diseases such as facial swelling, acute pericoronitis, chronic infection, and local inflammatory mass.[5] Moreover, the presence of impacted lower third molar may be connected with periodontal disease of the next second molar and with various associated types of cysts and odontogenic tumors (particularly ameloblastomas and keratocysts). Finally, the infection of the area surrounding the impacted teeth can lead to neurogenic disorders such as pain and paresthesia.[2,3,4,6,7]

The ectopic placement of third molars is a condition in which the tooth is located in a non-physiologic area.[1–4,6,7] It is relatively rare and often a dentigerous cyst may be associated with it. This represents the most frequent cause of dislocation of the third molar. The presence of a related tumor usually drives the tooth to an ectopic location (secondary ectopy). Otherwise, if the cause of the dislocation of the tooth cannot be deduced, the diagnosis may be defined ''primary ectopy'' (pure ectopy).[8,9]

The reason related to the third molar primary ectopy is still unknown. However, trauma, aberrant eruption, and anomalous development of the tooth germ are conditions that seem to influence it.[8,10]

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Clinicians can manage this atypical condition by following 2 basic therapeutic options: if there are no symptoms or complications, it could be considered a no surgical treatment.[1,10–12] In the cases characterized by symptoms, associated pathology, or also when the patient is requiring tooth removal, the treatment of choice is surgery by using intra- or extraoral approach.[2,13,14] An x-ray OPT investigation is generally sufficient for making the diagnosis of an ectopic third molar.[3,6] However, only the clinical examination allows evaluating symptoms like pain, trismus, the hypomobility of the temporomandibular joint,[15] and the strength of the bone that includes the third molar. Therefore, x-ray computed tomography (x-ray CT) scans or software Dentascan images are needed to determine a precise 3-dimensional anatomical position of wisdom tooth and its relation to surrounding anatomical structures.

Other criteria for the lower third molar extraction are prophylactic removal after orthodontic, prosthodontic, or restorative indications.[16] Third molar removal is a frequent procedure in oral surgery and the lower third molar extraction is still the most common.[2,17,18] It is usually performed by intraoral approach depending on their position, based on the direction of the crown of the tooth, often buccally or, more rarely, lingually. The surgical buccal approach is the most common and it is based on the possibility of avoiding surgical damage of the lingual nerve.[19,20] The lingual approach is indicated when the crown of the tooth is in lingual version or pushed to the lingual side; special care is requested in those cases to protect the lingual nerve.[19,20] In some particular cases, such as for a double molar inclusion, only the surgical intraoral approach is suggested; in fact, the surgical removal of 2 adjacent teeth involves a great loss of bone. Intraoral surgery is able to maintain the continuity of the mandibular cortical bone, and its thickness provides adequate bone healing.[21] Through a careful and complete surgical planning, it is also possible to determine the placement of maxilla-mandibular fixation using specific surgical titanium plates or miniplates to prevent postoperative fractures.[6]

Impacted teeth are recorded according to the Pell, Gregory, and Winter classification.[2,3,6,18] A new classification has been proposed after the data from the examination of x-ray OPT relative to the position of impacted third lower molar and considering the ratio between the roots and the mandibular canal (third-molar classification: TMC operative). This classification records 3 positions: the roots are above the bony canal, the roots are directly projected on the mandibular canal, or the roots are located immediately below it and the third molar is placed below the bony canal. This classification could represent a guide for the surgeon's decision-making, and it is useful to show the patient the real difficulty and the risks associated with the operation.

The inferior alveolar nerve is located in the mandibular canal and it usually represents an important anatomical limit that may strongly influence the success of the surgery. The mandibular canal is buccally placed in 60% of the cases; it has a lingual position in the other 40%.[3,9] Relating to the ectopic third lower impacted molars, the Scientific Literature Community underlined less than 30 cases until 2013.[1,12,22] These reports do not include the cases in which the third molar is located to the lower border of the mandible.[1,6,7,23] Various surgical approaches have been described to remove ectopic mandibular teeth[1,10]: intraoral,[1–4,13,20,24] particularly by using a sagittal split osteotomy (SSO[6,25]), and extraoral, more recently including endoscopic[1,3,23,26–28] preauricular-endaural, retromandibular, and submandibular surgery [6,9,29]

MATERIALS AND METHODS

The surgery involved the mandibular body and ramus by using an extraoral surgical access[14,15,29] that may be performed through retromandibular or submandibular way. The first one is considered the best access to approach subcondylar pathologies as fractures or tumors, although it has also been proposed to achieve pathologies of the mandibular body.[30] The incision of the skin begins approximately 1 cm below the lobe of the ear and 1 cm posterior to the mandible ramus. During the opening flap, the parotid gland and its anatomical content above all the facial nerve and its neural branches have to be considered. Even if this skin incision is more aesthetic, the surgical act is more complex.[15,29] The extraoral surgical access can be performed by the mandibular body and angle, and those regions are considered unsuitable by the intraoral approach.[15,29] The extraoral one provides optimal vision of the inferior mandible border and a good view of the underside of the mandible lingual cortex. Moreover, it consents to achieve surgical targets as mandibular ramus, angle, and subcondylar region.[23] The structures that must be saved during this dissection are the marginal mandibular branch of the facial nerve (CN VII) and the facial artery and vein. The unintentional injury of the facial nerve is manifested by a paralysis of several muscles like the depressor anguli oris, the depressor labii inferioris, the mentalis, and the orbicularis oris.[30–32] When the facial nerve is injured, it implies a typical unilateral pathological facial expression, almost like a faint smile on opening the mouth[33,34]; a temporary injury can give neuropraxia which usually ends after a few weeks. The correction of a permanent deformity needs complicated surgical interventions such as the use of the anterior body of the digastric muscle or the extensor of the digitorum brevis that can be repositioned for compensating the action of the paretic muscle.[31]

The marginal mandibular nerve departs from the nervous cervical branch and moves towards the mandibular body; it can be composed of a single ramus in a minority of cases (about 35%[35]), but usually it is constituted by 2 (about 65%[32,35]) or, seldom, more ramifications.[31,32] Its extraparotid route starts from the front edge of the parotid capsule (about 90%[29–31]), and the anatomic landmark is the masseter tuberosity.[34] It passes near the mandible angle about 1.5 cm below the lower margin[31](for this reason, the skin incision in this area must be placed at least 2 cm below the mandibular border). The nerve branch lies on the outer surface of the masseter muscle below the superficial musculo-aponeurotic system (in a minority of cases, about 30%, it can run below the inferior border) and then it crosses the "facial vessels" (an important anatomic landmark for searching marginal mandibular nerve[31,32]) passing above them or, sometimes, between them with the vein superficially and artery internally placed. Then, it becomes superficial (approximately 3 cm anterior to the masseteric tuberosity and about 2 cm to the corner of the mouth[34]) and penetrates the muscle depressor of the mouth. Finally, it disappears from the surgical field. The fiber of the platysma is intermingled with the fiber of the "depressor labii inferioris" at the corner of the mouth. A lesion of the platysma could be misinterpreted as a loss function of the depressor labii inferioris.[31] The projection on the skin of the middle area of the mandibular body is an area defined as "the danger zone" (2 cm posterior to the oral commissure).[31,34]

At first, particular care should be given to the presence of the jugular vein running just around the angle of the mandible. If the patient is undergoing general anesthesia, by infiltrating a physiological solution with vasoconstrictor, a hemostatic effect is obtained and the skin incision can be made 2 to 3 cm below the inferior border of the mandible (conventionally under the distance of one finger breadth). The incision could be normally 4 to 5 cm in length[6]; it can be extended in either direction in cases of inadequate exposure or, for aesthetic reason, it can be extended posteriorly and curved with the angle of the mandible (Risdon's procedure).[15]

The search of the "skin crease" is important to prevent postoperative scarring, but in adolescents it is difficult due to the

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elasticity of the subcutaneous tissue; in such situations, reference is made to the Langer lines.[15]

Eighty percent of the examined cases[36] showed the facial nerve (CN VII: which has a thickness of approximately 1 mm[30]) passing up to the lower border of the posterior portion of the mandibular body with a small branch that connects this branch to the main trunk of the CN VII. Therefore, an anastomosis with the mental nerve commonly occurs.[31] The marginal branch runs below the platysma muscle resting on the fascia of the masseter muscle (superficial musculo-aponeurotic system). Then it reaches the facial artery and vein from the antero-inferior angle of masseter and it penetrates the buccal region[35] emitting branches to mimic muscles of the lip; some neural branches run below the mandible innervating the platysma and not the depressors of the lower lip. The remaining ~20% of cases underlined how the marginal branch forms a loop downwardly convex, with a lowest point placed 1 cm below the lower mandibular border[36]; submandibular lymph nodes can be found right posteriorly to the artery (they are a constant anatomic landmark for searching the marginal mandibular nerve). When the "surgical act" regards pathologic conditions whose extension is not predictable, the facial vessels in its entirety can be achieved by ligature avoiding their surgical amputation; in this situation, the marginal branch is included and protected in the superior flap.[15]

As soon as the skin and subcutaneous tissues incision is made, clamping controls the bleeding of small vessels. The next surgical step is tissue dissection to expose the deep layers: the cervical fascia and platysma muscle. Guided by an undermined surgical protector to avoid cutting the marginal mandibular branch of the nerve, the fascia and muscle are divided with a scalpel, usually 2 to 3 cm below the mandibular border, not at the same level of the skin incision. It is advisable to protect the vascular bundle using the traction of a Langbeck retractor distally (for protecting jugular vessels) or proximally placed (for protecting facial vessels). The exact position of the surgical cut is related with the location of the tooth to be removed. Underneath this layer, the surgeon should be able to catch a glimpse of the facial vessels crossed by the marginal mandibular nerve. After reaching the muscular plane of the pterygomasseteric sling, the periosteum is emphasized by cutting those structures. Then, the periosteum is incised with a scalpel to the lower mandibular border to expose the pathological site at the level of the mandibular body and angle.

The aim of this paper is to present cases successfully treated by "extraoral submandibular approach" presented with an incision reduced in length, "extraoral submandibular reduced". The operations were performed under local anesthesia; the skin incision was planned and executed in length reduced to approximately 2 cm if compared to those reported in literature (4–5 cm). The deep layers crossed in the first study case have involved the masseter muscle and the superficial musculo-aponeurotic system while in the other one the cervical fascia and the platysma muscle only. In both cases, complete odontectomy was made with surgical revision without performing the ligation of the facial vessels. The suture of deep tissue was performed in several layers. The skin was sutured by using intradermal technique; lastly, a tubular drain was inserted.

A 48-year-old male patient presented with swelling of the right cheek and difficulty opening the mouth with a purulent drainage on the left side of the face (Fig. 1). Facial asymmetry was noted; no paresthesia or nervous alterations of the branches of the facial nerve was related. The patient presented an x-ray OPT (Fig. 2) which showed an ectopic impacted lower right third molar located in the mandibular body below the mandibular canal. The tooth was placed next to the lower border of the mandible overcoming it and having a direct contact with the soft subcutaneous tissue and muscle layers. In the bone, a radiolucency area resembling a cyst crowned it. A preoperative CT examination was performed (Fig. 3). The treatment

FIGURE 1. Clinical evaluation of the patient showing a fistula localized in the lower left mandible border.

FIGURE 2. Orthopantomographic x-ray evaluation underlined the lower left third molar localized in an uncommon anatomical position.

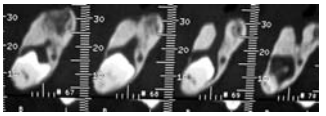

FIGURE 3. Computed tomography x-ray evaluation is requested to evaluate the anatomical limit of the tooth localized in the left mandible border.

proposed to the patient was an extraoral approach: by mini-submandibular access in conjunction with concomitant removal of the skin fistulous area; the mini-access was planned in relation to the extension of inflamed skin area. The patient undersigned an informed consent and an extraoral surgical access was chosen. Bacterial contamination was not regarded as a contraindication.

The pre-existing soft-tissue fistulous area was utilized as a way to access the mandible. A skin incision of about 2 cm in length was performed as follows: an upper cutaway 2 cm from the lower edge of the mandible was carried out along the upper perimeter of the inflamed skin area; then, a second incision was made to circumscribe that lower perimeter, and the 2 incisions were joined together. The skin area was removed. The platysma muscle was identified and incised through this incision and by using the traction of a Langbeck retractor situated in proximal position of the surgical cut (protecting the facial vessels), the periosteum was reflected to exhibit the tooth and the cortical bone of the lower border of the mandible. Through an easy osteotomy around the impacted tooth by utilizing a bone-drill handpiece, it was removed (Figs. 4, 5). The coronal soft tissue was enucleated. After the irrigation with antibiotic solution, the

FIGURE 4. Surgical extraoral approach involving the tooth crown. A flap is opened to perform the tooth extraction.

FIGURE 5. The entire tooth is removed and no bleeding is presented at the time of the surgery.

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FIGURE 6. Orthopantomographic x-ray evaluation showing the lower left third molar localized in an uncommon anatomical position.

periosteum and the muscle bundles were sutured in layers. Before suturing, an extraoral drain was inserted for the evacuation of secretions and blood and for irrigating the area with antibiotic solution during the postoperative period. The skin was sutured using nylon for maximum cosmetic benefit. Because of an established and heavy infection, the patient was treated with amoxicillin for 10 days. The patient reported no paresthesia.

A 59-year-old female patient presented an x-ray OPT that showed an impacted third molar at the lower border of the mandible on the right side of the face. She reported only a continuous pain due to the presence of an indolent mass that seemed to swell occasionally on the same side of the impacted tooth. Oral examination of the patient underlined how the lower left third molar was unerupted. Radiographic examination (Fig. 6) showed a deeply impacted third molar immediately located to the inferior dental nerve and the lower border of the mandible which was situated beyond the mandibular cortex contracting relationships with the soft tissues (with the root portion almost suspended in soft tissues); no relationship with the neurovascular bundle was recorded. In the bone, it was possible to notice a radiolucent area resembling a cyst associated with the crown of the tooth.

The patient undersigned an informed consent and it was planned to remove the tooth under local anesthesia through an extraoral approach. By using a skin marker, an incision line was scored 2 cm below the lower mandible border, between the angle and 3 cm before the masseteric tuberosity of the masseter muscle. The skin incision was about 2 cm length in total. A dissection was carried out down to the platysma muscle. The muscle was carefully bisected using blunt scissors, and the cervical fascia was carefully cut avoiding the marginal branch of the facial nerve until the superficial surface of the masseter muscle was exposed. The masseter muscle was then horizontally incised just above the lower mandible border and dissection was carried out to the periosteum to expose the mandible; an adequate exposure for odontectomy was achieved (Fig. 7). The extraction of impacted tooth was successfully performed (Fig. 8). The coronal soft tissue was enucleated. After the irrigation with antibiotic solution, the periosteum and the muscle bundles were sutured in layers. Before suturing, an extraoral drain was inserted for the evacuation of secretions and blood and for irrigating the area with antibiotic solution during the postoperative period. The skin was sutured by intradermal technique using nylon thread for maximum cosmetic benefit (Figs. 9, 10). The patient reported no paresthesia.

RESULTS

In both cases of our study, an incision reduced in length seems to give excellent aesthetic results. Moreover, it allows the achievement of the ectopic tooth. Odontectomy was successfully performed in both treated cases. The suture was removed after 10 days and the healing was regular. By applying an intradermal suture, aesthetic result required through time was obtained.

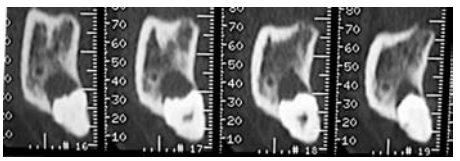

FIGURE 7. Computed tomography x-ray evaluation is requested to evaluate the anatomical limit of the tooth localized in the right mandible border.

FIGURE 8. The flap design is made accordingly to the anatomical vessels and nerve to control the bleeding during the surgery.

FIGURE 9. After the tooth removal, cleaning of the infected area is a fundamental condition to have a good healing after the operation.

FIGURE 10. Closure of the flap. A particular vision of the drainage medical device used for reducing the postoperative swelling.

DISCUSSION

The surgical technique for the removal of impacted lower third molar is usually associated with a more conservative intraoral approach even if the tooth is located in an ectopic area. This reason is due to the possibility of avoiding (using an extraoral approach) facial nerve cervical and marginal branch (CN VII) damage and preventing a cutaneous postoperative bad scarring. However, in some cases, the intraoral approach is inadequate because the surgical target lacks visualization and surgical field is limited. Moreover, sometimes the tooth is placed in an inaccessible region such as the angle and lower border of the mandible. Cases of severe restriction of jaw opening and cases of serious trismus can result in a next difficult surgery. In case of the third molar placed in a deep region such as in the condyle, coronoid, angle, and lower border of the mandible, the extraoral approach can be a valid therapeutic option for managing the uncommon tooth position. Often it ensures a reduced loss of mandibular bone by using the drill because the target is more close to the surgical act so as to define it "more efficient" as in our cases. The most used extra oral approach for mandible surgery is associated with retromandibular or submandibular pathway. In particular, the second one seems to be particularly efficient if it is well managed by the surgeon in the planning phase. In fact, it is possible to operate both on the edge of the mandible and on the lingual side, which reach the angle and the branch, sometimes even without applying the phase of surgical ligation and surgical resection of

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facial vessels. Mainly in case of an operation that requires a large surgical field, such as a tumor or fracture, this phase should be put in place. Thus, for the surgery of ectopic teeth placed at the lower border of the mandible, a ''mini-skin incision'' can be made distally or proximally to the facial vessels protecting those anatomical structures with an appropriate surgical instrument (like a Langbeck retractor). About 2 cm below and following the lower border of the mandible, a 2-cm-length incision can be made (mini-submandibular incision); in this way, the muscle layers are overcome by the surgical incision avoiding the anatomical structures that must be saved and the surgical field is reached. With particular care, it is possible to remove enough bone around the tooth to release and remove it. The cosmetic squeal of the skin scar can be avoided by using an intradermal aesthetic suture. Because of the diffusion of cosmetic surgery, a precise incision on the skin with an excellent healing is what the patients are requiring. Hence, the skin incisions must be made according to the basic anatomical knowledge. The surgical approach to ectopic teeth must provide a preoperative planning in which, after assessing the negative and positive factors of the case in clinical examination and the related operation, the oral surgeon will expose the patient to the possibility of success and possible negative contingencies. In the case of ectopic third molars located close to the lower border of the mandibular body, knowing the anatomical landmarks of the submandibular area, it is possible to use a small-size incision. It allows the oral surgeon to reach easily the pathological target and to complete the operation successfully and with minimal chance of injuring the anatomical structures that must be saved. Especially in the case of an ectopic impacted lower third molar located in the mandibular border, the partial localization beyond the bone and below the lower edge of the mandible constitutes a serious justification to induce the choice of the route of surgical extraoral submandibular approach by using a mini-skin incision (mini-submandibular pathway).

