A Rare Cause of Horner Syndrome: Pediatric Lipoblastoma of the Neck

To the Editor: Less than 10% of soft tissue neoplasms in the first and second decades of life have an adipose phenotype, and most are benign. The most common are various types of lipoma and lipoblastoma.[1] Lipoblastomas originate from embryonal fat tissue. Even if biologically benign, these neoplasms can be difficult to resect completely because of their undefined margins. The recurrence rates are between 14% and 24%. In the literature, there are few published case series of lipoblastoma/lipoblastomatosis.[2–4] Lipoblastomatosis has been defined as the diffuse multifocal type of lipoblastoma.[1] The majority of these tumors occur superficially in the subcutaneous tissues of the extremities and trunk and rarely at other sites including the face, scrotum, parotid gland, and mediastinum.[3] In the head and neck, these lesions are extremely rare.[4] Histologically, lipoblastomas are now categorized into two types, the encapsulated and diffuse types.[4] Cervical lipoblastoma typically presents as a rapidly enlarging, painless neck mass. Symptoms, however, can occur from compression of cervical structures, including Horner syndrome, hemiparesis, and respiratory compromise.[5–8] Diagnosis is often suggested by CT and MRI findings but can only be confirmed by microscopic examination.[6] The recommended treatment is complete surgical excision.[9]

An 8-month-old girl presented with a 2-month history of an enlarging, painless, left side neck mass and at the same side drooping of the eyelid (Figs. 1A, B). There was no history of trauma, infection, dysphagia, dyspnea, stridor, or fever. Medical history was unremarkable. Physical examination revealed a well-developed and well-nourished child with a semi-mobile, rubbery mass, approximately 5 cm in diameter, located in the left anterolateral side of the neck, starting 2 cm inferiorly of the angulus mandibulae and extending along the sternocleidomastoid (SCM) muscle. It was nontender and nonerythematous. At the same side with mass, left eye had ptosis, myosis, and enopthalmos. MRI research demonstrated a well-defined $5.3 \times 3.5 \times 3.7$-cm mass in the anterolateral region of the neck with anterior displacement of the carotid artery and compression of the trachea and esophagus (Fig. 2). Mass was isointense on T1-weighted images and hyperintense on T2-weighted images. Fine-needle aspiration biopsy revealed fibroadipose tissue.

Surgical excision via a lateral cervical approach demonstrated a well-encapsulated, soft, yellowish-white mass deep to the SCM muscle. The mass was successfully dissected away from the prevertebral fascia and cervical sympathetic ganglion chain and lung apex at the inferior (Fig. 3). Pathological evaluation of the tumor demonstrated immature adipocytes surrounded by myxoid material, and lobulation characteristic of lipoblastoma (Fig. 4).

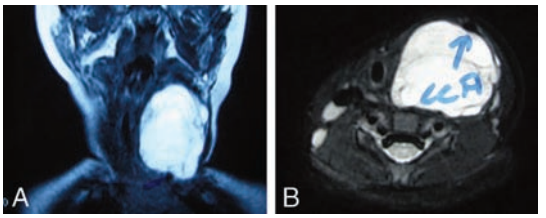

FIGURE 2. A, MRI T1 sequence, showing a well-defined $5.3 \times 3.5 \times 3.7$-cm mass in the anterolateral region of the neck with anterior displacement of the carotid artery and compression of the trachea and esophagus. B, The arrow points to the left common carotid artery.

At 2 years of follow-up, the child is healthy and developing normally without recurrent tumor. Ptosis, myosis, and enopthalmos of the left eye completely healed.

Lipoblastomas are rare benign tumors arising from embryonal white fat cells typically found in the axilla, mediastinum, retroperitoneum, prevertebral areas, and extremities, with only few cases reported in the neck (6). Most of them occur before 3 years of age.[7] Lipoblastomas can present anywhere in the body, but most of the lipoblastomas occur mainly in the extremities and the trunk and have rarely been described in the head and neck area. In the head and neck area, cervical region was the most common location, and the parotid gland, the cheek, the skin, and the orbit were also reported.

Lipoblastomas of the neck are frequently asymptomatic.[2] The most common presentation of cervical lipoblastoma is a rapidly growing, painless mass. However, in some cases, as the tumor became larger, it could compress the adjacent structures and cause specific symptoms such as respiratory insufficiency, Horner syndrome, and weakness of the upper extremity.[6]

According to the review of 48 cervical lipoblastoma cases from 1982 to 2009, Pham et al reported that the most common presenting symptoms for neck lipoblastomas were painless enlarging neck mass (53%) and respiratory distress (12%).[4] They found out 1 case presenting with Horner syndrome.[5] O'Donnell et al also reported a lipoblastoma case with Horner syndrome.[10]

CT and MRI can be used to identify lipoblastomas. MRI may be chosen because there is no radiation and the mass can be seen with high signal intensity in T1- and T2-weighted images. But the definitive diagnosis is always pathological examination of the mass.