REFERENCES

1. Ahmed NM, Speculand B. Removal of ectopic mandibular third molar teeth: literature review and a report of three cases. *Oral Surg* 2012;5: 39–44
2. Archer WH. Oral and Maxillofacial Surgery, 5th ed. Philadelphia: WB Saunders Company; 1975
3. Momen AA. Diagnostic accuracy of panoramic radiography in determining relationship between inferior alveolar nerve and mandibular third molar. *J Oral Maxillofac Surg* 2010;68:74–82
4. Torres AMF, Albiol JG, Aytes LB, et al. Evaluation of the indication for surgical extraction of third molars according to the oral surgeon and the primary care dentist. *Med Oral Patol Oral Cir Bucal* 2008;13:499–504
5. Scott N, Bater M, Wilson A. An unusual cause of a facial swelling. *J Surg Case Rep* 2010;10:10–13
6. Naaj IA, Braun R, Leiser Y, et al. Surgical approach to impacted mandibular third molars—operative classification. *J Oral Maxillofac Surg* 2010;68:628–633
7. McCrea S. Adjacent dentigerous cysts with the ectopic displacement of a third mandibular molar and supernumerary (fourth) molar: a rare occurrence. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2009;107:15–20
8. Kaur M, Shefali S. Molar impactions: etiology, implications and treatment modalities with presentation of an unusual case. *J Orofac Res* 2012;2:171–173
9. Salmeron JI, Del Amo J, Plasencia R, et al. Ectopic third molar in condylar region. *Int J Oral Maxillofac Surg* 2008;37:398–400
10. Wang C, Kok S, Hou L, et al. Ectopic mandibular third molar in the ramus region: report of a case and literature review. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2008;105:155–161
11. Hill CM, Walker RV. Conservative, non surgical management of patients presenting with impacted third molars: a 5-year study. *Br J Oral Maxillofac Surg* 2006;44:347–350
12. Kupferman SB, Schwartz HC. Malposed teeth in the pterygomandibular space: report of 2 cases. *J Oral Maxillofac Surg* 2008;66:167–169
13. Singh V, Alex K, Pradhan R, et al. Techniques in the removal of impacted mandibular third molar. *Eur J Gen Dent* 2013;2:25–30
14. Rivera JC, Estrada HI, de Jesús Ibarra González F. Tercer molar ectópico en región condilar. Presentación de caso clínico y revisión bibliográfica. *Rev Mex Cir Bucal Max* 2013;9:32–35
15. Patnaik VVG, Singla RK, Sanjus B. Surgical incisions—their anatomical basis: part 1—head and neck. *J Anat Soc India* 2000;49: 40–42
16. Adeyemo WL. Do pathologies associated with impacted lower third molars justify prophylactic removal? A critical review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2006;102:448–452
17. Almendros-Marqués N, Berini-Aytés L, Gay-Escoda C. Evaluation of intraexaminer and interexaminer agreement on classifying lower third molars according to the systems of Pell and Gregory and of Winter. *J Oral Maxillofac Surg* 2008;66:893–899
18. Juodzbalys G, Daugela P. Mandibular third molar impaction: review of literature and a proposal of a classification. *J Oral Maxillofac Res* 2013;2:1–12
19. Jerjes W, Upile T, Shah P, et al. Risk factors associated with injury to the inferior alveolar and lingual nerves following third molar surgery—revisited. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2010;109:335–345
20. Pippi R, Perfetti G. Lingual displacement of an entire lower third molar. Report of a case with suggestions for prevention and management. *Minerva Stomatol* 2002;51:263–268
21. Boffano P, Gallesio C, Bianchi F, et al. Surgical extraction of deeply horizontally impacted mandibular second and third molars. *J Craniofac Surg* 2010;21:403–406
22. Wong YK, Liow J, Tsui S, et al. Ectopic molar near the coronoid process: case report. *Quintessence Int* 2007;38:597–600
23. Milner N, Baker A. Extraoral removal of a lower third molar tooth. *Br Dent J* 2005;199:345–346
24. Gadre KS, Waknis P. Intra-oral removal of ectopic third molar in the mandibular condyle. *Int J Oral Maxillofac Surg* 2010;39:294–296
25. Jones TA, Garg T, Monaghan A. Removal of a deeply impacted mandibular third molar through sagittal split ramus osteotomy approach. *Br J Oral Maxillofac Surg* 2004;42:365–368
26. Suarez-Cunquiero MM, Schoen A, Schramm A, et al. Endoscopic approach to removal of an ectopic mandibular third molar. *Br J Oral Maxillofac Surg* 2003;41:340–342
27. Pace C, Holt D, Payne M. An unusual presentation of an ectopic third molar in the condylar region. *Aust Dent J* 2010;55:325–327
28. Bortoluzzi MC, Manfro R. Treatment for ectopic third molar in the subcondylar region planned with cone beam computed tomography: a case report. *J Oral Maxillofac Surg* 2010;68:870–872
29. Manisali M, Aghabeigi B, Newman L. Retromandibular approach to the mandibular condyle: a clinical and cadaveric study. *Int J Oral Maxillofac Surg* 2003;32:253–260
30. Sicher H. Oral Anatomy, 4th ed. Saint Louis: CV Mosby Co; 1965
31. Batra AP, Mahajan A, Gupta K. Marginal mandibular branch of the facial nerve: an anatomical study. *Indian J Plast Surg* 2010;43: 60–64
32. Balagopal PG, George NA, Sebastian P. Anatomic variations of the marginal mandibular nerve. *Indian J Surg Oncol* 2012;3:8–11
33. Larrabiee FW, Makielski KH, Henderson JL. Surgical Anatomy of the Face, 2nd ed. Philadelphia: Lippincott Williams & Wilkins; 2004
34. Karapinar U, Kilic C, Cetin B, et al. The course of the marginal mandibular branch of the facial nerve in adult cadavers. An anatomic study. *Saudi Med J* 2013;34:364–368
35. Hazani R, Chowdhry S, Mowlavi A, et al. Bony anatomic landmarks to avoid injury to the marginal mandibular nerve. *Aesth Surg J* 2011;31:286–289
36. Dingman RO, Grabb WC. Surgical anatomy of the mandibular ramus of the facial nerve based on the dissection of 100 facial halves. *Plast Reconstr Surg* 1962;29:266–272

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Unilateral Mandibular Advancement With Bilateral Intraoral Vertical Ramus Osteotomy

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Abstract: Intraoral vertical ramus osteotomy (IVRO) is an effective surgical procedure that is used for the correction of mandibular prognathism. However, application of IVRO for mandibular advancement has been limited because of the instability of the proximal segments caused by the gap between the distal and proximal segments. We report a case of unilateral mandibular advancement with bilateral IVRO for the correction of facial asymmetry. This case shows possible application of bilateral IVRO for unilateral mandibular advancement without any means of fixation.

Key Words: Unilateral mandibular advancement, facial asymmetry, intraoral vertical ramus osteotomy

Intraoral vertical ramus osteotomy (IVRO) is known as an effective surgical procedure for the correction of mandibular prognathism. Bone healing is known to occur successfully by cortical overlapping of the distal and proximal segments without any means of fixation.[1] However, IVRO has not been preferred for the treatment of mandibular retrognathism because of the high risk of surgical relapse.[2] We report a case of unilateral advancement of the mandible with bilateral IVRO with successful result.

CLINICAL REPORT

This study followed the Declaration of Helsinki on medical protocol and ethics, and the regional Ethical Review Board of Yonsei Dental Hospital approved the study (institutional review board no. 2-2011-0040).

An 18-year-old man was referred from the Department of Orthodontics for correction of facial asymmetry. The patient presented severe chin deviation to the left side with clicking and pain on the left temporomandibular joint (TMJ). The panoramic radiograph showed shorter length of the left condyle compared with that of the right (Fig. 1).

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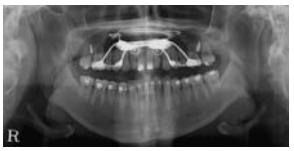

FIGURE 1. Preoperative panoramic radiograph showing facial asymmetry with different length of the condyles.

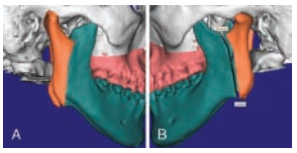

FIGURE 2. Three-dimensional virtual surgical simulation showing (A) an estimated setback of the distal segment on the right side with IVRO, (B) an estimated advancement of the distal segment on the left side with IVRO.

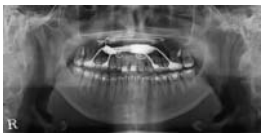

FIGURE 3. Panoramic radiograph taken at postoperative 2 weeks, showing a bony gap between the distal and proximal segments on the left side.

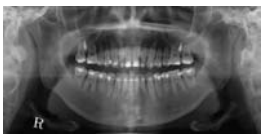

FIGURE 4. Panoramic radiograph taken at postoperative 12 months, demonstrating an evidence of bone regeneration in the gap between the segments.

Three-dimensional virtual surgical simulation was conducted with the reconstructed computed tomography scan images using Mimics software (Materialise Dental, Leuven, Belgium). Simulation showed an estimated setback of the distal segment on the right side, but advancement on the left side after horizontal rotational movement after bilateral IVRO (Fig. 2).

The patient underwent orthognathic surgery including Le Fort I osteotomy for maxillary cant correction and bilateral IVRO. No fixation was followed after IVRO. Maxillomandibular fixation (MMF) with stainless steel wires (0.15 mm) was maintained for 14 days immediately after the surgery. Postoperative panoramic radiograph taken on the day of MMF removal showed bone overlapping on the right side, but a gap between the segments on the left side (Fig. 3).

After the release of MMF, active physiotherapy was conducted as recommended for general IVRO[1] until postoperative 1 month. Postoperative periodic follow-up was done on 3, 6, and 12 months. Panoramic radiograph taken at 12 months postoperatively verified an evidence of complete bone healing in the gap between the segments (Fig. 4). Clicking and pain on the left TMJ were completely resolved after the operation.

Postoperative skeletal stability was evaluated by superimposition of the lateral cephalograms taken at postoperative 2 weeks and 3, 6, and 12 months. There was no difference in the anterior-posterior position of pogonion until after postoperative 12 months.

DISCUSSION

Intraoral vertical ramus osteotomy is a useful technique that can be used for the correction of mandibular prognathism. This

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procedure offers several advantages over sagittal split ramus osteotomy (SSRO), including shorter surgical time because of the technical simplicity and lower incidence of inferior alveolar nerve damage.[3] Also, IVRO is reported to be effective in improving TMJ symptoms.[4–7]

Conventional IVRO entails lateral overlapping of the proximal segments.[1] Recently, Arimoto et al[8] reported the evidence of bone healing in the cleavage space between the bone segments after distal and medial positioning of the proximal segment. However, application of IVRO for mandibular advancement has not been preferred because of the instability of the bone segments and high risk of postoperative skeletal relapse.[2] This instability is caused by the severe tension of the extended soft tissue envelope and the suprahyoid muscle group exerted on the advanced distal segment[9] during and after the physiotherapy.

Correction of asymmetric mandibular prognathism or facial asymmetry involves horizontal rotational movement of the distal segment toward the nondeviated side. Such rotation of the distal segment would occasionally result in an advancement of the distal segment on the deviated side after bilateral IVRO depending on the amount of the rotational movement. For advancement of the distal segment, SSRO with rigid fixation would provide wide surface contact with the proximal segment. However, application of SSRO after the horizontal rotational movement of the distal segment is often limited by the large bony interference with the proximal segments on the deviated side.[10]

Despite the estimated unilateral advancement of the distal segment, bilateral IVRO was implemented for the treatment of preexisting TMJ symptoms. Fixation had not been performed in order to promote physiologic repositioning of the condyle on the advancement side. After the surgery, counterclockwise rotation of the proximal segment was noted on the advancement side, making bone contact with the advanced distal segment at the inferior aspect with the distal segment. This phenomenon probably resulted from the anterior pulling action exerted by the pterygomasseteric sling. Because of the counterclockwise rotation of the proximal segment on the left side, amount of gap between the segments was smaller than had been expected from the three-dimensional virtual simulation.

Bone healing was evident in the gap between the segments at postoperative 12 months. Also, left TMJ symptoms were resolved after the operation. Skeletal stability was maintained until after postoperative 12 months.

This case shows that bilateral IVRO can be considered as a surgical strategy for unilateral mandibular advancement without any means of fixation, as well as for improving TMJ symptoms. Further investigation is required to determine possible amount of advancement and verify the quality of the healed bone in the gap.

REFERENCES

1. Fonseca RJ, Marciani RD, Turvey TA, et al. Oral and Maxillofacial Surgery. Vol. 3. Orthognathic Surgery, Esthetic Surgery, Cleft and Craniofacial Surgery. St Louis, MO: Saunders Elsevier; 2009
2. Hara S, Mitsugi M, Kanno T, et al. Clinical approach for mandibular advancement by intraoral vertical ramus osteotomy with endoscopically assisted intraoral fixation of an L-shaped compact lock plate. *J Craniofac Surg* 2013;24:545–547
3. Ghali GE, Sikes JW Jr. Intraoral vertical ramus osteotomy as the preferred treatment for mandibular prognathism. *J Oral Maxillofac Surg* 2000;58:313–315
4. Bell WH, Yamaguchi Y, Poor MR. Treatment of temporomandibular joint dysfunction by intraoral vertical ramus osteotomy. *Int J Adult Orthodont Orthognath Surg* 1990;5:9–27
5. Ueki K, Hashiba Y, Marukawa K, et al. The effects of changing position and angle of the proximal segment after intraoral vertical ramus osteotomy. *Int J Oral Maxillofac Surg* 2009;38:1041–1047
6. Ueki K, Marukawa K, Shimada M, et al. Condylar and disc positions after intraoral vertical ramus osteotomy with and without a Le Fort I osteotomy. *Int J Oral Maxillofac Surg* 2007;36:207–213
7. Jung HD, Jung YS, Park HS. The chronologic prevalence of temporomandibular joint disorders associated with bilateral intraoral vertical ramus osteotomy. *J Oral Maxillofac Surg* 2009;67:797–803
8. Arimoto S, Hasegawa T, Kaneko K, et al. Observation of osseous healing after intraoral vertical ramus osteotomy: focus on computed tomography values. *J Oral Maxillofac Surg* 2013;71:1602e1601–1602e1610
9. Gassmann CJ, Van Sickels JE, Thrash WJ. Causes, location, and timing of relapse following rigid fixation after mandibular advancement. *J Oral Maxillofac Surg* 1990;48:450–454
10. Yang HJ, Lee WJ, Yi WJ, et al. Interferences between mandibular proximal and distal segments in orthognathic surgery for patients with asymmetric mandibular prognathism depending on different osteotomy techniques. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2010;110:18–24

The Le Fort I Osteotomy as Surgical Approach of an Extensive Pleomorphic Adenoma of the Palate

Jacqueline Pascale Sotong, MD, Emanuele Zavattero, MD,† Paolo Garzino-Demo, MD,* Francesca Antonella Bianchi, MD,‡ and Guglielmo Ramieri, MD, DDS**

Abstract: Pleomorphic adenoma (PA) is the most common benign mixed salivary gland tumor.

We describe here a peculiar case of a 72-year-old woman with a PA of the palate resected with palatal approach combined with Le Fort I osteotomy, adding to the currently limited scientific literature on intraoral approach for removal of extensive tumors of the midface. The Le Fort I osteotomy approach allows direct visualization of the tumor and ensures a wide excision, increasing safety and minimizing the possibility of recurrence.

Key Words: Pleomorphic adenoma, Le Fort I, surgery, maxillofacial

Pleomorphic adenoma (PA) of the palate is the most common benign mixed salivary gland neoplasm that accounts for 60% of all benign salivary glands tumors.[1] It is mostly seen in women,

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FIGURE 1. Clinical presentation of the palatal lesion.

prevalent in the fourth through sixth decades of life. PA is usually solitary and presents a slow growth.[2]

Fine-needle aspiration is mandatory for the diagnosis; information about the location, size, and extension of tumor are provided by computed tomography and magnetic resonance imaging.[3]

Surgical excision is the treatment of choice for PA of the palate. The periosteum or bone should be removed if involved. Different techniques could be used for excision. The approach depends on the tumor location and extension. The lesion should be excised with a margin of surrounding tissue, as opposed to simple enucleation, to minimize recurrence.[4]

The surgical objective becomes a formidable task when the tumor is large on initial presentation or when it extends posteriorly and superiorly into the palate. The inability to delineate adequate posterior and superior margins is a major drawback of the usual anterior permucosal approach to excise the tumor.[2,3] To ensure wide excision, some authors suggest full-thickness resection of the palate with immediate reconstruction, but this procedure is radical and requires increased operating time.[5]

Here, we report a case of pleomorphic adenoma of the palate involving the nasal cavity that was removed using palatal approach combined with Le Fort I osteotomy.

CLINICAL REPORT

A 72-year-old female patient was referred at the Division of Maxillofacial Surgery, Città della Salute e della Scienza Hospital "Molinette", University of Torino, Italy for evaluation of a painless lesion on the hard left palate.

Clinical examination showed (Fig. 1) a palpable mass in the middle of the palate. The lesion was anteriorly covered with intact mucosa except for a small ulcerated posterior area.

An incisional biopsy was performed and a diagnosis of pleomorphic adenoma was rendered.

Computed tomography showed a solid oval mass on the posterior aspect of the hard palate, which involved the palatine bone and maxillary process of the hard palate, with extension to the nasal cavity (Figs. 2A, B).

An excision of the mass was planned under general anesthesia. On the basis of the location and extension of the tumor, both palatal approach and Le Fort I osteotomy were performed to completely remove the lesion.

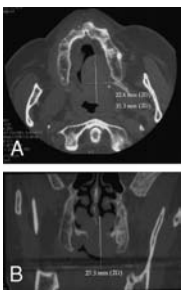

FIGURE 2. Preoperative CT scan, axial view (A) and coronal view (B) showing the pleomorphic adenoma situated in the middle of the palate and extending in the nasal cavity.

FIGURE 3. Intraoperative view showing the palatal approach. A parasagittal incision was performed to control the inferior boundaries of the tumor.

FIGURE 4. Intraoperative view in the caudal direction of the tumor in the maxilla after Le Fort I osteotomy and mobilization of the maxilla with a hook. The excision of the tumor can be performed under optimal visualization.