We did not have any recurrence so far in this case, but local recurrences up to 25% are reported.[11]

When treating head and neck masses in children, we must always keep in mind that differential diagnosis is crucial. Cystic hygroma/lymphangioma, vascular anomalies, mature lipoma, liposarcoma, lipofibroma, and thyroglossal duct cysts are to be considered and differentiated in pediatric masses.

This case shows us that although lipoblastoma is a rare and pathologically benign tumor, it is important to consider it in differential diagnosis of neck masses due to its critical anatomical localization. Lipoblastoma is better known in the last 15 years, but

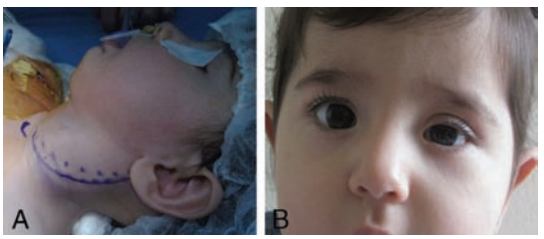

FIGURE 1. A and B, Left side neck mass with ptosis and myosis on the same side.

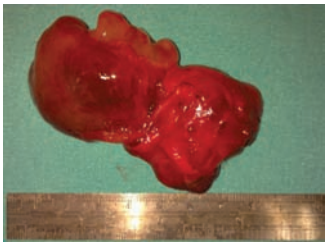

FIGURE 3. Lipoblastoma surgically excised.

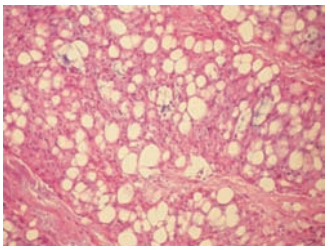

FIGURE 4. Specimen demonstrated immature adipocytes surrounded by myxoid material, and lobulation characteristic of lipoblastoma.

recognizing it as a cause for Horner syndrome is rare and important. This should be kept in mind when diagnosing pediatric neck masses.

Denizhan Dizdar, MD
Gaziosmanpaşa Taksim Training and Research Hospital
Department of Otorhinolaryngology
Istanbul, Turkey
denizhandizdar@hotmail.com

Senem Kurt Dizdar, MD
Asli Batur Çaliş, MD
Berna Uslu Coşkun, MD
Hamidiye işli Etfal Training and Research Hospital
Department of Otorhinolaryngology
Istanbul, Turkey

The authors report no conflicts of interest.

ACKNOWLEDGMENT

A written informed consent from the patient's parents is obtained for the use of photos in this article.

REFERENCES

1. Coffin CM, Alaggio R. Adipose and myxoid tumors of childhood and adolescence. *Pediatr Dev Pathol* 2012;15:239–254
2. Torre M, Borel C, Saitua F, et al. Lipoblastoma with unique localization requiring tracheal and esophageal resection. *J Pediatr Surg* 2010;45:e21–e23
3. Al-Salem AH, Al-Nazer M. Mesenteric lipoblastoma in a 2-year-old child. *Pediatr Surg Int* 2003;19:115–117
4. Pham NS, Poirier B, Fuller SC, et al. Pediatric lipoblastoma in the head and neck: a systematic review of 48 reported cases. *Int J Pediatr Otorhinolaryngol* 2010;74:723–728
5. Gammelgaard N, Jorgensen K, Lund C. Benign lipoblastoma in the neck causing respiratory insufficiency. *Laryngoscope* 1983;93:935–937
6. Brodsky JR, Kim DY, Jiang Z. Cervical lipoblastoma: case report, review of literature, and genetic analysis. *Head Neck* 2007;29:1055–1060
7. Choi HJ, Lee YM, Lee JH, et al. Pediatric lipoblastoma of the neck. *J Craniofac Surg* 2013;24:507–510
8. Rasmussen IS, Kirkegaard J, Kaasbøl M. Intermittent airway obstruction in a child caused by a cervical lipoblastoma. *Acta Anaesthesiol Scand* 1997;41:945–946
9. Dilley AV, Patel D, Hicks MJ, et al. Lipoblastoma: pathophysiology and surgical management. *J Pediatr Surg* 2001;36:229–231
10. O'Donnell KA, Caty MG, Allen JE, et al. Lipoblastoma: better termed infantile lipoma? *Pediatr Surg Int* 2000;16:458–461
11. Collins MH, Chatten J. Lipoblastoma/lipoblastomatosis: a clinicopathologic study of 25 tumors. *Am J Surg Pathol* 1997;21:1131–1137

Clinical Oral Findings in Patients With End-stage Renal Disease Receiving Hemodialysis

To the Editor: In a period of 1 year, we investigated the presence of oral lesions in 21 patients with end-stage renal disease (ESRD) receiving hemodialysis. The knowledge of oral lesions in these patients is important to establish strategies in an attempt to improve the quality of life. All patients were submitted to hemodialysis 3 times weekly, with mean of 4 hours each. Hypertensive glomerulonephritis was the most common cause of chronic renal failure and the mean period in dialysis was 13.7 months (range: 03–30 months). After clinical diagnosis, the lesions were grouped in (1) periodontal alterations, (2) dental or mucosal color changes, (3) uremic stomatitis, (4) infections, (5) premalignant and malignant lesions, (6) hemorrhage, and (7) others. Details of gender, age, etiology of renal disease, time of hemodialysis, and use of medications were also collected.