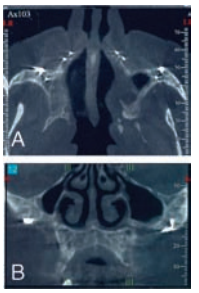

FIGURE 5. Postoperative CT scan, axial view (A) and coronal view (B), showing no evidence of recurrence.

First, the tumor was partially resected using a parasagittal palatal incision (Fig. 3). Adequate anterior, lateral, and posterior margins were been defined. The dissection was then carried superiorly and the Le Fort I osteotomy approach was used to ensure an adequate superior margin (Fig. 4). The Le Fort I osteotomy was performed in the standard lateral approach as described by Obwegeser.[6] A peripheral ostectomy was performed in areas suspected for bony involvement. The maxilla was repositioned in its original position using titanium plates (Synthes, Oberdorf, Switzerland). Four titanium miniplates with screws were used to osteosynthesize the maxilla.

The incision line in the palatal soft tissue was checked after the downfracture of the maxilla with visualization of the contour of the tumor in the nasal cavity.

Finally, a buccal fat flap was harvested to cover the hard palate and separate the nasal cavity. The nasal soft tissue layer was created by mobilization of the buccal fat pad which was fixed to the nasal mucosa with resorbable suture.

To ensure the wound healing of the palate, a prefabricated acrylic intraoral splint was used for 10 days.

The entire specimen containing overlying mucosa and underlying periosteum and bone was submitted for histopathologic examination.

No intraoperative complications were recorded. The postoperative period was uneventful. No evidence of recurrence was observed at 1-year follow-up (Figs. 5A, B).

The result of the histopathologic examination confirmed the diagnosis of pleomorphic adenoma.

A satisfactory postoperative functional and morphological result was obtained (Fig. 6).

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FIGURE 6. Postoperative view. A satisfactory clinical and functional result was obtained.

DISCUSSION

The case described in this article illustrates the feasibility and versatility of the Le Fort I osteotomy approach for wide excision of an extensive PA of the palate involving the nasal cavity. The technique allows direct visualization of the tumor from the superior surface, which helps to assure a wide extracapsular excision minimizing the possibility of recurrence.

Different approaches for removal of tumors in the palate and nasopharynx have been described during the past. Transoral approaches have often been inadequate because of technical difficulties in defining tumor margins and assuring adequate excision.[7–9]

Complete excision at the initial surgical procedure is mandatory because the recurrences are multifocal and therefore more difficult to treat.[1]

The Le Fort I approach provided a safe and excellent exposure of the tumor in this peculiar case.

Le Fort I osteotomy has been commonly used to correct midfacial skeletal deformities.[7] This technique maintains a good exposure of the surgical field. Von Langenbeck in 1859 and Cheever in 1867 have been the first to describe Le Fort I osteotomy as a surgical approach for removing tumors of the midface, particularly nasopharyngeal tumors.[8]

Sailer described in 1986 a Le Fort I approach for the removal of pathological conditions within the maxilla and reconstruction of the nasal layer covering the bony reconstruction with a buccal fat flap after a partial maxillectomy.[8] Alkan and Inal described a case of closure technique with buccal fat flap in palatal defects after excision of palatal PA.[9]

In this case, an intraoral approach through the palate was used to resect the inferior portion of the tumor.[10] To control the superior part of the mass, the Le Fort I osteotomy approach was preferred to partial maxillectomy. It has excellent acceptance by the patient because of the ability to return to normal function immediately after the procedure. It is a good choice for the surgeon because of the excellent exposure and control of the tumor and the safety of the procedure. It is quite a simple procedure in the hands of a maxillofacial surgeon.

This approach allows the removing of the nasal structures giving access and direct view into the ethmoid complex without affecting the integrity of the outer nasal skeleton and maxillary boundaries.[8]

Therefore, Le Fort I approach could facilitate the reconstructive procedures because it allows excellent access to mobilize and fix the buccal fat flap to the defect after the resection to cover and protect the nasal floor.[8,9]

Although not necessary in this case, its application must not be limited to the removal of benign tumor, and the level of the Le Fort I osteotomy could be adapted to the resection borders of the tumor and the additional cranial lateral and central approaches could be used if necessary.

To conclude, the transoral approach using Le Fort I osteotomy offers excellent access and direct vision of the operative field to remove palatal tumors involving the nasal cavity without creating aesthetic drawbacks.

REFERENCES

1. Sahoo NK, Rangan MN, Gadad RD. Pleomorphic adenoma palate: major tumor in a minor gland. *Ann Maxillofac Surg* 2013;3:195–197
2. Erdem MA, Cankaya AB, Guven G, et al. Pleomorphic adenoma of the palate. *J Craniofac Surg* 2011;22:1131–1134
3. Patigaroo SA, Patigaroo FA, Ashraf J, et al. Pleomorphic adenoma of hard palate: an experience. *J Maxillofac Oral Surg* 2014;13:36–41
4. Eneroth CM, Hertman L, Moberger L. Salivary gland tumors of the palate. *Acta Otolaryngol* 1972;73:305–315
5. Northington P. The management of the palatal pleomorphic adenoma. *Br J Oral Surg* 1974;12:132–140
6. Obwegeser HL. Surgical correction of small or retrodisplaced maxillae: the "dish face" deformity. *Plast Reconstr Surg* 1969;43:351–365
7. Alvi A, Myssiorek DJ, Schwartz M. Resection of a recurrent nasal tumor via Le Fort I osteotomy approach. *Am J Otolaryngol* 1995;16:418–421
8. Sailer HF, Haers PE, Gratz KW. The Le Fort I osteotomy as a surgical approach for removal of tumors of the midface. *J Craniomaxillofac Surg* 1999;27:1–6
9. Moloney F, Worthingtong P. The origin of the Le Fort I maxillary osteotomy: Cheever's operation. *J Oral Surg* 1981;39:731–734
10. Alkan A, Inal S. Closure of palatal defects following excision of palatal pleomorphic adenoma. *J Contemp Dent Pract* 2008;9:99–107

Study of Condylar Asymmetry in Angle Class III Malocclusion With Mandibular Deviation

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Objective: Differences in the condylar between both sides in class III cases with mandibular deviated are important for the diagnosis, treatment, and retention in this kind of patient. To detect the subtle differences, we analyzed the size, shape, and bone density with three-dimensional reconstructed technology.

Methods: The symmetry group and the asymmetry group each with 20 cases were chosen according to some standards. The computed tomographic data obtained and three-dimensional model were built with SimPlant software. The distance and angle were measured in the three-dimensional model, and the bone density was measured with the SimPlant software. The differences between the separated side in each group and the difference between the 2 groups were analyzed.

Results: In the asymmetry group, some measurement projects of the bilateral condyles showed significant differences, such as the ramal height, condylar perpendicular height, the area of maximum cross section of condylar, condylar medialateral diameter, length of

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posterior slope, and angle of posterior slope. When the asymmetry group was compared with the symmetry group, the condyles of the asymmetry group showed more asymmetrical variations in morphology, such as the ramal height, the condylar perpendicular height, the area of maximum cross section of condylar, the medialateral diameter, the length of anterior slope, and angle of posterior slope. The bone mineral density of the condylar anterior and condylar medial point was higher in the nondeviated side, and the bone mineral density of the condylar posterior was higher in the deviated side, and no statistically significant difference was found in the symmetry group.

Conclusions: In class III malocclusion with mandibular deviation patients, the three-dimensional morphology and bone density of condylar on the deviated side differ from the nondeviated side, which indicates the association between asymmetrical jaw function and joint remodeling

Key Words: mandibular deviated, condylar, three-dimensional reconstruction, bone density, orthognathics

Facial asymmetry as 1 of the most common craniofacial deformities is prevalent in patients with skeletal class III malocclusion and with a prevalence about 40%.[1–3] Recent studies showed that most of facial asymmetry was caused by unilateral mandibular hyperplasia[4,5] and was the common cause of temporomandibular joint (TMJ) disorders, imbalance of craniofacial muscle strength, and even scoliosis and trunk imbalance.[6–9] Mandibular condylar plays an important role in the development of oral-facial complex and has a special multidirection capacity for the growth and adaptive remodeling of TMJ.[10,11] Condylar shape and bone density were correlated with the pathogenesis of mandibular asymmetry and bilateral imbalanced occlusal force,[12] It was demonstrated by Saccucci et al[13] that the optimum size of the mandibular condylar was indicative and predictive of a precise clinical situation. In patients with mandibular asymmetry, imbalanced occlusion can cause abnormal stress distribution on articular surfaces and dysfunctional osseous remodeling of condylar, causing the internal derangement and functional impairment of the TMJ.[14] Akahane and colleagues'[15] study with cephalometric laminography showed that the head of the condylar in the deviated side was significantly narrow and shorter than the nondeviated side. You conducted a study using the three-dimensional computed tomography method and reported that there were significant differences in condylar size and condylar volume between the nondeviated and deviated sides compared with the symmetry group.[3] Skeletal class III malocclusion tends to show the distinguishing feature not only the mandibular but also the condylar inclination compared with classes I and II malocclusion.[17] But the characteristic of the condylar asymmetry about the class III malocclusion with mandibular deviation had been discussed little.

Based on the computed tomographic (CT) data, three-dimensional reconstruction is considered an effective tool to study asymmetry.[18] Computed tomographic images are reconstructed into three-dimensional image; errors caused by magnification and distortions are decreased, and quantitative measurements of the anatomic structures of interest are possible.[19] Surgeons can get real anatomic surface landmarks, accurate dimensional measurements, angle measurements, and even the volumetric measurements.[20,21] Therefore, three-dimensional reconstruction can provide more information about the configuration and bone changes of condyles other than simple distance and angle measurement reported by the previous researchers.[22]

In order to detect the subtle differences in patients of class III malocclusion with mandibular deviation, the distance, angle measurement, and bone mineral density (BMD) of three-dimensional reconstructed condylar models on both nondeviated and deviated sides were analyzed. The results are discussed with the aim of providing a more objective quantification way for evaluating the facial asymmetry and more useful information for the treatment of class III malocclusion with mandibular deviation.

FIGURE 1. The front view of the asymmetry cases. MD indicates the perpendicular distances between point Me to the midsagittal reference line.

MATERIALS AND METHODS

As the chin is strongly related to the perception of facial asymmetry, facial asymmetry is defined by the degree of menton deviation (MDs) from the midsagittal reference line (Fig. 1).[23,24] The midsagittal reference line is defined with the methods recommended by Grummons and Kappeyne van de Coppello. According to the MDs, the present study were divided into 2 groups,the symmetry group and the asymmetry group, both with 20 cases. The symmetry group was composed of 20 volunteers from Qingdao University (10 men and 10 women; average age, 18 years) without asymmetry, whose MDs were less than 2 mm. The asymmetry group with 20 cases (10 men and 10 women; average age, 20 years) was selected from the patients with skeletal class III (ANB <-4 degrees) who had undergone a CT scan for the purpose of presurgical evaluation at the Department of Oral Surgery, Qingdao Municipal Hospital, between 2011 and 2013, with MDs of more than 5 mm. All the 40 patients studied had no systemic disease and no degenerative disease of the TMJ. The patient's characteristics in the symmetry and asymmetry groups are listed in Table 1.

TABLE 1. Patient Characteristics in the Symmetry and Asymmetry Groups

Variable	Mean	SD	Minimum	Maximum
Symmetry group (n = 20)				
Age, y	19	2	18	20
ANB, degrees	1.5	2.6	−1.5	2.6
Pog to N perpendicular, mm	1.4	3.3	−1.8	3.5
MD, mm	1.2	0.5	0.4	2.0
Asymmetry group (n = 20)				
Age, y	20	6.2	17	26
ANB, degrees	−4.3	2.6	−2.5	−8.1
Pog to N perpendicular, mm	7.0	3.4	5.1	13.0
MD, mm	6.7	3.4	5	12

TABLE 2. Description of Landmarks

Landmark	Definition
Me (menton)	The most inferior midpoint on the symphysis
Aco	The most anterior point of the condylar head
Sco	The most superior apex of the condylar head
Pco	The most posterior point of the condylar head
Mco	The most medial point of the condylar head
Lco	The most lateral point of the condylar head
Inf (simoid notch)	The lowest point of the sigmoid notch
Gomid (gonion midpoint)	The midpoint between Go post and Go inf on the mandibular angle

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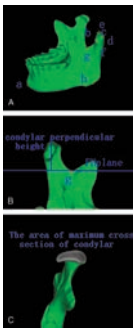

FIGURE 2. A, Landmarks of the mandibular: a, Me; b, Aco; c, Sco; d, Pco; f, Lco; g, Inf; h, Gomid. B, The condylar perpendicular height measurement method. C, The area of maximum cross section of condylar measurement method.

The CT scans were obtained by using a spiral CT scanner (SOMATOM Definition AS+; Siemens, Berlin, Germany) with a 512×512 matrix, 120 kV, and 200 mA. The thickness of the axial image was 1.0 mm. The patients were positioned with the Frankfort horizontal plane perpendicular to the floor and the facial midline coinciding with the long axis of the CT machine.[22] The DICOM (digital imaging and communication in medicine) images were created in 1.0-mm slice thicknesses after scanning. The DICOM data were reconstructed into three-dimensional images using SimPlant software. The mandibles were separated from the whole images.

Landmarks (Table 2 and Fig. 2) were designated on the reconstructed three-dimensional surface model, and their positions were verified on the axial, coronal, and sagittal slices. The following bilateral measurements were made (Table 3): (1)ramal height; (2) mandibular body length; (3) the condylar perpendicular height (Fig. 2B); (4) the area of maximum cross section (Fig. 2C); (5) condylar anteroposterior diameter; (6) condylar medialateral diameter; (7) length of anterior slope; (8) length of posterior slope; (9)angle of anterior slope; (10) angle of posterior slope; (11) condylar axis angle. The data were measured in units of 0.01 mm or 0.01 degree. Each item was measured 3 times by the experimenter, and the mean was obtained.

The bone density analysis tool of the SimPlant software was used to measure the BMD of the point: condylar anterior (Aco), condylar posterior (Pco), condylar superius (Sco), condylar medial point (Mco), and condylar lateral point (Lco) in the 2 groups. The area of the measurement ellipse was designated as 3.78 mm^2, and every point was measured in 3 different slices in the axial plane (Fig. 3).

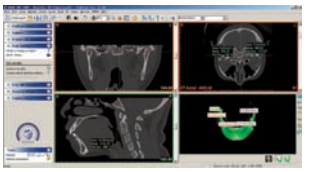

FIGURE 3. The measurement of the condylar BMD.

Statistical Analysis

The data were statistically analyzed using SPSS 18.0 for Windows (SPSS Inc, Chicago, IL). At first, paired-samples *t* test was used to analyze the difference between the left side and the right side in the symmetry group or the difference between the deviated side and the nondeviated side in the asymmetry group. Then, using independent-samples *t* test compared the differences of left minus right sides in symmetry group with deviated side minus nondeviated side in the asymmetry group.

RESULTS

In the symmetry group, the bilateral condylar, no significant differences were found by using paired-samples *t* test ($P > 0.05$). In the asymmetry group, some measurement projects of the bilateral condyles showed significant differences, such as the ramal height ($P < 0.05$), condylar perpendicular height ($P < 0.01$), the area of maximum cross section of condylar ($P < 0.01$), condylar medialateral diameter ($P < 0.01$), length of posterior slope ($P < 0.01$), and angle of posterior slope ($P < 0.05$). When the asymmetry group was compared with the symmetry group, it was found that the condylar of the asymmetry group occurred to asymmetrical variations in morphology by using independent-samples *t* test. There are some measurement projects with significant differences such as the ramal height ($P < 0.01$), the condylar perpendicular height ($P < 0.01$), the area of maximum cross section of condylar ($P < 0.01$), the medialateral diameter ($P < 0.01$), the length of anterior slope ($P < 0.01$), and angle of posterior slope ($P < 0.01$), confirming the overall asymmetrical morphology of the condylar (Table 4).

In the asymmetry group, the BMDs of the Aco and Mco were higher in the nondeviated side ($P < 0.01$), and the BMDs of the Pco were higher in the deviated side ($P < 0.05$), and no statistically significant difference was found in the symmetry group (Figs. 4A, B).