Of the 21 ESRD patients, 19 (90.5%) presented oral lesions. Calculus and gingival inflammation were the most common findings, followed by color changes in mucosal or dental tissues. Three cases of uremic stomatitis were observed, 2 in the ulcerative form and 1 erythemopultaceous. All infections were of fungal nature. The case of spontaneous hemorrhage was noted in the marginal gingiva of the right inferior canine and occurred immediately after hemodialysis section. No malignancies were diagnosed, but 2 cases of homogeneous leukoplakia in the palate and retromolar region, respectively, were observed (Table 1). The features of the patients are summarized in Table 2.

Chronic kidney disease (CKD) affects a range of tissues and systems and may cause neurological, cardiovascular, respiratory, immunological, hematopoietic, gastrointestinal, skin, and oral complications, either related to disease itself or by the renal replacement therapy (RRT). In a previous study,[1] we investigated the prevalence of oral lesions in renal transplant patients, and it was found that 81% of the sample showed some type of alteration. However, in the PubMed database, few studies about oral lesions in renal patients receiving hemodialysis were found. The high prevalence of oral

TABLE 1. Distribution of Oral Lesions in Patients Receiving Hemodialysis

Category	Absolute Frequency	Relative Frequency
Periodontal findings		
Calculus	11	26.8
Gingivitis	06	14.7
Color changes		
Petechiae	05	12.2
Pallor	04	9.8
Dental pigmentation	04	9.8
Uremic stomatitis		
Ulcerative	02	4.9
Erythemopultaceous	01	2.4
Infections		
Pseudomembranous candidiasis	03	7.3
Premalignant and malignant lesions		
Leukoplakia	02	4.9
Hemorrhage	01	2.4
Others		
Aphtous stomatitis	01	2.4
Traumatic ulcer	01	2.4
Total	41	100

TABLE 2. Clinical Features of Patients Receiving Hemodialysis

Patient	Age (yr)/Gender	Etiology of Renal Disease	Time in Hemodialysis (mo)	Oral Findings
1	62/M	Hypertensive nephrosclerosis	7	Pallor, calculus, pseudomembranous candidosis, ulcerative uremic stomatitis
2	55/F	Diabetic nephropathy	23	Calculus, pallor
3	35/F	Diabetic nephropathy	10	Pallor, dental pigmentation
4	56/M	Hypertensive nephrosclerosis	4	Calculus
5	37/F	Hypertensive nephrosclerosis	30	Calculus, gingivitis
6	63/M	Unclear	4	Calculus, gingivitis
7	26/F	Lupus nephritis	5	Ulcerative uremic stomatitis, calculus, gingivitis
8	45/M	Hypertensive nephrosclerosis	22	Petechiae
9	53/F	Hypertensive nephrosclerosis	3	—
10	51/F	Hypertensive nephrosclerosis	9	Pseudomembranous candidosis
11	63/F	Diabetic nephropathy	5	Leukoplakia, petechiae, dental pigmentation, gingivitis
12	75/F	Drug-induced nephrotoxicity	25	Leukoplakia, erythemopultaceous uremic stomatitis, petechiae
13	18/M	Chronic glomerulonephritis	27	Gingival ulcerations, dental pigmentation
14	28/F	Hypertensive nephrosclerosis	28	Calculus, gingivitis
15	49/F	Hypertensive nephrosclerosis	8	Calculus
16	37/F	Hypertensive nephrosclerosis	28	Petechiae
17	32/F	Diffuse glomerulosclerosis	10	Calculus
18	17/M	Chronic glomerulonephritis	14	Petechiae, spontaneous gingival bleeding
19	34/F	Unclear	8	Pallor, dental pigmentation, pseudomembranous candidosis, calculus
20	19/F	Chronic glomerulonephritis	12	Calculus, gingivitis
21	37/F	Diffuse glomerulosclerosis	6	—

clinical findings in the present study is similar to the one reported by De Rossi and Glick,[2] which showed that at least 90% of patients under hemodialysis present several oral alterations.

Infection is a frequent concern in patients with CKD and constitutes the second leading cause of death in patients undergoing RRT. It has been proposed that immunological and inflammatory disturbances, including defects in granulocytes, impaired B- and T-cell functioning, and impaired phagocytosis, occur at an early stage in the course of CKD, worsen with the progression of uremia, and are exacerbated by the dialysis procedure.[3] Therefore, the decreased immunological response in ESRD patients receiving hemodialysis can explain the high prevalence of gingival inflammation and the cases of fungal infection observed in the present study. Because fungal colonization may have potential consequences like sepsis,[2] these patients should be followed regularly leading to the early detection and treatment of fungal infections.