DISCUSSION

The etiology of facial asymmetry has been studied by predecessors, and the common view was that the facial asymmetry was caused by genetic factors or the acquired environmental factors, even both working together. The present study shows that misregulation of

TABLE 3. Description of Measurement Project

Measurement Project	Definition
Ramal height	The distance between Sco-Gomid
Mandibular body length	The distance between Gomid-Me
Condylar perpendicular height	The vertical height of the Sco to the plane parallel with the Frankfort horizontal plane, tangency to the Inf
Condylar volume	The volume of the part of condylar above the plane parallel with the Frankfort horizontal plane, tangency to the Inf
The area of maximum cross section of condylar	The area of maximum cross section of condylar tangency to the plane parallel with the Frankfort horizontal plane
Condylar anteroposterior diameter	The distance between Aco-Pco
Condylar media lateral diameter	The distance between Mco-Lco
Length of anterior slope	The distance between Sco-Aco
Length of posterior slope	The distance between Sco-Pco
Angle of anterior slope	The angle between line of Sco-Aco with Frankfort horizontal plane
Angle of posterior slope	The angle between line of Sco-Pco with Frankfort horizontal plane
Condylar axis angle	The angle between line of Mco-Lco and the midsagittal reference plane in the slice

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TABLE 4. Statistic Analysis of the Condylar Measurement of the Symmetry Group and Asymmetry Group

	Symmetry Group (n = 20)			Asymmetry Group (n = 20)			
	Left Side	Right Side	Difference (Left Side–Right Side)	Deviated Side	Nondeviated Side	Difference (Deviated Side–Nondeviated Side)	Independent-Samples *t* Test
Measurement	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	*P*
Ramal height, mm	61.71 (6.4)	62.15 (5.3)	−0.44 (1.43)	61.32 (4.17)	70.08 (9.82)	−8.76 (4.30)*	<0.01
Mandibular body length, mm	91.2 (6.3)	89.54 (5.8)	1.66 (2.97)	86.14 (11.16)	90.39 (6.40)	−4.26 (6.40)	>0.05
Condylar perpendicular height, mm	19.50 (2.66)	19.82 (2.70)	−0.32 (1.41)	20.33 (2.98)	26.22 (3.33)	−5.89 (1.34)†	<0.01
The area of maximum cross section of condylar, mm^2	110.97 (10.4)4	116.78 (9.87)	−4.19 (9.64)	109.67 (12.07)	120.36 (10.49)	−10.70 (7.1)†	<0.01
Condylar anteroposterior diameter, mm	13.64 (1.26)	13.48 (1.22)	1.96 (2.33)	12.22 (2.03)	14.31 (2.42)	−2.26 (2.19)	>0.05
Condylar medialateral diameter, mm	19.26 (1.66)	18.97 (1.68)	0.49 (0.43)	17.82 (2.33)	22.68 (1.68)	−5.23 (1.17)†	<0.01
Length of anterior slope, mm	6.81 (0.62)	6.82 (0.78)	−1.21 (2.56)	5.51 (0.55)	6.82 (0.78)	−1.52 (1.70)*	>0.05
Length of posterior slope, mm	11.01 (1.16)	10.68 (1.04)	−0.33 (0.78)	10.01 (1.16)	6.68 (1.04)	3.33 (1.55)†	<0.01
Angle of anterior slope, degrees	47.6 (5.7)	48.1 (5.9)	−0.5 (5.4	56.63 (5.7)	62.17 (5.9)	−5.54 (5.4)	>0.05
Angle of posterior slope, degrees	53.9 (4.7)	55.8 (5.7)	−1.9 (5.1)	53.91 (4.74)	49.8 (3.72)	4.10 (2.50)†	<0.01
Condylar intersection angle, degrees	69.8 (5.6)	70.6 (6.2)	−0.8 (1.14)	74.59 (4.75)	72.93 (5.59)	1.56 (3.49)	>0.05

* $P < 0.05$;
† $P < 0.01$.

growth after birth was regarded as a major reason leading to mandibular deviation.[25] The cause-and-effect relationship between the mandibular deviation and the morphology of the condylar has been discussed in the previous studies. As we learn from the study of Mongini and Schmid,[26] in the early stage of mandibular deviation such as the mixed dentition, there was no significant difference in the morphology of the condylar between the deviation side and the nondeviation side. If the deviation was not corrected timely, the difference would become obvious along with the age growth. Zhao and colleagues'[27] study suggested that the reduction of the condylar size occurred as an adaptive change to mandibular deviation. During this process, the muscle plays an important role in the change of the condylar morphology.[28] The muscle's contraction and stretching force worked on the mandibular condylar will change the growth environment of the condylar cartilage and generate gradually variation on the morphology of the condylar.[29] As the primary center of mandible growth, the condyle undergoes a remodeling process as the responses to continuous stimuli during jaw movements.[30] But asymmetrical jaw function alters the intra-articular mechanical dynamics, which changed the persistent or renewed activity in 1 or both of the condyles. And in our study, the condylar expresses corresponding difference in the deviated side and the nondeviated side. Statistically

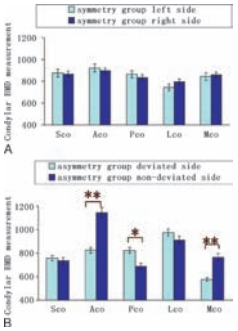

FIGURE 4. Bone mineral density measurement of the condylar in different groups. A, Bone mineral density measurement of the condylar of the symmetry group. B, Bone mineral density measurement of the condylar of the asymmetry group (*$P < 0.05$, †$P < 0.01$).

significant differences were found in the size (the ramal height, condylar perpendicular height, the area of maximum cross section of condylar, condylar medialateral diameter, length of posterior slope) and the angle (angle of posterior slope). In the nondeviated side, the condylar was pushed forward and downward, so under the work of stretching force, the head of the condylar in the nondeviated side became significantly wider and bigger than the deviated side, which was corresponding with the study of Akahane et al.[15] When the contour of the condylar was analyzed, the posterior slopes were more flat and short in the nondeviated group, and there were no significant differences in the condylar intersection angle between the 2 groups.

Mechanical effects of loading as the initial factor in bone remodeling not only produce alterations in the contour and shape of the subchondral bone but also affect the bone mass.[31] Significant difference was detected in the BMD measurement between the 2 sides in the asymmetry group, and we also found that in different locations of the condylar, there were statistically different BMD expressions. Higher BMD was obtained on the most anterior point of the condylar head and the most medial point of the condylar head of the nondeviated side, but lower BMD was obtained on the most posterior point of the condylar head. But there were no statistically significant differences in the BMD of the most superior apex of the condylar head between the deviated side and the nondeviated side. The higher bone density of anterior slope in the nondeviated side compared with the deviated side may be explained by anterior slope role as the main burdened surface during the jaw movement and the anterior slope of the nondeviated side bears more strength than the offside. The lower bone density on the posterior slope on the nondeviated side shows the posterior slope endures lower strength than does the deviated side. Hence, the minor trauma of the anterior slope on the nondeviated side always acts as the initial sign of TMJ dysfunctions.[32] And a well-balanced position of the condyle relative to the glenoid fossa may be critical to the ordinate function of the TMJ, which should be implicated in the teeth alignment, occlusal treatment, and orthotherapy. When the class III malocclusion with mandibular deviation patients was treated by orthognathics, the differences of the shape, size, and bone density in condylar between both sides still exist, and the imbalance of the muscle would not disappear just after the operation, so the retention was important

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before the new remolding of the condylar to adjust the new position of the mandibular.

CONCLUSIONS

In class III malocclusion with mandibular deviation patients, the three-dimensional morphology and bone density of condylar on the deviated side differ from the nondeviated side, which indicates the association between asymmetrical jaw function and joint remodeling.

REFERENCES

1. Severt TR, Proffit WR. The prevalence of facial asymmetry in the dentofacial deformities population at the University of North Carolina. *Int J Adult Orthod Orthognath Surg* 1997;12:171–176
2. Samman N, Tong AC, Cheung DL, et al. Analysis of 300 dentofacial deformities in Hong Kong. *Int J Adult Orthod Orthognath Surg* 1992;7:181–185
3. You K-H, Lee K-J, Lee S-H, et al. Three-dimensional computed tomography analysis of mandibular morphology in patients with facial asymmetry and mandibular prognathism. *Am J Orthod Dentofacial Orthop* 2010;138:540.e1–540.e8
4. Obwegeser HL, Makek MS. Hemimandibular hyperplasia–hemimandibular elongation. *J Maxillofac Surg* 1986;14:183–208
5. Yoon KS, Jung YS, Kang GC, et al. Facial asymmetry with mandibular prognathism: a new trial of classification and interpretation. *J Korean Assoc Oral Maxillofac Surg* 2004;30:108–120
6. Ferro F, Spinella P, Lama N. Transverse maxillary arch form and mandibular asymmetry in patients with posterior unilateral crossbite. *Am J Orthod Dentofacial Orthop* 2011;140:828–838
7. Fudalej P, Kokich VG, Leroux B. Determining the cessation of vertical growth of the craniofacial structures to facilitate placement of single-tooth implants. *Am J Orthod Dentofacial Orthop* 2007;131:S59–S67
8. Chung KR, Kim SH, Choo H, et al. Distalization of the mandibular dentition with mini-implants to correct a class III malocclusion with a midline deviation. *Am J Orthod Dentofacial Orthop* 2010;137:135–146
9. Shuncheng Zhou, Juanjuan Yan, Hu Da, Yang Yang, et al. A correlational study of scoliosis and trunk balance in adult patients with mandibular deviation. *PLoS One* 2013;8:e59929
10. Zane Krisjane, Ilga Urtane, Gaida Krumina, et al. Condylar and mandibular morphological criteria in the 2D and 3D MSCT imaging for patients with class II division 1 subdivision malocclusion. *Stomatol Baltic Dent Maxillofac J* 2007;9:67–71
11. Hans MG, Enlow DH, Noachtar R. Age-related differences in mandibular ramus growth: a histologic study. *Angle Orthod* 1995;65:335–340
12. Han Lin, Ping Zhu, Yi Lin, et al. Mandibular asymmetry: a three-dimensional quantification of bilateral condyles. *Head Face Med* 2013;9:42
13. Saccucci M, Polimeni A, Festa F, et al. Do skeletal cephalometric characteristics correlate with condylar volume, surface and shape? A 3D analysis. *Head Face Med* 2012;8:15
14. Ueki K, Nakagawa K, Marukawa K, et al. Stress change on the temporo-mandibular joint in mandibular prognathism subjects with asymmetry after orthognathic surgery. *Eur J Orthod* 2010;V32N5:522–529
15. Akahane Y, Deguchi T, Hunt NP. Morphology of the temporomandibular joint in skeletal class III symmetrical and asymmetrical cases: a study by cephalometric laminography. *J Orthod* 2001;28:119–128
16. Li ZB, Sun GW, Dong YJ, et al. The orthognathic treatment of mandibular asymmetry. *Hua Xi Kou Qiang Yi Xue Za Zhi* 2004;22:484
17. Kwon TG, Park HS, Ryoo HM, et al. A comparison of craniofacial morphology in patients with and without facial asymmetry—a three-dimensional analysis with computed tomography. *Int J Oral Maxillofac Surg* 2006;35:43–48
18. Letzer GM, Kronman JH. A posteroanterior cephalometric evaluation of craniofacial asymmetry. *Angle Orthod* 1967;37:144–158
19. Pirttiniemi P, Miettinen J, Kantomaa T. Combined effects of errors in frontal-view asymmetry diagnosis. *Eur J Orthod* 1996;18:629–636
20. Van Elslande DC, Russett SJ, Major PW, et al. Mandibular asymmetry diagnosis with panoramic imaging. *Am J Orthod Dentofacial Orthop* 2008;134:183–192
21. Grayson B, Cutting C, Bookstein FL, et al. The three-dimensional cephalogram: theory, technique, and clinical application. *Am J Orthod Dentofacial Orthop* 1988;94:327–337
22. Haraguchi S, Takada K, Yasuda Y. Facial asymmetry in subjects with skeletal class III deformity. *Angle Orthod* 2002;72:28–35
23. Ahn JS, Lee KH, Hwang HS. Relationship between perception of facial asymmetry and posteroanterior cephalometric measurements. *Korean J Orthod* 2001;31:489–498
24. Obwegeser HL. Mandibular growth anomalies: terminology, aetiology, diagnosis, treatment Berlin, Germany: Springer-Verlag; 2001:43–353
25. Grummons DC, Kappeyne van de Coppello MA. A frontal asymmetry analysis. *J Clin Orthod* 1987;21:448–465
26. Mongini F, Schmid W. Treatment of mandibular asymmetries during growth—a longitudinal study. *Eur J Orthod* 1987;9:5–1
27. Zhao C, Kurita H, Kurashina K, et al. Temporomandibular joint response to mandibular deviation in rabbits detected by 3D micro-CT imaging. *Arch Oral Biol* 2010;9:929–937
28. Yoshino T. Effects of lateral mandibular deviation on masseter muscle activity. *Kokubyo Gakkai Zasshi* 1996;63 (1):70–80
29. Kawakami M, Yamada K, Inoue M, et al. Morphological differences in the temporomandibular joints in asymmetrical prognathism patients. *Orthod Craniofacial Res* 2006;9:71–76
30. Tecco S, Saccucci M, Nucera R, et al. Condylar volume and surface in Caucasian young adult subjects. *BMC Med Imaging* 2010;10:28
31. Zhang J, Jiao K, Zhang M, et al. Occlusal effects on longitudinal bone alterations of the temporomandibular joint. *J Dent Res* 2013;92:253–259
32. Alexiou K, Stamatakis H, Tsiklakis K. Evaluation of the severity of temporomandibular joint osteoarthritic changes related to age using cone beam computed tomography. *Dentomaxillofac Radiol* 2009;38:141–147

Lower Eyelid Reconstruction for Iatrogenic Eyelid-to-Globe Malposition

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Abstract: An 86-year-old man with iatrogenic eyelid-to-globe malposition had a combination of medial tarsal strip, lateral periosteal sling, and inferior tarsal support with the ear cartilage. An appropriate apposition between the globe and lower eyelid was established postoperatively with good tear meniscus and acceptable cosmetic results.

Key Words: Ear cartilage, eyelid-to-globe malposition, lateral periosteal sling procedure, lower eyelid ectropion, medial tarsal strip procedure

Appropriate eyelid-to-globe apposition is essential for lower eyelid reconstruction, if not iatrogenic dry eye may occur.

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The authors report no conflicts of interest.

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The medial and lateral canthal supports originate from the posterior lacrimal crest and the Whitnall tubercle, respectively.[1] If the lower eyelid is not refixated on to these sites during reconstruction, the medial or lateral area of the lower eyelid will "float" from the globe.

We present a case of an iatrogenic lower eyelid-to-globe malposition resulting from an anatomically unacceptable ear cartilage graft fixated on the medial and lateral orbital rims. We stressed the importance of refixating the medial and lateral canthi to their origins in apposing the reconstructed eyelid to the globe.

CASE REPORT

An 86-year-old man had a right facial nerve palsy after excision of right parotid gland tumor 7 years ago. He was presented to us with right ocular pain and epiphora. Before that, he had had the Kuhnt-Szymanowski-Smith procedure[2] with ear cartilage implantation to the ectropic lower eyelid.

On initial examination, the right lower eyelid was straight and rigid, and was not in contact with the globe in both medial and lateral areas (Fig. 1A). Slit lamp examination demonstrated diffuse superficial punctate keratopathy and conjunctival injection on the right eye. Visual acuity on right eye was no light perception because of an old retinal detachment.

We performed the lower eyelid reconstruction under local anesthesia. An initial subciliary incision was made and reached the lower tarsal surface. We exposed and removed the ear cartilage that was fixated on to the medial, lower central and lateral orbital rims, and lower tarsal plate. As the lacrimal drainage does not function in a facial palsy case, we first performed the medial tarsal strip procedure[3] which involves denuding the medial tarsal edge, with incision of the lower lacrimal canaliculus, then the tissue from the caruncle to the posterior lacrimal crest is undermined via the transcaruncular approach (Fig. 1B).[4] Finally, the medial edge of the lower tarsal plate was fixated to the periosteum of the posterior lacrimal crest with 5-0 PDS-II (Johnson & Johnson K.K., Tokyo, Japan) suture (Fig. 1C). Then, we performed a lateral canthoplasty with a periosteal flap from the lateral orbital rim with its base reaching 2 mm behind the lateral orbital rim at the Whitnall tubercle (Fig. 1D). The periosteal flap was sutured to the lower tarsal plate with 5-0 PDS-II suture (Fig. 1E).[5] Midface lift procedure, which was performed by the suborbicularis oculi fat lifting to the inferior orbital rim with 2 points sutures of 5-0 PDS-II, was added to secure a sufficient volume of the lower eyelid skin (Fig. 1F). The removed cartilage was trimmed as a rectangular shape (8×17 mm) and inserted between the lower tarsal plate and lower eyelid retractors.[6] Finally, the wound was closed with interrupted 6-0 PDS-II sutures.

After 2 months postoperatively, the lower eyelid was apposed well on the ocular surface, by which the corneal condition was completely resolved, although epiphora was persistent. The lower eyelid curvature was acceptable without complication such as lower eyelid entropion, ectropion, or retraction (Fig. 1G).

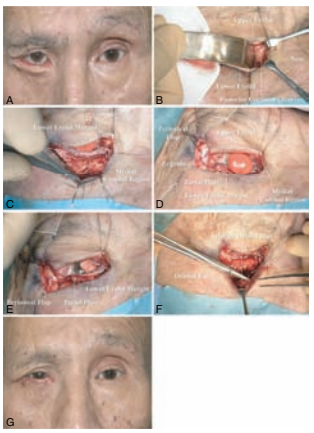

FIGURE 1. A, Preoperative photograph. The right lower eyelid is straight and rigid, with loss of contact with the globe. B–F, Intraoperative photographs (upper: superior, right: nasal). B, The posterior lacrimal crest is exposed through transcaruncular approach. C, The medial tarsal edge is sutured to the periosteum of the posterior lacrimal crest. D, Marking periosteal flap on the zygomatic bone. The design is always toward superiorly to have a sling effect. E, The periosteal flap is sutured to the lower tarsal plate. F, Midface lift procedure. The sub-orbicularis oculi fat is raised to the inferior orbital rim with 2 points lifting sutures of 5-0 PDS-II. G, Postoperative photograph. Appropriate apposition between the globe and lower eyelid is shown.

DISCUSSION

Appropriate eyelid-to-globe apposition was regained through the anatomically reasonable canthal fixations with vertical ear cartilage support.

The posterior lacrimal crest is the origin of the medial canthal support. Therefore, connection of the lower tarsal plate to the posterior lacrimal crest appropriately apposes the lower eyelid and the globe.[7] On the other hand, medial canthal fixation on to the orbital rim, which was present preoperatively in our case, caused anterior displacement of the lower eyelid margin and resulted in dry eye.[8]

The medial tarsal strip procedure was performed to reconstruct the medial canthus,[4] as the lower tarsal length was too short in our case to perform the lateral tarsal strip procedure.[9] We used, therefore, a periosteal flap,[5] and sutured it to the lateral area of the lower tarsal plate. The base of the periosteal flap was set on the Whitnall tubercle, which is the origin of the lateral canthal support.[1] This regained an appropriate apposition of the globe and the lateral area of the lower eyelid.[5]

The removed cartilage was again implanted to the lower eyelid after shaving to an appropriate size using a surgical technique for a lower eyelid lengthening.[6] Although this technique was originally described for the thyroid-related lower eyelid retraction and cicatricial entropion,[8] we used it successfully in this case. The rationale for using the cartilage was to support the lower eyelid against gravity and prevent cicatricial contracture.

The lower eyelid skin is continuous to the midface and is affected by gravity.[10] Therefore, the midface lift procedure as well as the lower eyelid lengthening was crucial to the current procedure to maintain a stable position of the lower eyelid. The atonic lower eyelid and cheek, if not corrected simultaneously, would result in lower eyelid sagging or ectropion postoperatively.[10]

In conclusion, understanding the anatomical basis for the medial and lateral canthoplasty is crucial in establishing an appropriate apposition between the globe and the lower eyelid to achieve good functional and cosmetic outcomes.

REFERENCES

1. Kakizaki H, Malhotra R, Madge SN, et al. Lower eyelid anatomy: an update. *Ann Plast Surg* 2009;63:344–350
2. Smith B, Cherubini TD. Oculoplastic surgery. St Louis, MO: CV Mosby; 1970:92–94
3. Jordan DR, Anderson RL, Thiese SM. The medial tarsal strip. *Arch Ophthalmol* 1990;108:120–124
4. Fante RG, Elner VM. Transcaruncular approach to medial canthal tendon placation for lower eyelid laxity. *Ophthal Plast Reconstr Surg* 2001;17:16–27
5. Maloof A, Ng S, Leatherbarrow B. The maximal Hughes procedure. *Ophthal Plast Reconstr Surg* 2001;17:96–102
6. Kakizaki H, Zako M, Iwaki M. Lower eyelid lengthening surgery targeting the posterior layer of the lower eyelid retractors via a transcutaneous approach. *Clin Ophthalmol* 2007;1:141–147

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7. Kakizaki H, Zako M, Nakano T, et al. Direct insertion of the medial rectus capsulopalpebral fascia to the tarsus. *Ophthal Plast Reconstr Surg* 2008;24:126–130
8. Demirci H, Hassan AS, Elner SG, et al. Comprehensive, combined anterior and transcaruncular orbital approach to medial canthal ligament plication. *Ophthal Plast Reconstr Surg* 2007;23:384–388
9. Anderson RL, Gordy DD. The tarsal strip procedure. *Arch Ophthalmol* 1979;97:2192–2196
10. Elner VM, Mauffray RO, Fante RG, et al. Comprehensive midfacial elevation for ocular complications of facial nerve palsy. *Arch Facial Plast Surg* 2003;5:427–433

Surgical Repair of Lacerated Anterior Cerebral Artery Presented With Massive Intracerebral Hemorrhage

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Objective and Importance: Traumatic intracranial aneurysms present diagnostic and therapeutic challenges. Owing to their fragile nature, endovascular intervention has become the first-line treatment; however, direct surgery has an advantage in certain cases.
Clinical Presentation: A 34-year-old man in coma was admitted after a motor vehicle accident. Brain computed tomographic scans revealed deep bifrontal, left intraventricular, and subarachnoid hemorrhages. Three-dimensional computed tomographic angiography and digital subtraction angiography revealed an aneurysm arising from the left pericallosal artery.
Intervention: A massive intracerebral hematoma prompted us to perform emergency surgical intervention. We immediately removed the hematoma and extirpated the aneurysm. After hematoma evacuation via the interhemispheric approach, a pulsating red sphere projecting from the pericallosal artery, with no obvious solid wall or neck, was encountered. While retracting the frontal lobe, it suddenly ruptured. Under temporary trapping of the parent artery, the point of bleeding was identified. No aneurysm wall or fibrous tissue was present, whereas a 1.5-mm laceration was observed at the pericallosal artery close to its branching point. The laceration was sutured with 10-0 nylon. Postoperative digital subtraction angiography confirmed patency of the pericallosal artery.
Conclusions: Although recent technologic advances of intravascular surgery have enabled successful treatment of traumatic pseudoaneurysms, open surgical intervention still has some advantages of providing definitive hemostasis, allowing for parent artery reconstruction, and facilitating mass reduction. The case in the current study was quite unusual in that angiographic aneurysm had disrupted easily, leaving arterial laceration. This finding implies the probability of unavoidable parent artery occlusion when endovascular treatment is applied.

Key Words: Anterior cerebral artery, direct surgery, massive cerebral hemorrhage, traumatic brain injury, traumatic pseudoaneurysm

Intracranial aneurysms after closed head injuries are difficult to diagnose and challenging to treat. These fragile aneurysms frequently rerupture during surgery, causing considerable surgical risk.[1,2] As a result of recent technologic advances of intravascular surgery, there is an ongoing paradigm shift toward minimally invasive management of these lesions.[3,4] Recently, successful endovascular treatment of intracranial pseudoaneurysms has been reported.[3–9] However, direct surgical procedure may be indicated in certain cases. Here, we report the case of a traumatic pseudoaneurysm of the left distal anterior cerebral artery (ACA) immediately operated on. Associated massive intracerebral hematoma prompted us to perform emergency surgical intervention.

CLINICAL REPORT

A 34-year-old man in coma was referred to our hospital after a motor vehicle accident. When the emergency physician arrived at the scene, the patient was in a deep coma associated with massive oronasal bleeding and showed paradoxical breathing. His consciousness level was assessed at 3 on the Glasgow Coma Scale. He was intubated at the initial hospital, and profound epistaxis was controlled with staple sliver. Brain computed tomographic (CT) scans revealed deep bifrontal, left intraventricular, and subarachnoid hemorrhages. A dot-shaped high density was noted close to the cerebral falx (Fig. 1). Three-dimensional computed tomographic (CT) angiography (CTA) (Fig. 2A) and digital subtraction angiography (DSA) (Fig. 2B) revealed a left ACA distal aneurysm. Fractures of the nose, zygoma, right radius, and ulna were also confirmed. The patient was transferred to our hospital for neurosurgical treatment.

On arrival, he was intubated and sedated. His blood pressure was 128/80 mm Hg and his heart rate was 92/min; both his pupils were reactive. Repeated brain CT scans revealed no enlargement of the hematoma. On the basis of radiologic findings, the aneurysm of the distal ACA, presumably of a traumatic nature, was considered to be the causative lesion. Emergency operation was performed to remove the hematoma and to prevent rebleeding. Using the interhemispheric approach, the hematoma was evacuated and the

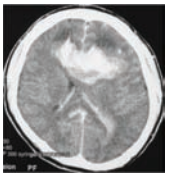

FIGURE 1. Initial brain CT scan on arrival showing interhemispheric and deep frontal hematoma associated with subarachnoid and intraventricular hemorrhage.

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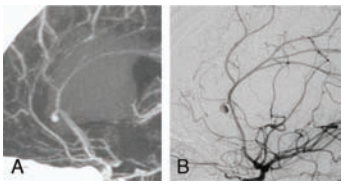

FIGURE 2. Three-dimensional CTA (A) and left carotid angiogram (B) revealing an aneurysm of the left pericallosal artery.

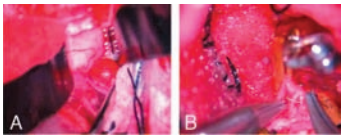

FIGURE 3. Intraoperative photographs. A, A pulsating red sphere was detected in the interhemispheric fissure. B, The laceration of the pericallosal artery was sutured. No residual neck or fibrin clot was identified around the laceration site.

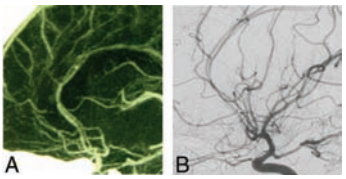

FIGURE 4. Three-dimensional CTA (A) and left carotid angiogram (B) demonstrating disappearance of the aneurysm and patency of the left pericallosal artery.

aneurysm was explored. A pulsating red sphere projecting from the pericallosal artery was encountered. Because it had no solid wall or neck, neck clipping seemed to be impossible (Fig. 3A). While retracting the frontal lobe, it suddenly ruptured. Under temporary trapping of the pericallosal artery and its cortical branch, the bleeding point was explored. Notably, no aneurysm wall or fibrous tissue was present, whereas a 1.5-mm laceration was observed at the pericallosal artery close to its branching point (Fig. 3B). The laceration was sutured with 10-0 nylon. Postoperative CT scans showed no residual hematoma or ischemic lesion (Fig. 4A). Postoperative three-dimensional CTA and DSA confirmed patency of the ACA (Fig. 4B). He underwent a ventriculoperitoneal shunt for progressive hydrocephalus. He was able to eat and obey commands at the time of discharge.

DISCUSSION

Traumatic cerebral aneurysm is uncommon, accounting for less than 1% of all intracranial aneurysms.[2] Skull-base arteries and the distal segment of the ACA are frequently affected in trauma cases. They may occur after a blunt closed head injury,[1,3,4,10–14] a penetrating wound,[3,4,15] or iatrogenic arterial damage.[4,6,16] Traumatic distal ACA aneurysms are predominantly seen in children and young adults[13,17] after severe trauma such as motor vehicle accidents or falls[10,12–14,18]; even minor injuries could be a causative event.[19] Occasionally, it is difficult to define the relationship between trauma and the preexisting intracranial saccular aneurysm.[10,20,21] Traumatic aneurysms may be confirmed through autopsy or forensic study.[22] Their typical clinical course is a delayed intracerebral hemorrhage a few weeks after trauma.[2,3,12,15] Hemorrhage in the interhemispheric fissure, corpus callosum, or deep frontal lobe is a suspicious finding of traumatic distal ACA aneurysm.[9,12] Screening with three-dimensional CTA is useful to observe vascular injury before catheter angiography. Of note, initial angiographic studies may not have disclosed an aneurysm and subtle angiographic changes may have been overlooked.[1,3,8,11,13] Some authors emphasize repeated angiography because angiography undertaken on the day of trauma identifies aneurysms in less than half of the cases.[11] Angiographic characteristics include a nonbranching location, irregular outline of the sac wall, delayed filling and emptying of the sac, as well as less opacification.[15,22] The vast majority of reported cases are diagnosed and treated several days or weeks after trauma. The case in the current study was unusual in that the aneurysm was visualized on the day of injury. Thus, rupture of a preexisting aneurysm should be differentiated.

Traumatic aneurysms can be histologically classified as true, false, dissecting, or mixed on the basis of the disrupted extent of the arterial wall.[2,11,15] Involvement of the media, internal elastic lamina, and media with intact adventitia leads to a true aneurysm, whereas disruption of all layers results in a false aneurysm. In our case, it had neither a true nor fibrous wall but consisted of a very thin fragile membrane that ruptured very easily. To our knowledge, such a lesion has not been reported thus far. It may be an initial stage of traumatic pseudoaneurysm. The laceration proximity to the branching point suggested shearing forces between the pericallosal artery and its branch to impose injury.

Frequent rupture has been reported for traumatic aneurysms, causing a high mortality of up to 50%.[2] Thus, once an aneurysm is detected, procedures to prevent rebleeding should be initiated. Surgical interventions include neck clipping, aneurysm resection and suture, as well as trapping with or without vascular reconstruction. Parent artery occlusion might be unavoidable in the treatment of pseudoaneurysm. Endovascular procedures seem safer and valuable because surgical exploration of this aneurysm carries a significant risk. Recently, a successful endovascular obliteration of the aneurysm[4,7,9] and the parent artery[8] has been reported. In the current study, the aneurysm was so fragile that the fibrin clot covering the arterial laceration was easily taken off spontaneously, leaving a bleeding orifice. Obviously, rerupture would have occurred if coil embolization of the aneurysm was attempted; thus, parent artery occlusion might be inevitable. Discrepancies between preoperative neuroimaging and surgical findings should be noted in the treatment of vascular injury on an emergency basis.

In conducting a direct surgical approach, surgeons should bear in mind (1) that traumatic pseudoaneurysms are very fragile, limiting the possibility of ordinary neck clipping, and (2) that vascular reconstruction with bypass surgery may be necessary in cases where simple suture is unavailable.[1] Egashira et al[18] reported clipping after A3 side-to-side anastomosis, and Dunn et al[16] described autogenous arterial graft interposition with resection of the aneurysm. Although the management of traumatic vascular injury has evolved with the development of intravascular surgery, open surgical intervention still has some advantages of providing definitive hemostasis, allowing for parent artery reconstruction, and facilitating mass reduction.

REFERENCES

1. Amagasa M, Onuma T, Suzuki J. Surgical treatment of intracranial traumatic aneruysm [in Japanese]. *Surg Cereb Stroke* 1987;15:205–208
2. Larson PS, Reisner A, Morassutti DJ, et al. Traumatic intracranial aneurysms. *Neurosurg Focus* 2000;8:1–6
3. Bell RS, Vo AH, Roberts R, et al. Wartime traumatic aneurysms: acute presentation, diagnosis, and multimodal treatment of 64 craniocervical arterial injuries. *Neurosurgery* 2010;66:66–79
4. Cohen JE, Rajz G, Itshayek E, et al. Endovascular management of traumatic and iatrogentic aneurysms of the pericallosal artery. *J Neurosurg* 2005;102:555–557
5. Cohen JE, Gomori JM, Segal R, et al. Results of endovascular treatment of traumatic intracranial aneurysms. *Neurosurgery* 2008;63:476–486

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6. Horowitz M, Sharts M, Levy E, et al. Endovascular management of ventricular catheter-induced anterior cerebral artery false aneurysm: technical case report. *Neurosurgery* 2005;57:374
7. Medel R, Crowley RW, Hamilton DK, et al. Endovascular obliteration of an intracranial pseudoaneurysm: the utility of Onyx: technical note. *J Neurosurg Pediatr* 2009;4:445–448
8. Sim SY, Shin YS, Yoon SH. Endovascular internal trapping of pericallosal pseudoaneurysm with hydrogel-coated self-expandable coil in a child: a case report. *Surg Neurol* 2008;69:418–422
9. Yang TO, Lo YL, Huang YC, et al. Traumatic anterior cerebral artery aneurysm following blunt craniofacial trauma. *Eur Neurol* 2007;58:239–245
10. Cummings TJ, Johnson RR, Diaz FG, et al. The relationship of blunt head trauma, subarachnoid hemorrhage, and rupture of pre-existing intracranial saccular aneurysms. *Neurol Res* 2000;22:165–170
11. Komiyama M, Morikawa T, Nakajima H, et al. Early" apoplexy due to traumatic intracranial aneurysm case report. *Neurol Med Chir (Tokyo)* 2001;41:264–270
12. Sasaoka Y, Kamada K, Kenemoto Y, et al. Ruptured traumatic aneurysm of the peripheral anterior cerebral artery: study of delayed hemorrhage after closed head injury [in Japanese]. *No Shinkei Geka* 1997;25:337–344
13. Yokota H, Tazaki H, Murayama K, et al. Traumatic intracranial aneurysm—discussion about ninety-four cases including five of our own. *No Shinkei Geka* 1983;11:521–528
14. Yuge T, Shigemori M, Tokutomi T, et al. Diffuse axonal injury associated with multiple traumatic aneurysms of the distal anterior cerebral artery—case report. *Neurol Med Chir (Tokyo)* 1990;30:412–416
15. Benoit BG, Wortzman G. Traumatic cerebral aneurysms. Clinical features and natural history. *J Neurol Neurosurg Psychiatry* 1973;36:127–138
16. Dunn IF, Woodworth GF, Siddiqui AH, et al. Traumatic pericallosal artery aneurysm: a rare complication of transcallosal surgery. *J Neurosurg* 2007;106 (2 suppl pediatrics):153–157
17. Asari S, Nakamura S, Yamada O, et al Traumatic aneurysm of peripheral cerebral arteries. Report of two cases. *J Neurosurg* 46:795–803.
18. Egashira Y, Ikegame Y, Aki T, et al. Surgical treatment of a ruptured traumatic anterior cerebral artery aneurysm: report of two cases [in Japanese]. *Jpn J Neurosurg (Tokyo)* 2005;14:635–640
19. Senegor M. Traumatic pericallosal aneurysm in a patient with no major trauma. *J Neurosurg* 1991;75:475–477
20. Hsieh CT, Lin EY, Tsai TH, et al. Delayed rupture of pre-existing cerebral aneurysm in a young patient with minor head trauma. *J Clin Neurosci* 2007;14:1120–1122
21. Tokuno T, Ban S, Shingu T, et al. Traumatic anterior cerebral artery aneurysm difficult to distinguish from congenital cerebral aneurysm. *Case report [in Japanese] No Shinkei Geka* 1994;11:1073–1076
22. Opeskin K. Traumatic pericallosal artery aneurysm. *Am J Forensic Med Pathol* 1995;16:11–16

Investigation of Nasal Mobility in Asians and its Change After Alloplastic Augmentation

Rui Hou, MD, Dong Li, MD, Jie Yuan, MD, PhD, Liang Xu, MD, PhD, Jieneng Wu, MD, and Min Wei, MD, PhD

Abstract: Augmentation rhinoplasty is one of the most popular cosmetic surgeries in Asia, and both dorsal augmentation and mobile nose modifications are emphasized for the characters of the Asian nose and for Asians who are fond of the Western nasal shape. The goal in rhinoplasty is to create a balanced and natural appearance. Therefore, the cosmetic aspects, postoperative complications, and functional aspects should be regarded as a whole. In fact, the first 2 aspects had been widely reported in a large number of literatures. However, the functional aspects about mobility have been ignored by most surgeons. A sample method was used in this article to investigate the range of mobility of normal nasal tip in Asians and analyze the correlations of the nasal mobility and alloplastic materials. A total of 170 healthy adult subjects aged from 18 to 35 were included and were classified into 3 groups standing for normal nose and rhinoplasty with L-shaped silicon implants with e-PTFE implants. A 3D raster surface scanner was used to capture the images of the subjects accurately and rapidly in this detailed anatomical study. The range of nasal mobility (to right or left) in Asian normal women was 35.53 ± 4.84 degrees, and the length of mobile part was 16.62 ± 1.94 mm, which accounts for $40\% \pm 5\%$ of nasal length. The L-shaped alloplastic implants would reduce the nasal mobility, and there had been differences among the changes caused by different implants, which might cause unsatisfactory outcomes in patients. Results of the study indicated that augmentation rhinoplasty should be performed as well as augmented mammaplasty which attaches importance not only to the static and dynamic appearance of the nasal tip but also to the feeling when it is touched.

Key Words: Nasal mobility, rhinoplasty, silicon, e-PTFE

Rhinoplasty is one of the most popular cosmetic surgeries in Asia, and anatomical differences between the Asian nose and the Western nose have already been generally described in the literature. Typically, the Asian nose, which needs rhinoplasty, is characterized by a broad low dorsum, poor tip projection, a wide lobule, thick lobular skin with abundant subcutaneous fatty tissue, alar flaring, a retracted columella, and a small osteocartilaginous framework.[1,2] The lower lateral cartilages are often weak and thin, resulting in reduced support and projection. Therefore, Asian patients prefer rhinoplasty augmentation which can bring them a higher and narrower nasal dorsum, and more projected and distinct nasal tip.[2]

There are 2 components of augmentation: the rigid dorsal component and the mobile nasal component.[2] Both dorsal augmentation and the mobile nose modifications are emphasized for characters of the Asian nose. Therefore, an "L"-shaped implant, continuous from nasal dorsum to the mobile tip, is tailored to achieve satisfactory augmentation. Silicone and expanded polytetrafluoroethylene (e-PTFE, Tisuthes; SuoKang Medical Materials Co., Ltd., Shanghai, China) are the most commonly used alloplastic materials.[3] Despite the procedures and the materials, rhinoplasty operations have to consider functional outcomes as well as cosmetic results, which have been evaluated by photographs in many documents.[4]

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FIGURE 1. Diagram of nasal measurement.

FIGURE 2. Nasal shape of normal people when their nasal tip is being touched.

FIGURE 3. Nasal shape of the people who accepted augmentation rhinoplasty with L-shaped silicon implants when their nasal tip is being touched.

FIGURE 4. Nasal shape of the people who accepted augmentation rhinoplasty with L-shaped e-PTFE implants when their nasal tip is being touched.

However, the assessment of functional outcomes—the mobility of the nasal tip—is rarely reported in the English-language literature. Typical values of the mobile nose and a proper method to evaluate it are necessary. The purpose of this article is to use a simple method to investigate the range of mobility of normal nasal tip in Asians and analyze the correlation of the nasal mobility and alloplastic materials.