Uremic stomatitis is an uncommon complication of uremia resulting from advanced renal failure. Although the low incidence of this condition in patients with CKD can be attributed to the RRT,[4] some cases may be diagnosed in patients receiving hemodialysis, as observed in this study. It has been suggested that uremic stomatitis is a consequence of raised levels of ammonia compounds formed by the hydrolysis of bacterial ureases in saliva.[2] Uremic stomatitis is not considered a life-threatening condition, but patients may present xerostomia, uriniferous breath odor, unpleasant taste, and burning sensation, leading to a poor quality of life.

According to Bradford et al,[5] the risk of oral cancer in patients receiving hemodialysis is similar to that of otherwise healthy individuals in the general population. We did not observe any cases of oral cancer in the present study, but 2 patients were diagnosed with oral leukoplakia. Because 65% of oral homogeneous leukoplakia presents some degree of dysplasia,[6] and up to 18% of lesions progress to carcinoma,[7] patients with ESRD should be carefully monitored for signs of oral leukoplakia and treated promptly when these lesions are diagnosed.

In summary, it could be concluded that oral cavity can harbor a variety of manifestations in patients with ESRD receiving hemodialysis, being necessary to establish a preventive oral care program to improve the quality of life of these patients.

Luiz Carlos Ferreira da Silva, DDS, MSc
Manoel Pacheco de Andrade-Júnior, MD
Marta Rabello Piva, DDS, MSc
Paulo Ricardo Saquete Martins-Filho, DDS, MSc
Federal University of Sergipe
Sergipe, Brazil
martins-filho@ufs.br

Roseana de Almeida Freitas, DDS, MSc
Federal University of Rio Grande do Norte
Natal, Brazil

REFERENCES

1. Da Silva LCF, de Almeida Freitas R, de Andrade MP, et al. Oral lesions in renal transplant. *J Craniofac Surg* 2012;23:e214–e218
2. De Rossi SS, Glick M. Dental considerations for the patient with renal disease receiving hemodialysis. *J Am Dent Assoc* 1996;127:211–219
3. K/DOQI clinical practice guidelines for chronic kidney disease: evaluation, classification, and stratification. *Am J Kidney Dis* 2002;39:S1–S266
4. Broscious SK, Castagnola J. Chronic kidney disease: acute manifestations and role of critical care nurses. *Crit Care Nurse* 2006;26:17–2022–27; quiz 28
5. Bradford CR, Hoffman HT, Wolf GT, et al. Squamous carcinoma of the head and neck in organ transplant recipients: possible role of oncogenic viruses. *Laryngoscope* 1990;100:190–194
6. Schepman KP, van der Meij EH, Smeele LE, et al. Malignant transformation of oral leukoplakia: a follow-up study of a hospital-based population of 166 patients with oral leukoplakia from The Netherlands. *Oral Oncol* 1998;34:270–275
7. Reibel J. Prognosis of oral pre-malignant lesions: significance of clinical, histopathological, and molecular biological characteristics. *Crit Rev Oral Biol Med* 2003;14:47–62

A Patient with Pott Puffy Tumor With Pansinusitis and Orbital Involvement in an Immunocompromised Patient

To the Editor: Orbital cellulitis is a serious disease that can lead to permanent vision loss, intracranial extension, and death. It is occasionally encountered in young children, as a complication of frontal sinusitis. However, this orbital process is often an extension of primary sinus disease and can manifest with a subperiosteal abscess (SPA) and rarely with intracranial complications of sinusitis and Pott puffy tumor (PPT).[1–11] Pott puffy tumor is an extracranial, frontal SPA, resulting from frontal osteomyelitis.[12] The tumor has been reported mostly in the pediatric population and adolescents and only rarely in adults.[13–15]

A literature review revealed that no report has been previously issued on PPT with orbital involvement in an immunocompromised adult. We present a rare case of PPT with orbital involvement in an immunocompromised adult and include a brief literature review.

A 55-year-old man presented at our clinic with right periorbital swelling, which had persisted for 3 days. He had a history of mitral valve replacement 2 years previously and had been on immunosuppressant treatment since. While under immunosuppressant treatment, pansinusitis developed and spread to the maxilla, palate, and nasal septum. Subtotal maxillectomy was performed, and a histopathologic biopsy confirmed osteomyelitis.

At this presentation, he had frontal swellings on both sides of the forehead and right upper lid swelling and redness (Fig. 1). Best-corrected visual acuity was 20/32 in the right eye and 20/25 in the fellow eye. His intraocular pressures were normal, pupils were isochoric and reactive to light, and he had full-range extraocular movement without proptosis.

An enhanced computed tomography (CT) scan revealed severe bone destruction combined with adjacent soft tissue swelling on both frontal, ethmoid, nasal, maxillary, and palatine bones. An SPA was also found on the forehead (Fig. 2). Magnetic resonance imaging revealed a preseptal abscess, enhancement of both frontal convexities, and dural thickening, suggesting osteomyelitis (Fig. 3). A bone scan showed increased uptake in the maxillae and frontal bone.