SUBJECTS AND METHODS

In this clinical trial, a total of 170 healthy adult subjects aged from 18 to 35 were included. They were all Chinese women and were classified into groups N, S, and P. Group N is composed of 100 subjects who had a normal nose and never underwent any nasal surgery. Group S comprised 40 subjects, and all the subjects had primary cosmetic rhinoplasty through vestibular incision with L-shaped silicon graft (WanHe; WanHe Plastic Materials Co., Ltd., Guangzhou, China). Group P had 30 subjects who underwent primary cosmetic rhinoplasty through vestibular incision with L-shaped e-PTFE graft (Tisuthes; SuoKang Medical Materials Co., Ltd., Shanghai, China). The subjects from group S and P were operated on from January 1, 2012 through December 31, 2012 and were followed periodically 3 to 14 months after surgery. All subjects had no complaints about the static nasal morphology or injection in this study.

All subjects were scanned using Axis Three 3D scanner (Axis Three XS-400; Axis Three, UK), and assessments were carried out on the raster scan image. To optimally reduce the variable pressure based on a swab, the maximum dynamic degree of the nasal tip was pushed by the same researcher with basically the same force. Outcomes were evaluated by both objective and subjective measures of cosmetic and functional (mobility) outcome. The following data were collected and analyzed: mobility of the nose, the length of the mobile part, the nasal length, and report of subjective outcome for group S and P at final evaluation.

Images were obtained according to standardized techniques of clinical photography. Before taking the images, hair covering the face and eyeglasses were removed. The subject sat up and was instructed to keep her eyes level with the horizontal line in a natural condition. For standard clinical image, a constant brightness and exposure were used. The distance between the subject and the scanner was 80 to 120 cm. The scanning area was from hairline to the thyroid cartilage. The subjects were demonstrated to hold a swab and put the tip to the position between the nasal tip and the edge of the right (or left) alar groove, give a hard squeeze at this region, and make the nasal tip have a maximum skewing, and then the image should be taken (Figs. 2, 3, 4).

Measurements were carried out on the images by the software Axis Three version 2.7.2 used to simulate plastic surgery procedures developed by Rose Bellingham etc. Along the dorsal midline, n and prn stand for the nasion and the nasal point, respectively, while m shows the turning point of the mobile nasal component. The angle α indicated the mobility of the nose. The length of d and the n-m length as well as the nasal length L was calculated. The proportion of d in L was calculated too. These measurements were carried out twice, and the average values were recorded (Fig. 1).

Subjective measures were carried out by a series of questionnaires for group N, S, and P, including whether the subject paid attention to the mobility of the nose or not and how the subject felt about it.

Statistical Analysis

Data of the measurements from group N, S, and P were expressed as the average value and standard deviations ($\bar{x} \pm s$), which were analyzed using one-way analysis of variance test. A P value of less than 0.05 was considered statistically significant (SPSS, version 16.0; SPSS, Inc.).

RESULTS

The objective outcomes are demonstrated in Table 1. Compared with group N, the nasal mobility of group S had a significant decline; the position of m moved up and extended the length of the mobile nose, resulting in an unnatural appearance. In some subjects,

TABLE 1. Measurements of the Nasal Mobility, the Length of the Mobile Part, and the Nasal Length Between Group N, Group S, and Group P

	Group N (n = 100)	Group S (n = 40)	Group P (n = 30)	*P* Value*		
	Mean SD	Mean SD	Mean SD	Groups N vs. S	Groups N vs. P	Groups P vs. S
α (degrees)	35.53 4.84	22.63 8.06	15.97 4.77	<0.0001 s	<0.0001 s	<0.0001 s
d (mm)	16.62 1.94	26.04 10.17	13.31 2.20	<0.0001 s	0.011 s	<0.0001 s
L (mm)	41.74 3.84	42.05 4.09	45.18 3.93	0.919	<0.0001 s	0.005 s
d/L	0.4 0.05	0.63 0.27	0.29 0.04	<0.0001 s	0.002 s	<0.0001 s

* Statistical analysis by one-factor ANOVA (post hoc: Scheffe) significance level (s) $P < 0.05$.

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an obvious activity of the whole silicone graft could be observed, making m overlap with n, which was one of the symptoms of an unnatural appearance. Compared with group N and S, group P had the minimum mobility of the nose. The position of m moved down, leading to a decrease in the proportion of the mobile nasal length. A rigid nasal tip could be found, and only the soft tissue from the nasal tip had a slight activity in some subjects. Images taken from the 3 groups are shown in Figures 2, 3, and 4.

In group N (n = 100), most subjects had awareness about the mobility of the nose (71%). When asked whether they care about the decline of the nasal activity caused by nasal rhinoplasty with alloplastic materials or not, the rate of YES was 26% versus 37% of NO, and the other subjects denoted no clear answer about that. Although no one required removal of the implant, there had been some complaints in group S and P. In group S (n = 40), 2 subjects were told about the decline of the nasal mobility (5%) preoperation. However, there were more subjects who realized the changing of the nasal mobility (27.5%). Some of them complained about the rigid and inflexible nasal tip and were therefore unsatisfied with the surgery (22.5%); subjects among them indicated that it was awkward to touch the nasal tip in public (15%), and one of them showed a serious attitude toward that variation leading to a lack of courage to rub the nose or blow it (2.5%). However, in group P (N = 30), these things seemed to be more serious, and the data were 3.33%, 56.67%, 26.67%, 30%, and 10% in order.

DISCUSSION

Nose, a conical organ located in the central position of the face, plays an important role in facial aesthetics. The defects of Asian nasal form and respects for the Caucasoid nasal shape are the main reasons for the popularity of nasal augmentation in Asia. There are 2 components that need to be augmented: the rigid dorsal component and the mobile nasal component. Nasal bones, frontal process of the maxilla, and the nasal process of the frontal bone comprise the framework that supports part of the nasal dorsum and makes it rigid and fixed. The other part of the nasal dorsum, however, consists of lateral nasal cartilage and alar cartilage, which have certain flexibility and mobility. Also, because of the abundant subcutaneous tissue, the nasal tip has more significant activity.

The goal in rhinoplasty is to create a balanced and natural appearance that the nose blends in with the face and essentially disappears. A perfect postoperative nose is one wherein no one notices it, which appear natural (look non-surgical) with good symmetry and appropriate length and rotation.[5] Therefore, the cosmetic and functional aspects and postoperative complications should be considered as one and regarded as a whole. In fact, there are a large number of literature reports about the cosmetic aspects and postoperative complications, whereas the functional aspects about mobility have been ignored by most surgeons. This can be reflected by the results in this study that most common women have a consciousness of nasal mobility (71%), but only a few subjects were told about the changing preoperation (5% and 3.33%), though inquiries about whether the postoperative nose has the same activity as a preoperative one or not were often presented by the ones who intend to have rhinoplasty. The following questions caused concern and were studied in this literature: (1) what is the range of normal nasal mobility?; (2) would alloplastic materials change the mobility or not, and what change could it bring about?; and (3) is there any difference among the changing caused by different implants?

A 3D raster surface scanner was used to capture the images of the subjects accurately and rapidly in this detailed anatomical study. Studying a 3-dimensional raster scanning image on the monitor is similar to handling a physical model of the nose which can be rotated or tilted to achieve the most suitable view for carrying out assessments. Also, the feature makes it far superior to the conventional 2D images and both qualitative and quantitative assessments can be carried out. The 3D images can make up for the deficiency of the conventional inaccuracy of 2D images.

Although autogenous augmentation is the gold standard in Caucasian patients, indications in Asians are mostly reserved for the patients explicitly requesting autogenous augmentation, for failed primary implant rhinoplasty, and for severe nasal deformities such as in Binder syndrome or in cleft lip patients.[6] Standard cantilever bone grafts or cartilage techniques have been found to provide inferior aesthetic results for reasons that bone grafts always undergo resorption and large cartilage grafts over the dorsum often warp over time.[7] They are therefore infrequently used. Alloplastic implant rhinoplasty is the preferential choice for Asian rhinoplasty, and the reasons are multifactorial: superior aesthetic results to autologous augmentation, acceptable price of implants, the straightforward procedure, no donor-site morbidity, relatively low complications, and it does not cut bridges for a salvage autologous rhinoplasty in case of complications.[7]

As we mentioned above, the mobility of the nasal tip is known to be an important part for functional rhinoplasty. In terms of this aspect, 2-piece rhinoplasty where the tip plasty with cartilage works and dorsal augmentation with alloplastic material seems better regarding the mobility of nasal tip compared with 1-piece type rhinoplasty. However, according to our follow-up, nasal mobility of 2-piece rhinoplasty with prosthesis and cartilage is the same as 1-piece rhinoplasty except for a natural soft tip. The reason is that prosthesis has to extend to the mobile nasal component so as to obtain a good dorsum radian. In addition, some patients are reluctant to take the cartilage. Technically, 2-piece operation is more complex than one. Besides an affected donor site, it is difficult to fix and easy to move. What is more, only a few experienced doctors can successfully implement it with a perfect shape. However, it is effective with the L-shaped prosthesis because it is accurate, easy to operate, does not require special fixing, achieves good effect after the operation, and the donor site is not impaired. The L-shaped implant provides both dorsum and tip augmentation. The columellar extension provides support for tip projection and can also provide volume when there is little columellar show. Thus, L-shaped implants such as silicon and e-PTFE seem to be the most commonly used implant shape.[8,9] This explains why both 2 implants with L shape were studied in the research.

The data in Table 1 could answer the questions posed above. The range of nasal mobility (to right or left) in Asian normal women was 35.53 ± 4.84 degrees, the length of mobile nose was 16.62 ± 1.94 mm which accounts for 0.4 ± 0.05 of nasal length, and point m is located in a lower position near the center of the dorsum. The L-shaped alloplastic implants changed the nasal mobility by reducing it, consistent with experiences of some surgeons.[2] In silicon implant group, the mobility declined about 12.9 degrees; nonetheless, the mobile nasal length increased over 9.4 mm, resulting in a weird dynamic appearance of unsuitable proportion.

The foot process of the silicon L-shaped implant usually rests between the medial crurae, and a deeply placed implant should benefit from having thicker soft tissue coverage; for tough periosteal tissue, it is hoped to provide better implant fixation and prevent undesirable migration.[10] However, the covering nasal periosteum was always torn at its periphery and disconnected from the surrounding bone. Therefore, lateral sides of the inserted silicones were not immediately covered with the periosteum, so the implant was not fixed and still had migration.[11] Although the peri-implant fibrous capsule will be formed, it cannot completely prevent the implant from sliding. As a result, a rigid nose with a mobile implant is generated.

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Data in Table 1 also indicated that there were differences among the changes caused by different implants. In e-PTFE implant group, the mobility declined to 15.97 ± 4.77 degrees and the mobile nasal length decreased to 13.31 ± 2.20 mm, so the nasal dynamic appearance was unnatural and the nasal tip was more rigid, although it was widely believed that it had a main advantage of creating a natural look in static. It might be caused by its characteristic of vascularization. In contrast to silicon, e-PTFE implants are less likely to form capsules and less likely to migrate, and healing is by tissue adhesion and not through encapsulation.[12] Thus, a rigid nose with a rigid implant is formed.

It was obviously demonstrated from the outcomes of subjective measures that a decline in nasal mobility was not accepted by most subjects (37% vs. 26%). A certain misunderstanding happened in the communication between surgeon and patient, which resulted in the complaints of unable to move the nasal tip (22.5% and 26.67%). Social activities would be interfered with less frequency of touching the nose (15% and 30%), and a serious attitude could be caused easier by e-PTFE than by silicon (10% vs. 2.5%). The outcomes implied a concept that has been ignored for a long time: augmentation rhinoplasty should be treated as well as augmentation mammaplasty which attaches importance not only to the appearance but also to the feeling when you touch it.

CONCLUSION

While requesting an adorable appearance, most patients in Asia want to conceal rhinoplasty surgery due to the conservative and shy characters. Therefore, a successful rhinoplasty should model a natural nasal appearance under static and dynamic. The mobile aspects should be considered as important as the cosmetic aspects and postoperative complications. Although suitable procedures and materials were not discussed, this literature provided the normal range of nasal mobility in Asian women and confirmed that the most popular L-shaped implants would reduce the nasal mobility, which could cause unsatisfactory outcomes with rhinoplasty in patients. Aesthetic surgeons might be implied that an estimate toward the nasal mobility of the patient should be done according to the normal range of it, and the patient ought to be informed about its changing; great importance would be attached to psychological feelings of patients for a better satisfaction of surgery. Further progresses are still needed to improve the rhinoplasty methods in the future.

REFERENCES

1. Sanchez JP, Balingit JS. Oriental rhinoplasty: a logical and systematic approach. Quezon City, Philippines: JMC Press Inc; 2000
2. Wong JK. Aesthetic surgery in Asians. *Curr Opin Otolaryngol Head Neck Surg* 2009;17:279–286
3. Ahn JM. The current trend in augmentation rhinoplasty. *Facial Plast Surg* 2006;22:61–69
4. Kim YH, Kim BJ, Jang TY. Use of porous high-density polyethylene (Medpor) for spreader or extended septal graft in rhinoplasty. *Ann Plast Surg* 2011;67:464–468
5. Kwon TK. Complications found in Asian tip surgery. *Facial Plast Surg* 2012;28:238–242
6. Bergeron L, Chen PK. Asian rhinoplasty techniques. *Semin Plast Surg* 2009;23:16–21
7. Daniel RK. Discussion: Silicone augmentation rhinoplasty in an Oriental population. *Ann Plast Surg* 2005;54:6–7
8. Tham C, Lai YL, Weng CJ, et al. Silicone augmentation rhinoplasty in an Oriental population. *Ann Plast Surg* 2005;54:1–5discussion 6–7.
9. Zhanqiang L. Carving Gore-Tex reinforced sheets for augmentation rhinoplasty in Chinese patients. *Plast Reconstr Surg* 2006;117:326–328
10. Tsai FC, Liao CK, Fong TH, et al. Analysis of nasal periosteum and nasofrontal suture with clinical implications for dorsal nasal augmentation. *Plast Reconstr Surg* 2010;126:1037–1047
11. Rojvachiranonda N, Pyungtanasup K, Siriwan P, et al. Cadaveric study of the nasal periosteum and implant position after augmentation rhinoplasty. *J Craniofac Surg* 2012;23:1163–1165
12. Yap EC, Abubakar SS, Olveda MB. Expanded polytetrafluoroethylene as dorsal augmentation material in rhinoplasty on Southeast Asian noses. *Arch Facial Plast Surg* 2011;13:234–238

Successful Conservative Management of a Large Congenital Scalp and Skull Defect

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Abstract: The authors report a case of a female baby born with large congenital scalp and skull defects with exposure of the cranial arachnoid mater.

The female patient was delivered by Caesarean section following diagnosis of a large cranial skin defect by ultrasound. The skin defect was present at the time of birth, measured 8.5 cm × 8.0 cm, and involved the full thickness of the cranium. The cranial arachnoid mater was exposed and veins running on the surface of the brain were observed.

There was no means to cover the large defect surgically and conservative treatment was initiated. Complete epithelialization of the scalp defect was acquired without fatal problems. In the course of treatment, Mepilex Transfer was very effective.

The repair of congenital defects of the scalp and skull is a major challenge not only because of the rarity but also because of severe morbidity. A surgical or conservative approach is chosen according to the situation. This rare case highlights two important clinical issues: that the congenital defects of the scalp and skull could not be managed with a surgical approach and in its place, a conservative approach using the special dressing Mepilex Transfer proved highly effective.

Key Words: Large scalp and skull defects, cleft palate, conservative treatment, Safetac technology, Mepilex Transfer

The repair of congenital defects of the scalp and skull is a major challenge not only because they occur rarely, but also because

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morbidity can be severe. There are various types of scalp and skull defects described in the literature, among them are aplasia cutis congenita, congenital calvarial giant foramina, atypical congenital calvarial defects, and amniotic bands. Perlyn's classification of scalp and skull defects as follows: Class I defects (open defects of the scalp alone), Class II defects (open defects of the skull alone), and Class III defects (open defects of both the skull and scalp).[1]

Here, we present a case of a female baby born with large congenital scalp and skull defects with exposure of the cranial arachnoid mater. The defect was 8.5 cm × 8.0 cm and classified as Perlyn's Class III, although it was atypical in terms of size. Accordingly, a decisive approach according to the classification was not chosen. In addition, cleft palate was also present. The management of this rare condition is discussed below.

CLINICAL REPORT

A female baby was delivered by Caesarean section following diagnosis of a large cranial skin defect by ultrasound. The mother was 23-years old (gravida 1, para 1). There was no maternal use of alcohol or drugs. Family history was unremarkable. The newborn infant weighed 2314 g and Apgar scores were 7 and 9 at 3 and 5 min, respectively. The skin defect was present at the time of birth, measured 8.5 cm × 8.0 cm, and involved the full thickness of the cranium. The cranial arachnoid mater was exposed and veins running on the surface of the brain were observed (Fig. 1A). There was a white, thin membrane believed to be the dura mater at the margin of the skin defect. There was no immediate leakage of cerebrospinal fluid (CSF). Three-dimensional computed tomography revealed dysplastic corpus callosum, and the overlying bones including the frontal, parietal, and temporal bones were missing (Fig. 1C–E). In physical examination, a hard and soft cleft palate was present (Fig. 1B). On the other hand, cardiovascular, musculoskeletal, respiratory, gastrointestinal, and genitourinary anomalies were not present.

There was no means to cover the defect surgically. Given there was no sign of CSF leakage, the lesion was dressed once a day with nonadhesive gauze with ointment to keep the wound moist and aseptic. Intravenous antibiotics and phenobarbital were administered to prevent infection and seizures.

On day 14, CSF leakage of was observed. Neurosurgeons placed a sheet of an absorbable reinforcement material made from polyglycolic acid, NEOVEIL (GUNZE, Kyoto, Japan), on several surface deficits (Fig. 1F). In addition, systemic antimicrobial treatment was started. On day 24, CSF leakage recurred and the same treatment was performed. The cause of the leakage was thought to be friction between the fragile surface of the defect and the gauze. It was necessary, therefore, to reduce the risk of bleeding from the fragile surface. As the patient often moved her head, leading to friction, we applied Mepilex Transfer (Mönlycke Health Care, Tokyo, Japan; Fig. 1G) to the scalp defect as an adhesive dressing, which had the advantages of covering a wide area, keeping the wound moist precisely, and reducing the friction of the movement with a cushion effect. Mepilex Transfer itself had Safetac Technology and was very gentle but adhesive on the surface of the wound. After Mepilex Transfer was applied, there was no further CSF leakage.

This conservative treatment was continued until complete epithelialization of the scalp defect on day 212 (Fig. 1H). To prevent direct insults to the wound, a special head-cap was made and attached. Long-term follow-up is necessary to observe development of the skull defect.

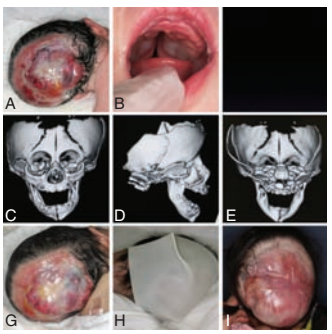

FIGURE 1. (A) Presentation at 4 days. Large scalp and skull defects and (B) hard and soft cleft palate. (C) AP view of three-dimensional computed tomography of the skull defect, including frontal, parietal, and temporal bones, (D) lateral view, (E) PA view. (F) Treatment using a patch of lyophilized dura mater to manage leakage of cerebrospinal fluid. (G) The scalp defect covered with Mepilex Transfer. (H) On day 212, epithelialization was achieved.

DISCUSSION

This rare case highlights two important clinical issues: that the congenital defects of the scalp and skull could not be managed with a surgical approach and in its place, a conservative approach using the special dressing Mepilex Transfer proved highly effective.

In regard to the first clinical issue, most of the several variations in congenital hard- and soft-tissue defects of the head (eg, aplasia cutis congenita, congenital calvarial giant foramina, atypical congenital calvarial defects, and amniotic bands) are usually incompatible with life as they are associated with brain anomalies, and they all differ in cause, size, and neurovisceral involvement. The few long-term follow-up studies in the literature present a limited number of cases on the whole. Therefore, the etiology remains unknown and there is no uniform treatment plan. Perlyn et al proposed a classification system based on the defect type present and introduced useful algorithms for care based on the class of the defect. Our patient had a Class III defect based on this algorithm.

Numerous reports have been published on the treatment of neonates born with congenital defects of the scalp.[2–5] Early surgical treatment is widely recommended because of fears of infection,[6,7] although a conservative approach is generally recommended for large defects.[2,8–10] Perlyn et al's algorithm advocates early coverage of the brain for scalp and skull defects >5 cm. Furthermore, it advises a different operation if <50% of the normal skull remains because there may not be enough autologous calvaria available for coverage. In this situation, a custom-fabricated plastic implant cranioplasty is preferred. Perlyn et al advocate a composite scalp flap (skin, subcutaneous tissue, and galea) to cover the soft-tissue defect, but in our case the skull defect was too large; it was impossible to elevate the composite scalp flap and perform a split-thickness skin graft on the pericranium over the flap donor site. There was no option to perform calvarial reconstruction surgically.

Hobar et al[11] showed in an animal model that when dura from an infant was replaced by that from a mature animal, bone regeneration no longer occurred. They concluded that infant dura had a critical osteoprogenitor function that dissipated with aging, and that the presence of local growth factors may contribute to this process. Any skin graft, especially one involving a defect too large for the graft to cover, entails a high risk of partial or total graft failure because of its size or an associated abnormality of the adjacent skin.[12] Furthermore, the graft might not expand sufficiently as the brain grows.[13] On the basis of these considerations, we chose conservative treatment without touching the remaining dura on the margin of the defect surgically and managed to obtain epithelialization of the scalp defect without any critical complications.

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The second clinical issue highlighted by the present case is that conservative treatment using a special dressing—in this case, Mepilex Transfer—was very effective. Mepilex Transfer is an adhesive dressing (Safetac technology) that transfers exudate into an outer absorbent dressing and it consists of flexible and hydrophilic polyurethane foam coated with a soft layer of silicone.[14] This dressing was very useful for covering the exposed cranial arachnoid mater and veins running on the surface of the brain. Slight friction was all that was needed to injure the brain and cause bleeding in this case, and the features of Mepilex Transfer, namely its softness and ability to decrease stress from the outside and adhere strongly, prevented friction because of head movement. In addition, Safetac technology made it possible to change dressings without the slightest damage to the tissue underlying the defect. As Mepilex Transfer is excellent at absorbing and passing exudate from tissue, controlling exudate from the tissue was easy.

Our patient also had a cleft palate. The palate was high and narrow, which is not associated with cardiovascular, musculoskeletal, respiratory, gastrointestinal, central nervous system, and genitourinary anomalies. Most cleft palates are isolated, but some are part of a syndrome, such as Pierre Robin sequence, Sticker syndrome, velocardiofacial syndrome, Treacher-Collins syndrome, or Van der Woudes syndrome. However, none of these syndromes are associated with defects of the cranial bone. Jones et al[15] reported a case of aplasia cutis congenita with cleft palate which also involved hypoplasia of the distal phalanges of the hands, ectrodactyly of the feet, and epidermolysis bullosa of the extremities and lower trunk. It is speculated that this patient is close to the notion of Frieden's classification group 9: aplasia cutis congenital associated with syndromes of malformation.[16]

REFERENCES

1. Perlyn CA, Schmelzer R, Govier D, et al. Congenital scalp and calvarial deficiencies: principles for classification and surgical management. *Plast Reconstr Surg* 2005;115:1129–1141
2. Muakkassa KF, King RB, Stark DB. Nonsurgical approach to congenital scalp and skull defects. *J Neurosurg* 1982;56:711–715
3. Argenta LC, Dingman RO. Total reconstruction of aplasia cutis congenita involving scalp, skull, and dura. *Plast Reconstr Surg* 1986;77:650–653
4. Vinocur CD, Weintraub WH, Wilensky RJ, et al. Surgical management of aplasia cutis congenita. *Arch Surg* 1976;111:1160–1164
5. Leboucq N, Montoya y Martinez P, Montoya-Vigo F, et al. Aplasia cutis congenita of the scalp with large underlying skull defect: a case report. *Neuroradiology* 1994;36:480–482
6. Schneider BM, Berg RA, Kaplan AM. Aplasia cutis congenita complicated by sagittal sinus hemorrhage. *Pediatrics* 1980;66:948–950
7. Sela M, Sahar A, Lewin-Epstein J. Agenesis of parietal bones with restoration of the cranial vault. Case report. *J Neurosurg* 1979;50: 674–676
8. Sharma AK, Kothari SK, Agarwal LD, et al. Agenesis of the skull bones. *Pediatr Surg Int* 2001;17:452–454
9. Basterzi Y, Bagdatoglu C, Sari A, et al. Aplasia cutis congenita of the scalp and calvarium: conservative wound management with novel wound dressing materials. *J Craniofac Surg* 2007;18:427–429
10. Santos de Oliveira R, Barros Juca CE, Lopes Lins-Neto A, et al. Aplasia cutis congenita of the scalp: is there a better treatment strategy? *Childs Nerv Syst* 2006;22:1072–1079
11. Hobar PC, Schreiber JS, McCarthy JG, et al. The role of the dura in cranial bone regeneration in the immature animal. *Plast Reconstr Surg* 1993;92:405–410
12. Saraiya HA. Management of aplasia cutis congenita of the scalp: a continuing enigma. *Br J Plast Surg* 2002;55:707–708
13. Kim CS, Tatum SA, Rodziewicz G. Scalp aplasia cutis congenita presenting with sagittal sinus hemorrhage. *Arch Otolaryngol Head Neck Surg* 2001;127:71–74
14. Kinoshita O, Nishimura T, Kawata M, et al. Vacuum-assisted closure with Safetac technology for mediastinitis in patients with a ventricular assist device. *J Artif Organs* 2010;13:126–128
15. Jones EM, Hersh JH, Yusk JW. Aplasia cutis congenita, cleft palate, epidermolysis bullosa, and ectrodactyly: a new syndrome? *Pediatr Dermatol* 1992;9:293–297
16. Frieden IJ. Aplasia cutis congenita: a clinical review and proposal for classification. *J Am Acad Dermatol* 1986;14:646–660

The Application of a Carbon Dioxide Laser in the Treatment of Superficial Oral Mucosal Lesions

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Abstract: To evaluate the safety and advantages of using carbon dioxide (Co_2) laser in the treatment of oral mucosal lesion, including vascular malformations, precancerous lesions, and verrucous nevus, a retrospective analysis was conducted for 73 patients with oral mucosal lesions who received Co_2 laser treatment during June 2012 to December 2013, including 25 patients with vascular malformations, 22 patients with oral leukoplakia, 18 patients with oral mucosal lichen planus, and 8 patients with oral mucosal and labial mucosal verrucous nevus. The lesions ranged from 0.8×0.8 cm to 4×3 cm. Twenty patients with lesions removed using the traditional scalpel, assisted with an electric knife, were the control group. The operative time was from 3 to 10 minutes, with an average of 5.5 minutes; the average amount of intraoperative bleeding was 5 mL. None of the 73 patients had postoperative infections, and all wounds healed well after the surgery. The patients were followed up for 1 year. Two patients with oral leukoplakia showed recurrence after the surgery, and a reoperation achieved satisfactory treatment effects. The operative time of the control group was in the range of 4 to 15 minutes, with an average of 9.5 minutes, and the average amount of intraoperative bleeding was 10 mL. None of the 20 patients had postoperative infections, and the wounds also showed healing well after the surgery. The application of Co_2 laser in the treatment of oral mucosal lesions has the

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advantages of reduced bleeding, a clear view during surgery, and a shorter operative time.

Key Words: Carbon dioxide laser, oral mucosal, surgery

Lasers have been applied in medicine for many years. They were first used in the fields of stomatology, dermatology, ophthalmology, and general surgery.[1–5] At present, laser therapy is widely used in various fields of medicine. The application of lasers in stomatology has a history of more than 40 years.[1] The carbon dioxide (Co_2) laser is the most commonly used laser in oral and maxillofacial surgery. The Co_2 laser is a gas laser and primarily exhibits water absorption. It has a small light spot diameter and a shallow depth of functioning, and its interaction with tissues is based on the thermal effect of the laser. The heat coagulation range on the edge of resection is only approximately 0.1 mm and is thus most suitable for fine and precise cuts.[6] Studies have found that using Co_2 laser for the ablation of oral precancerous lesions can effectively prevent lesion recurrence[7] and that the laser can be used in the treatment of oral mucosal vascular disease.[8] In recent years, we have used Co_2 laser for the treatment of superficial oral mucosal lesions and have achieved good results, as reported below.

MATERIALS AND METHODS

Patients

We selected 73 patients with superficial oral mucosal lesions who were treated in our hospital during the period of June 2012 to December 2013, including 25 patients with vascular malformations, 22 patients with oral leukoplakia, 18 patients with oral mucosal lichen planus, as well as 8 patients with oral mucosal and labial mucosal verrucous nevus. The age of the patients ranged from 2 years to 78 years, with a mean age of 54.6 years. There were 40 cases in the male patients and 33 cases in the female patients. The venous malformation lesions were in the range of approximately 1.0×1.0 to 4×3 cm; the precancerous lesions were in the range of approximately 0.8×0.8 to 3.0×2.5 cm; and the verrucous nevus lesions were in the range of approximately 0.5×0.8 to 2.0×2.5 cm. The lesions in all 73 patients were located in the oral mucosa as well as in the cheek, tongue, floor of the mouth, gums, palate, and vermilion. All patients were newly diagnosed patients and were without systemic diseases. We also selected 8 patients with vascular malformations, 10 patients with oral precancerous lesions, and 2 patients with labial mucosal verrucous nevus for the control group, with the lesions removed using the traditional scalpel assisted with an electric knife. There were 12 males and 8 females. During the surgery, routine monitoring of the blood pressure, pulse, and oxygen saturation levels of the patients was conducted.

Instruments and Equipment

An Acupulse 40WG optic fiber Co_2 laser treatment system from the Lumenis medical laser company (Lumenis Ltd, Yokneam, Israel) was used.

Surgical Methods

Primacaine was used for local infiltration anesthesia or general anesthesia with intubation. Upon the successful performance of anesthesia, the surgical margin was marked with methylene blue. A Co_2 laser (with a continuous output mode and a power of 5.0–7.0 W) was used to conduct surgical resection along the surgical margin. After the surgery, nonabsorbable suture was used to close the wound, or the wound was not sutured and was left to heal naturally.

Statistics and Follow-up

We recorded the operative time and blood loss for each patient. The operative time was from the initiation of surgical resection of the lesion to the completion of suturing. The suture was removed during reexamination 1 week after the surgery. The follow-up time was in the range of 1 to 24 months, with an average follow-up time of 12 months. The SPSS 13.0 statistical package was used for statistical analysis, and $P < 0.05$ indicated statistically significant differences.

RESULTS

All patients had satisfactory anesthetic effects as well as smooth and stable surgical processes. The statistical analysis showed that the Co_2 laser treatment group and the control group had no significant differences in the composition of the cases. For the 73 patients in the Co_2 laser treatment group, the operative time was in the range of 3 of 10 minutes, with an average of 6.5 minutes. The intraoperative bleeding was in the range of 2 to 8 mL, with an average blood loss of 5 mL. None of the 73 patients had postoperative infections. The patients were followed up for 1 to 24 months, with an average follow-up time of 12 months. During the follow-up period, all patients showed satisfactory wound healing. Two patients showed disease recurrence, which was identified during reexaminations at 8 months and at 10 months after the surgery, and the postoperative pathologic results of these 2 patients both revealed leukoplakia. These 2 patients with recurrence were again treated by Co_2 laser, which achieved satisfactory effects, and no recurrence was detected during follow-up. The operative time for the 20 patients in the control group was in the range of 4 to 15 minutes, with an average of 9.5 minutes. The intraoperative bleeding ranged from 7 to 20 mL, with an average of 10 mL. None of the 20 patients had postoperative infections, and the wounds also showed healing by the first check after the surgery. One patient experienced recurrence and was treated again using surgical resection. Compared with the control group, the operative time using the Co_2 laser for the treatment of superficial oral mucosal lesions was shorter (6.5 min vs 9.5 min, $P < 0.05$) and the amount of bleeding was less (5 mL vs 10 mL, $P < 0.05$), with statistically significant differences. Video 1 (see Supplemental Digital Content, Video 1, http://links.lww.com/SCS/A105) showed the process of resection of venous malformation in the tongue with Co_2 laser. Video 2 (see Supplemental Digital Content, Video 2, http://links.lww.com/SCS/A106) showed the process of resection of basal cell adenoma in buccal mucosa with Co_2 laser.

DISCUSSION

Among the existing types of medical high-energy laser light sources, the Co_2 laser is one of the lasers with the longest history of application.[6] Over the years, owing to its powerful capabilities of vaporizing biologic tissues and its precise cutting action, the Co_2 laser has been widely used in surgery and has achieved significant treatment effects. The Co_2 laser was invented in 1963 by Patel.[6] Since the 1970 s, the Co_2 laser has been widely used in the excision of various lesions, including oral squamous cell carcinoma, and has achieved good results.[7–10] The Co_2 laser has been used in surgery for more than 40 years, but for a long time, its main applications have consisted of superficial body tissues, organs, or visceral lesions in open surgery. The direct reasons for these limitations are the comparatively insufficient hemostatic properties of the Co_2 laser and the lack of suitable energy transfer tools with the necessary

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flexibility; the fundamental reason for its use is the characteristic effects of Co_2 laser on tissues, which are determined by its inherent wavelength. The Lumenis Co_2 laser treatment system uses optic fibers with good flexibility and energy transmission, which effectively solves the Co_2 laser energy conduction issue.

Lasers have been applied in oral and maxillofacial surgery for many years.[1] It is well known that the oral and maxillofacial areas have rich blood supply and complex structures; the lesions are often located in small cavities, with limited space for operation, making surgical procedures more difficult. The Co_2 laser can be used to remove oral and maxillofacial lesions as well as head and neck lesions. Because the Co_2 laser can be focused to become a small light spot with a diameter of only approximately 0.1 to 0.2 mm, it is possible to conduct fine vaporization, cutting, or repair of diseased tissues while also obtaining a satisfactory hemostatic effect and fewer complications.[7,11] We have used a Co_2 laser with a continuous output mode to treat superficial lesions on the oral mucosa, including the excision of vascular malformations and precancerous lesions. Compared with the surgical approach of using a traditional scalpel assisted with an electric knife, the operative time of the Co_2 laser treatment is significantly shorter and the intraoperative bleeding is reduced. We analyzed the reasons, and we believe that, first, Co_2 laser treatment can precisely cut the lesions by being focused into a light spot, thus preventing both insufficient removal and excessive removal; second, while the Co_2 laser cuts the lesions, the laser simultaneously conducts hemostasis of the capillaries at the site of excision. Therefore, the application of the Co_2 laser for the excision of oral mucosa permits a clear view of the scope of the excised diseased tissues, reducing the operative time. Laser surgery can also shorten the length of the hospital stay. It has been reported that, when treating oral soft tissue lesions, laser and conventional surgery do not differ significantly in the effects of heat-mediated tissue damages on diagnosis, intraoperative and postoperative pain control, as well as postoperative complications.[12] Upon lesion recurrence, laser treatment can still be selected. Our cases showed that patients with recurrence still obtained satisfactory results by repeating the same laser treatments.

In summary, we believe that the Co_2 laser ablation of superficial oral mucosal lesions is safe, with reduced blood loss and a shorter operative time, and is suitable for clinical application.