At this time, his body temperature was 38.4°C, and laboratory findings showed pancytopenia and elevated erythrocyte sedimentation rate and C-reactive protein.

Methicillin-resistant *Staphylococcus aureus*, *S. aureus*, *Pseudomonas aeruginosa*, *Enterococcus faecium* (vancomycin-resistant enterococci), and *Aspergillus* were isolated from cultures of open pus and bone.

Histopathologic examination revealed numerous inflammatory cells in the abscess by hematoxylin-eosin staining and degenerated fungal hyphae (*Aspergillus*) in destroyed bone tissue by Grocott-Gomori methenamine silver staining (Fig. 4).

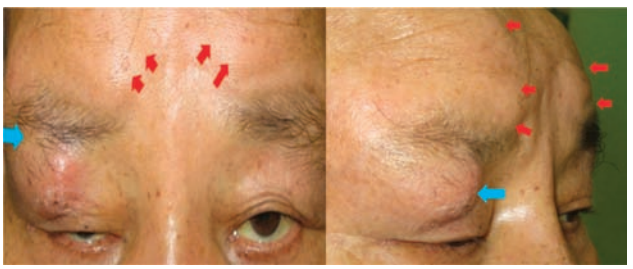

FIGURE 1. Photograph of the patient at initial presentation. The red arrows indicate frontal swellings on both sides of the forehead, and the blue arrow shows right upper lid swelling and redness.

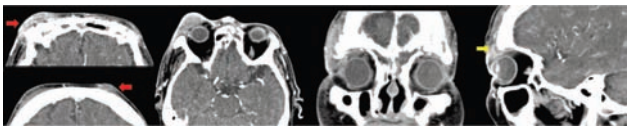

FIGURE 2. Enhanced CT findings. The red arrow indicates PPT on the forehead, and the yellow arrow indicates an SPA.

Based on clinical and histological findings, a diagnosis of PPT associated with osteomyelitis in frontal bone, pansinusitis, and orbital cellulitis with orbital roof involvement was reached.

After a thorough preoperative evaluation, surgical debridement and pus drainage were performed on pus-filled sequestra with combined bilateral frontal bone craniectomy. Following surgery, broad-spectrum antibiotics and antifungal treatment using third- and fourth-generation cephalosporins, quinolones, and voriconazole were administered for 4 weeks.

Within 1 month of surgery, the periorbital swelling had dramatically subsided, and only redness of the right upper eyelid remained (Fig. 5).

Pott puffy tumor is not a real tumor but a swelling of the forehead due to an SPA associated with osteomyelitis in frontal bone. It is classically caused by frontal sinusitis and can be complicated by orbital and intracranial involvement. The frontal sinus is most commonly associated with intracranial infection, followed by the ethmoid, sphenoid, and maxillary sinuses.[16,17] Furthermore, the intracranial complications of sinusitis may include meningitis, epidural abscess, subdural empyema, brain abscess, and dural and cavernous sinus thrombosis.[12]

The morbidity or mortality rate of patients with complications arising from sinusitis, including osteomyelitis, has been reported to range from 5% to 40%.[18,19] The incidence of orbital complications following sinusitis is highly variable but is usually more than 20%[20] and may reach 80% in immunocompromised patients.[19] However, the incidence of frontal osteomyelitis, as an intracranial complication of pansinusitis, is low at about 1.5% of all osteomyelitis cases.[21]

In immunocompromised patients, a small number of reports have described osteomyelitis elsewhere, such as in mandible[22] or spine,[23] but no report of PPT with orbital involvement due to frontal osteomyelitis has been issued.

The symptoms of frontal osteomyelitis vary from nonspecific, such as headaches, vomiting, and fever, to a localized lump and tender swelling over bone. In our case, the patient had frontal swelling on both sides of the forehead with focalized right upper lid swelling and redness.

Courses and response to therapy differ significantly between immunocompromised and immunocompetent hosts, and in the former, aggressive spread and high mortality rate require prolonged treatment with combinations of antibiotics. Immunocompetent hosts present a lower mortality rate and respond to shorter treatment, usually based on a single antibiotic.[24–27]

The frontal bone and maxilla are the 2 orbital bones most commonly involved in osteomyelitis.[28] In our case, CT revealed severe bone destructions on frontal, ethmoid, nasal, maxillary, and

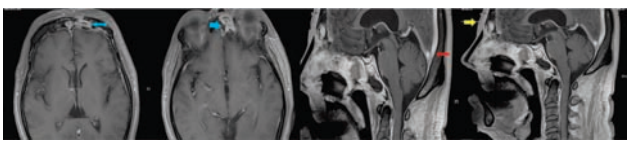

FIGURE 3. Magnetic resonance imaging findings. The blue arrow indicates a preseptal abscess due to osteomyelitis; the red arrow shows dural thickening, and the yellow arrow shows enhancement of both frontal convexities.

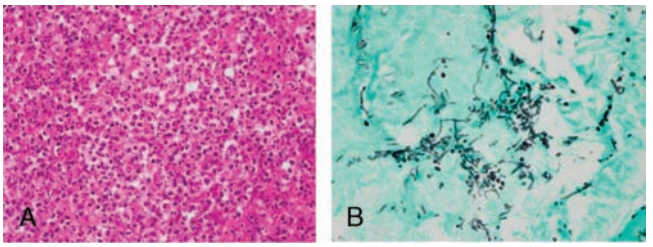

FIGURE 4. Histopathology. A, Photomicrograph showing the presence of numerous inflammatory cells in the abscess (hematoxylin-eosin stain, original magnification ×400). B, Degenerated fungal hyphae of aspergillosis in destroyed bone tissue (Grocott-Gomori methenamine silver, original magnification ×400).

palate bones, and bone scan showed increased uptake in both maxilla and frontal bone.

Chronic osteomyelitis is defined as a special inflammatory process involving cortical and cancellous bone that evolves over months or even years, and which is characterized by the persistence of microorganisms, low-grade inflammation, sequestrum, and fistulous tracts.[29,30] This type of bone infection is not curable with antibiotics and can be cured only if sequestrum is removed. In our case, surgical debridement and pus drainage of sequestra with pus and combined neurosurgery for necrotic bone and tissue removal of bilateral frontal bone were achieved by surgery and postoperative antibiotic therapy, and the patient recovered quickly with normal vision and few complications.

The most common bacteria isolated from this type of infection are *S. aureus*, streptococcus, and *Haemophilus influenzae*.[28,31,32] In our case, cultures on open pus and bone identified *S. aureus*, *P. vulgaris* (methicillin-resistant *S. aureus*), *P. aeruginosa*, *E. faecium* (vancomycin-resistant enterococci), common bacteria, and *Aspergillus*. The patient was under immunosuppressant treatment, which explains the uncommon culture findings.

The management of osteomyelitis includes symptomatic therapy, immobilization for some patients, adequate drainage of purulent material, and antibiotic therapy consisting of the parenteral administration of antibiotics for at least 4 to 8 weeks. In cases thus far reported, combination therapy for 2 to 6 months appears to be most successful, and surgical intervention, whenever possible, increases probability of definite cure.[33,34] In our case, aggressive treatment with 4 weeks of broad-spectrum antibiotics and antifungal treatment with third- and fourth-generation cephalosporins, quinolones, and voriconazole were initiated after surgery.

Pott puffy tumor is a rare disease and usually develops secondary to sinusitis in immunosuppressant patients. A high index of suspicion is required to achieve an early diagnosis of infection as PPT progresses rapidly. In addition, because PPT has high recurrence rate, early aggressive treatment with broad-spectrum antimicrobial agents and surgical debridement is needed.

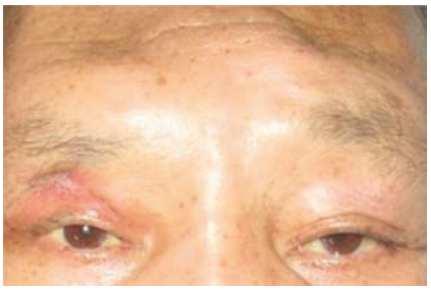

FIGURE 5. Photograph taken 1 month after surgery. The periorbital swelling dramatically subsided; only redness of the upper right eyelid remained.

Sang Hun Hyun, MD
Jee Woong Jung, MD
Mijung Chi, MD, PhD
Department of Ophthalmology
Gachon University Gil Hospital
Incheon, South Korea
cmj@gilhospital.com