REFERENCES

1. Roodenburg JL, Witjes MJ, de Veld DC, et al. Lasers in dentistry 8. Use of lasers in oral and maxillofacial surgery. *Ned Tijdschr Tandheelkd* 2002;109:470–474
2. Anderson RR. Laser medicine in dermatology. *J Dermatol* 1996; 23:778–782
3. Brauer JA, Gordon Spratt EA, Geronemus RG. Laser therapy in the treatment of connective tissue diseases: a review. *Dermatol Surg* 2014;40:1–13
4. Sherwood LA, Knowles G, Wilson RG, et al. Retrospective review of laser therapy for palliation of colorectal tumours. *Eur J Oncol Nurs* 2006;10:30–38
5. Linke SJ, Steinberg J, Katz T. Therapeutic excimer laser treatment of the cornea. *Klin Monbl Augenheilkd* 2013;230:595–603
6. Patel CKN. Continuous-wave laser action on vibrational-rotational transitions of CO2. *Phys Rev* 1964;136:1187–1193
7. Jerjes W, Hamdoon Z, Hopper C. Co2 lasers in the management of potentially malignant and malignant oral disorders. *Head Neck Oncol* 2012;4:17
8. Ma LW, Levi B, Oppenheimer AJ, et al. Intralesional laser therapy for vascular malformations. *Ann Plast Surg* 2014;73:547–551
9. Canis M, Ihler F, Martin A, et al. Transoral laser microsurgery for T1a glottic cancer—a review of 404 cases. *Head Neck* [published ahead of print March 13, 2014] doi: 10.1002/hed.23688
10. Prgomet D, Bacić A, Prstacić R, et al. Complications of endoscopic Co_2 laser surgery for laryngeal cancer and concepts of their management. *Coll Antropol* 2013;37:1373–1378
11. White JM, Chaudhry SI, Kudler JJ, et al. Nd:YAG and Co_2 laser therapy of oral mucosal lesions. *J Clin Laser Med Surg* 1998; 16:299–304
12. Tuncer I, Ozçakir-Tomruk C, Sencift K, et al. Comparison of conventional surgery and CO2 laser on intraoral soft tissue pathologies and evaluation of the collateral thermal damage. *Photomed Laser Surg* 2010;28:75–79

Sudden Hearing Loss: A Study of Prognostic Factors for Hearing Recovery

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Objective: Sudden hearing loss (SHL) is a disease, isolated or associated with vertigo, of unknown etiology. The aim of this study was to identify the prognostic factors for hearing recovery.
Methods: In this retrospective study, we analyzed 287 cases of SHL (mean age 42). Tonal threshold audiometry, BAEP, and RM were the diagnostic procedures for clinical balance of the patient. The therapy used the following: corticosteroids, osmotic diuretic infusion, and vasoactives. All the factors were evaluated through statistical tests, Spearman test, and linear logistic regression.
Results: In our study, we observed that 39.3% of patients improved, 27.9% remained unchanged, 29% had complete recovery, and 3.8% worsened. Therapy was not related to the degree of recovery from hearing loss, whereas young age, delayed diagnosis, and audiometric curve type were the three factors related to the degree of hearing recovery.
Conclusions: In our study, we report a better overall recovery rate compared with spontaneous recovery. In particular, early treated patients as well as patients with upsloping hearing loss frequently recovered after treatment. Age, time between onset and treatment, and audiogram type were shown to be significantly related to outcome.

Key Words: Sudden hearing loss, treatment, outcome, delay, age, recovery

Sudden hearing loss (SHL) is a condition that often affects the patients' relationship with other people, considered as serious disability, as its hearing loss may involve frequencies between 500

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and 2000 Hz, and its symptoms may include heavy deafness and tinnitus.[1–10]

Moreover, it represents a frequently encountered condition that often leads to hospital admission; it is also a good indicator of the Health education level and organizational efficiency of the health services (suspect of pathology, access to diagnostic structures, timeliness of intervention, and other factors).

The prevalence of SHL has been reported to be one in 3000 inhabitants, representing about 1% of all sensorineural hearing losses.[1–15] As for age, 75% of patients affected by this disease are more than 40 years (60% of patients are between 40- and 70-years old).[1–15]

Sudden hearing loss can affect males or females without noticeable differences of incidence between genders.

A great number of patients with SHL spontaneously recover; furthermore, some patients with partial SHL often tend to undervalue the symptoms and they do not go to hospital.[10–15]

As for etiology, several mechanisms, either isolated or combined, are believed to damage the cochlea, thus provoking SHL (infective, vascular, immunological, membranous breakage theory, and other factors), although the exact cause of most cases remains unknown.

Therefore, as SHL is usually idiopathic, its treatment still remains empirical and controversial. In fact, nowadays, no therapeutic standard protocol is universally accepted as the most effective in the treatment of this disease. In fact, in literature, several treatment protocols have been proposed, including vasodilators to improve hematic flow, corticosteroids for their antinflammatory immunomodulating and antiedema effects, hyperbaric oxygen therapy for cochlear vasodilatations, anticoagulants to reduce hematic viscosity, antiaggregating drugs, osmotic diuretics to reduce endolymphatic pressure, antivirals (when a viral origin is suspected, in particular the herpes virus), or rest in bed.[1–15]

Finally, there is no consensus about which factors can have a prognostic value for hearing recovery.

Therefore, the aim of this article was to present our experience in the management of SHL and to identify prognostic factors for hearing recovery in our study population.

MATERIALS AND METHODS

A retrospective chart review was performed to identify patients referred to our Department for Sudden Hearing Loss. Only patients presenting with the specific complaint of SHL were reviewed. Sudden hearing loss was defined as at least a 30-dB sensorineural hearing loss, occurring in at least 3 subsequent frequencies over a period shorter than 3 days.

The charts of all patients admitted with the diagnosis of SHL between 2001 and 2005 were reviewed.

The following data were collected for each patient: age, gender, thorough medical history, delay of presentation and diagnosis, initial and posttreatment pure tone audiometry (PTA), and MRI or auditory brainstem response (ABR).

All patients had denied having had any prior episodes of SHL.

All patients had provided a complete medical history and they had undergone physical and laboratory examinations as well as audiologic evaluations.

During the initial hospital visit, a PTA examination had been performed for each patient.

In addition to PTA, all patients had undergone an MRI for the purpose of excluding possible retrocochlear lesions, or an ABR testing if MRI was contraindicated or refused by the patient.

Initial pretreatment audiograms were categorized into 4 sensorineural types: upsloping (a "rising curve" involving a threshold shift for low-medium frequencies), downsloping (a "falling curve" coincided with a threshold shift for medium-high frequencies, such as 8 kHz), mid-frequency (a U-shaped curve, suggesting for a mid-frequency loss), and profound loss (a flat audiogram with a threshold shift greater than 90 dB in all frequencies).

Patients affected by idiopathic SHL who had agreed to be hospitalized and that had been followed-up after being discharged were considered.

All patients were hospitalized. Then, the treatment protocol was instituted. Three different therapeutic protocols were administered. The first group of patients were administered osmotic diuretic (250 mL of saline solution with mannitol/glycerol 18%) and corticosteroids (betamethasone disodium phosphate 4 mg in F.S. 100cc x i.v.); the second group was given osmotic diuretic, corticosteroids, and antiaggregating drugs; and the third group received osmotic diuretic and antiaggregating drugs. Treatment protocols including antiaggregating drugs (second and third groups) were administered to patients affected by cardiovascular comorbidities with tromboembolic risks or diabetes.

Siegel's criteria were used to assess the recovery outcome of the patients in each group as follows: "no improvement" (patients showing <15 dB gain); "slight improvement" (patients with 1–15 dB gain and final hearing poorer than 45 dB); "partial improvement" (patients showing >15 dB gain and final hearing level between 25 and 45 dB); and "complete improvement" (patients whose final hearing level was >25 dB regardless of the amount of gain). For statistical reasons, we divided our study members into 2 groups, the first with no or slight improvement and the second with partial or complete improvement.

To evaluate the existence of a relation of causality between a factor of exposure and the recovery from the disease, the x^2 *test* was done, and x^2 *for trend test* among the classes of variables. To analyze the relation between the variables and the event expressed as dichotomic variable (0 and 1, or sick and recovered), a model of linear logistic regression was used. The association was expressed in terms of Odds Ratio.

RESULTS

Between 2001 and 2005, 330 patients reporting SHL were referred to our department for assessment.

Following the performance of MRI or ABR, 43 cases were excluded with the diagnosis of Ménière's disease, vestibular schwannoma or neurinoma of the VIII cranial nerve, neurovascular conflict, malformation, vasculopathy, or other expansive lesion.

Therefore, 287 patients with idiopathic SHL were included in the study. On the whole, 130 subjects (45%) were male and 157 (55%) were female. The right ear was involved in 43% of patients and the left ear in 57%.

The mean age of the study members was 49 years, with a range of 18–85 years. In particular, according to age, 4 groups were identified: less than 20 years (4 patients, 1.4%), between 21 and 45 years (99 cases, 34.5%), between 46 and 60 years (90 patients, 31.3%), and over 60 years (94 cases, 32.7%).

On the whole, 75 patients (26.5%) presented comorbidities: arterial hypertension (35 patients), ischemic cardiopathy (24 patients), and diabetes (16 patients).

The delayed diagnosis, that is the time between the patient's awareness of the disease and the diagnosis with the subsequent start of therapy, was classified according to 5 classes, as in Table 1.

Table 1 lists the characteristics of the study population.

As for the audiometric configuration of SHL, the most common shape was a flat profound loss audiogram (48%), followed by an upsloping curve (32%), a downsloping configuration (14%), and U-shaped curve (6%).

On the whole, 73.5% of patients were treated according the first group protocol (osmotic diuretics and corticosteroids), the 14.3% of

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TABLE 1. Characteristics of the Study Population

Characteristic		Number of Patients	Percentage
Age	Mean value	49	
	Range	18–85	
Gender	Male	130	45.3%
	Female	157	54.7%
Audiometric configuration of SHL	Profound loss audiogram	138	48%
	Upsloping curve	92	32%
	Downsloping curve	17	14%
	U-shaped curve	40	6%
Type of treatment	DO + CS	212	73.9%
	DO + CS + AA	41	14.3%
	DO + AA	34	11.8%
Delayed diagnosis	0–2 days	52	18%
	3–7 days	120	41%
	8–15 days	43	15%
	16–30 days	29	10%
	More than 30 days	43	15%
Comorbidities	Arterial hypertension	35	
	Ischemic cardiopathy	24	
	Diabetes	16	

AA, antiaggregants; CS, corticosteroids; DO, diuretics osmotics; SHL, sudden hearing loss.

patients received osmotic diuretics, corticosteroids, and antiaggregating drugs (second group), and the 12.2% of patients were administered osmotic diuretics and antiaggregating drugs (third group).

Finally, according to treatment outcomes, the following results were obtained: 3.8% of patients showed "no improvement"; 27.9% reported "slight improvement"; 39.3% of subjects were considered in the "partial improvement" group; and the remaining 29% of the study population showed a "complete improvement."

Then, 4 variables were analysed in relation to treatment outcomes, dividing the patients into the 2 aforementioned groups (no or slight improvement; partial or complete improvement): patient's age, delayed diagnosis, typology of audiometric curve, and pharmacological therapy.

As for age, there appeared to be a clear relation between age and the possibility of functional recovery. There was a positive trend inversely proportional to ageing. The classes of age with significantly greater frequency of recovery were those of patients with an age lower or equal to 45 years (x^2 *for trend* = 10.9; $P = 0.004$). The Odds Ratio from the regression analysis ($OR = 0.58$; $IC = 95\%$; 0.36–0.93) was used to evaluate age increase in comparison with recovery probability: the value obtained showed that younger age classes seem to have higher recovery probability, in fact there was no association between age increase and improvement.

The delayed diagnosis variable, compared with the final result, showed that the percentage of improvement seemed to progressively decrease together with the increase of days spent by the patient to reach awareness and to contact a specialist (89% within the first 2 days, up to 41% after 30 days of delayed diagnosis). In this case too, from statistic analysis, a significant relation between delayed diagnosis and final result was found, and it was represented by the value x^2 *for trend* = 7.26; $P = 0.002$. The Odds Ratio ($OR = 0.57$; *IC* 95% 0.42–0.77) confirmed that there was no direct association between an increase in delayed diagnosis and improvement, but a negative trend with the increase of diagnosis time appeared to be evident.

As for the analysis of the morphology of audiometric curve, improvement occurred in 58% of cases for pantonal hearing losses, 71% for hearing losses on acute tones, and in 80% for hearing losses on low tones. In the relation between hearing loss on low tones and improvement, the significance emerged from *test* x^2 *value* $P = 0.001$, $OR = 5.0$; *IC* 95% 1.90–13.2.

Finally, as for pharmacological therapy in groups 1 and 3 the percentages of improved patients appeared to be higher, whereas in the patients that were administered the complete association of drugs, a noticeable improvement was found in just 50% of cases. However, statistical analysis revealed a $P = 0.29$, underlining no significance in data, and logistic regression ($OR = 1.02$; *IC* 95% 0.94–1.10) confirmed the absence of effective relevance.

x^2 *Test* was finally applied to all the variables, comparing fully recovered patients to all the others; statistically significant associations were observed between a full hearing recovery and the following variables: timing of diagnosis ($P = 0.001$), and audiometric curve type ($P = 0.000$).

DISCUSSION

Despite the limits represented by differences in methods (low number of patients, non-homogeneous groups), the trend of all the study is to consider SHL as a pathology of difficult assessment because despite extensive evaluation an etiology can only be found in 10%–15% of patients.[13] Furthermore, no standard definition or treatment protocol has been accepted.[13–15]

However, most of the literature deals with SHL addressing the problem of its treatment, whereas few articles are focused on prognostic factors influencing the likelihood of patients' hearing being restored.[1–9]

As for treatment of SHL, as aforementioned there is no consensus. The high spontaneous recovery rate is a confounding issue too: in fact, most studies report a spontaneous recovery in 45%–65% of patients.[1–15] Nevertheless, steroids have become widely accepted for the management of SHL on the basis of their antiinflammatory effect.[13–15]

The studies on idiopathic SHL had limited success in providing a standard definition or treatment protocols, partly because of the low incidence in the routine otologic caseload, the lack of etiologic specification, and the very same high spontaneous recovery rate.[13–15]

Overall, our treatment protocol allowed us to obtain a slight improvement of hearing in 27.9% of cases, a partial improvement in 39.3% of patients, and a complete improvement in 29% of the study population, with a whole recovery rate that is above the spontaneous recovery rate.

In literature, some authors support the utility of antivirals, while others consider them ineffective.[1–15] The same discordance of results concerns many other treatments, from the use of vasodilators and drugs which act on hematic viscosity, to the use of carbogen, to hyperbaric oxygen therapy.[1–15]

Concerning the use of mannitol, there are different opinions.[1–15] Recently, preliminary studies have applied transtympanic steroid instillations, but the results are not completely demonstrable.[1–15]

In the few previous articles about prognostic factors of hearing recovery in SHL, there is no unanimous consensus on which factors could have a predictive value on prognosis of SHL.

As for prognostic factors, we found significant associations were observed between a full hearing recovery and the variables "timing of diagnosis" and "audiometric curve type." We also demonstrated that patients younger than 45 have a higher recovery probability. Therefore, timing of diagnosis and the type of audiometric curve morphology seem to be significant prognosticators of recovery in our study, but the young age may have a role too.[1–15]

Time from onset of hearing loss to presentation is extremely significant in prognosis. Most studies have demonstrated that presentation after 7 to 10 days results in poor outcome. It has been

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proposed that this is because of a self-selection process, whereas those who recover quickly do not seek medical care.[13–15]

As for audiometric curve type, a statistically significant association was found between an upsloping curve (with a threshold shift for low-medium frequencies) and improvement.

We did not observe a significant advantage in treating patients with a profound pantonal hearing loss (flat curve) because the recovery rate of 58% was comparable with a spontaneous recovery rate.[13–15]

Instead, as in our study population, upsloping audiograms have already been reported to have a better prognosis for recovery.[13–15]

Finally, our statistical analysis revealed no significant differences between patients stratified according to different treatment protocols. At the time of study, the protocol used in our division usually provides a politherapy with betametasone sodium phosphate 4 mg in F.S. 100cc x i.v.+ mannitol 18% 250cc x i.v.+ sulodexide 600 uls/ 2 mL x 2 x i.v. The administration of the drugs was done during hospitalization, to oblige the patients to rest and to isolate the ear from environmental noises.

CONCLUSIONS

In our study population, we report a better overall recovery rate compared with spontaneous recovery. This is comparable to most prior studies that used steroid as a treatment.[7,9,10] In particular, early treated patients as well as patients with upsloping hearing loss frequently recovered after treatment.

Age, time between onset and treatment, and audiogram type were shown to be significantly related to outcome.

Further prospective studies are needed to evaluate the efficacy of each component of the treatment regimen, in addition to the effect of changing steroid dosages and length of use. The necessity for a therapeutic protocol universally accepted clearly emerges, as well as the necessity for more in-depth studies on the physiopathology of nervous transmission and cochlear neuropharmacology, patients' selection, standardization of treatment, and of result evaluation.

REFERENCES

1. Lee HS, Lee YJ, Kang BS, et al. A clinical analysis of sudden sensorineural hearing loss cases. *Korean J Audiol* 2014;18:69–75
2. Bear ZW, Mikulec AA. Intratympanic steroid therapy for treatment of idiopathic sudden sensorineural hearing loss. *Mo Med* 2014;111: 352–356
3. Bogaz EA, Suzuki FA, Rossini BA, et al. Glucocorticoid influence on prognosis of idiopathic sudden sensorineural hearing loss. *Braz J Otorhinolaryngol* 2014;80:213–219
4. Wittig J, Wittekindt C, Kiehntopf M, et al. Prognostic impact of standard laboratory values on outcome in patients with sudden sensorineural hearing loss. *BMC Ear Nose Throat Disord* 2014;14:6
5. Suzuki H, Tabata T, Koizumi H, et al. Prediction of hearing outcomes by multiple regression analysis in patients with idiopathic sudden sensorineural hearing loss. *Ann Otol Rhinol Laryngol* 2014;123: 821–825
6. Lionello M, Staffieri C, Breda S, et al. Uni- and multivariate models for investigating potential prognostic factors in idiopathic sudden sensorineural hearing loss. *Eur Arch Otorhinolaryngol* [published online ahead of print March 25, 2014] doi: 10.1007/s00405-014-2992-8
7. Wen YH, Chen PR, Wu HP. Prognostic factors of profound idiopathic sudden sensorineural hearing loss. *Eur Arch Otorhinolaryngol* 2014;271:1423–1429
8. Park KH, Lee CK, Lee JD, et al. Combination therapy with systemic steroids, an antiviral agent, anticoagulants, and stellate ganglion block for treatment of sudden sensorineural hearing loss. *Korean J Audiol* 2012;16:71–74
9. Jun HJ, Chang J, Im GJ, et al. Analysis of frequency loss as a prognostic factor in idiopathic sensorineural hearing loss. *Acta Otolaryngol* 2012;132:590–596
10. Enache R, Sarafoleanu C. Prognostic factors in sudden hearing loss. *J Med Life* 2008;1:343–347
11. Ceylan A, Celenk F, Kemaloğlu YK, et al. Impact of prognostic factors on recovery from sudden hearing loss. *J Laryngol Otol* 2007;121:1035–1040
12. Narozny W, Kuczkowski J, Kot J, et al. Prognostic factors in sudden sensorineural hearing loss: our experience and a review of the literature. *Ann Otol Rhinol Laryngol* 2006;115:553–558
13. Zadeh MH, Storper IS, Spitzer JB. Diagnosis and treatment of sudden-onset sensorineural hearing loss: a study of 51 patients. *Otolaryngol Head Neck Surg* 2003;128:92–98
14. Dispenza F, De Stefano A, Costantino C, et al. Sudden sensorineural hearing loss: results of intratympanic steroids as salvage treatment. *Am J Otolaryngol* 2013;34:296–300
15. Psifidis AD, Psillas GK, Daniilidis JCh. Sudden sensorineural hearing loss: long-term follow-up results. *Otolaryngol Head Neck Surg* 2006;134:809–815

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