REFERENCES

1. Kombogiorgas D, Solanki GA. The Pott puffy tumor revisited: neurosurgical implications of this unforgotten entity. Case report and review of the literature. *J Neurosurg* 2006;105:143–149
2. Ibarra S, Aguirrebengoa K, Pomposo I, et al. Osteomyelitis of the frontal bone (Pott's puffy tumor). A report of 5 patients. Enferm Infecc. *Microbiol Clín* 1999;17:489–492
3. Karaman E, Hacizade Y, Isildak H, et al. Pott's puffy tumor. *J Craniofac Surg* 2008;19:1694–1697
4. Durur-Subasi I, Kantarci M, Karakaya A, et al. Pott's puffy tumor: multi detector computed tomography findings. *J Craniofac Surg* 2008;19:1697–1699
5. Masterson L, Leong P. Pott's puffy tumor: a forgotten complication of frontal sinus disease. *Oral Maxillofac Surg* 2009;13:115–117
6. Lamoreau KP, Fanciullo LM. Pott's puffy tumor mimicking preseptal cellulitis. *Clin Exp Optom* 2008;91:400–402
7. Kaabia N, Abdelkafi M, Bellara I, et al. Pott's puffy tumor. a case report. *Méd Mal Infect* 2007;37:350–353
8. Bambakidis NC, Cohen AR. Intracranial complications of frontal sinusitis in children: Pott's puffy tumor revisited. *Pediatr Neurosurg* 2007;35:82–89
9. Younis RT, Lazar RH, Anand VK. Intracranial complications of sinusitis: a 15-year review of 39 cases. *Ear Nose Throat J* 2002;81:636–638
10. Moloney JR, Badham NJ, McRae A. The acute orbit. Preseptal cellulitis, subperiosteal abscess, and orbital cellulitis due to sinusitis. *J Laryngol Otol Suppl* 1987;12:1–18
11. Fearon B, Edmonds B, Bird R. Orbital-facial complications of sinusitis in children. *Laryngoscope* 1979;6:947–953
12. McDermott C, O'Sullivan R, McMahon G. An unusual cause of headache: Pott's puffy tumour. *Eur J Emerg Med* 2007;14:170–173
13. Shehu BB, Mahmud MR. Pott's puffy tumour: a case report. *Ann Afr Med* 2008;7:138–140
14. Upadhyay S. Recurrent Pott's puffy tumour: a rare clinical entity. *Neurol India* 2010;58:815–817
15. Bagdatoglu C, Guleryuz A, Ersoz G, et al. A rare clinical entity: Pott's puffy tumour. *Pediatr Neurosurg* 2001;34:156–158
16. Blitzer A, Carmel P. Intracranial complications of disease of the paranasal sinuses. In: Blitzer A, Lawson W, Friedman WH, eds. *Surgery of the Paranasal Sinuses.* Philadelphia, PA: Saunders; 1985: 328–337
17. Fairbank D, Vanderveen T, Bordley J. Intracranial complications of sinusitis. In: English GM, ed. Otolaryngology. Vol. 2. New York: Harper & Row, 1983:1–28
18. Maniglia AJ, Goodwin WJ, Arnold JE, et al. Intracranial abscesses secondary to nasal, sinus, and orbital infections in adults and children. *Arch Otolaryngol* 1989;115:1424–1429
19. Parikl SL, Venlkatraman G, DelGaudio JM. Invasive fungal sinusitis: a 15-year review from a single institution. *Am J Rhinol* 2004;18:75–81
20. Chandler JR, Langenbrunner DJ, Stevens ER. The pathogenesis of orbital complications in acute sinusitis. *Laryngoscope* 1970;80: 1414–1428
21. Osei-Yeboah C, Neequaye J, Bulley H, et al. Osteomyelitis of the frontal bone. *Ghana Med J* 2007;41:88–90
22. George R, Panayotis D, George G, et al. Acute osteomyelitis of the mandible caused by Rhodococcusequi in an immunocompromised patient: a case report and literature review. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2012;114:1–5
23. Robert LW, Melanie BF, Carolyn CM, et al. Fungal spinal osteomyelitis in the immunocompromised patient: MR findings in three cases. *Am J Neuroradiol* 1999;20:381–385

24. Verville TD, Huycke MM, Greenfield RA, et al. Rhodococcusequi infections of humans: 12 cases and a review of the literature. *Medicine (Baltimore)* 1994;73:119–132
25. Votava M, Skalka B, Hrstkova H, et al. Review of 105 cases of isolation of Rhodococcusequi in humans. *CasLekCesk* 1997;136:51–53
26. Weinstock DM, Brown AE. Rhodococcusequi: an emerging pathogen. *Clin Infect Dis* 2002;34:1379–1385
27. Kedlaya J, Ing MB, Wong SS. Rhodococcusequi infections in immunocompetent hosts: case report and review. *Clin Infect Dis* 2001;32:39–46
28. Kapoor S, Kapoor MS, Sood GC. Osteomyelitis of orbital bones. *J Pediatr Ophthalmol* 1977;14:171–175
29. Jauregui LE, Senour CL. Chronic osteomyelitis. In: Jauregui LE, ed. *Diagnosis and management of bone infections.* New York: Dekker; 1995:37–108
30. Caputo GM, Cavanagh PR, Ulbrecht JS, et al. Assessment and management of foot disease in patients with diabetes. *N Engl J Med* 1994;331:854–860
31. El-Toukhy E, Szal M, Levine MR, et al. Osteomyelitis of the orbit. *Ophthal Plast Reconstr Surg* 1997;13:68–71
32. Krishna G. Osteomyelitis of the supraorbital margin. *Ind J Ophthalmol* 1984;30:111–112
33. Van Etta LL, Filice GA, Ferguson RM, et al. Corynebacteriumequi: a review of 12 cases of human infection. *Rev Infect Dis* 1982;5:1012–1018
34. McGowan KL, Mangano MF. Infections with Rhodococcusequi in children. *Diagn Microbiol Infect Dis* 1991;14:347–352

A Modification of the Posterior Auricular Flap Used for Scapha and Helix Middle One-Third Defects of the Auricle

Two main reasons of the auricular defects are trauma and skin cancer.[1] Defects that are not reconstructed appropriately cause obvious deviations especially at the helical region, and these deviations cause psychological morbidity in both adults and children.[2]

An 84-year-old male patient represented with a 1×0.5-cm dysplastic lesion at his right scapha. The lesion and the underlying cartilage were excised with a clean margin of 1 cm. The posterior skin was preserved (Fig. 1). The posterior auricular flap was designed with a dimension of 3×5 cm. In fact, the width was equal to the defect width (3 cm), and the length was equal to the length between posterior auricular sulcus and flaps distal edge (5 cm). Posterior skin of the defect was incised from the cartilage margin, and a tunnel was created. The flap was passed from the tunnel, and the defect margins were marked on the flap. The skin that was lying anterior to the marked edge and the projection of this area at the posterior auricle was de-epithelialized (Fig. 2). The flap was passed from the tunnel (Fig. 3), and the inset was made. The donor site was reconstructed with the full-thickness skin graft, which was obtained from the de-epithelialization process.

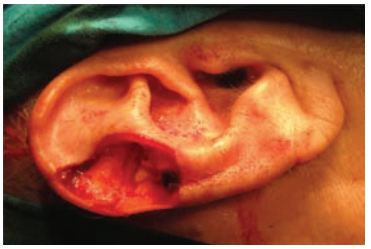

FIGURE 1. The defect after excision of the lesion.

FIGURE 2. The posterior auricular flap after de-epithelialization.

FIGURE 3. The flap that was passed from the tunnel.

FIGURE 4. Hair growth on the anterior surface of the ear.

The best known reconstruction options for the defects of the middle one-third helical and antihelical regions are wedge excision and primary repair, Banner flap, Anita Buch flap, and primary repair after stellate excisions. Disadvantages of these techniques are the need for further excisions from the nontumor sites and loss of height of the auricle.

Posterior auricular flap is an ideal option for wide defects of the middle one-third of the helix and antihelix. Main disadvantage of the standard posterior auricular flap is the necessity for a 2-stage procedure.[3] We describe the 1-stage usage of the posterior auricular flap. The disadvantages are the postauricular scar and the hair growth on the anterior surface of the ear (Fig. 4). We believe that this modification of the posterior auricular flap can be used successfully in consideration of less donor-site morbidity, lack of auricular height loss, and ability to restore support alternatively for cartilage because of the flap thickness.

Orhan Çizmeci, MD
Fethi Sarper Mete, MD
Erol Kozanoğlu, MD
Samet Vasfi Kuvat, MD
Department of Plastic, Reconstructive and Aesthetic Surgery
Fatih, Capa
Istanbul, Turkey
ocizmeci@istanbul.edu.tr

REFERENCES

1. Elhasy NI. Acquired ear defects. *Clin Plast Surg* 2002;29:175–186
2. Horlock N, Vogelin E, Bradbury ET, et al. Psychosocial outcome of patients after ear reconstruction: a retrospective study of 62 patients. *Ann Plast Surg* 2005;54:517–524
3. Cheney ML, Hadlock TA, Quatela VC. Reconstruction of the auricle. In: Baker SR, ed. *Local Flaps in Facial Reconstruction.* 2nd ed. Philadelphia, PA: Elsevier Inc; 2007:581–624

Complications After Mandibular Third Molar Surgery: A Case of Bulky Peripheral Osteoma

To the Editor: Many postoperative complications have been described after third-molar surgery. Osteoid osteoma after a third-molar extraction is very rare. The purpose of this letter is to report a case of bulky peripheral osteoma after third-molar surgery, rare for its histology, dimensions, growth, and teeth association.

A 35-year-old healthy man referred himself to the Department of Maxillofacial Surgery, University of Siena, for right mandibular swelling and bleeding: patient referred the beginning of his simptoms and the growth of the mass few weeks after right third-molar surgery. Clinical examination revealed a giant vestibular erythematous mass, bleeding with minimal pressure (Fig. 1). Patient history revealed no smoking habits and no previous oral or systemic neoformations. Because of its initial aggresive presentation, our first suspicion was for a malignant neoplastic mass. Computed tomography scan showed a 24 × 10-mm volume mass in contact with third-molar socket, infiltrating soft tissues, and first and second molars (Fig. 2). The Gardner syndrome was excluded. For this reason, we decided for a preliminary incisional biopsy, which excluded a malignant mass.

Afterward, we planned for a mass removal with bone debriding and first- and second-molar removal (Fig. 3). Histological examination revealed a complex mass compoud of pseudoepitelial hyperplasia, fibrosis, and necrosis areas, with a central dystrophic osteoid osteoma. Patient was discharged the day after surgery, and postoperative visits showed mucosal healing. Postoperative control 1 year after surgery showed complete recovery (Fig. 4).

Many authors agree that the differentiation between osteoma and osteoblastoma should be based on the size, site, and radiological appearance.[1] The differential diagnosis of this lesion includes cementoblastoma, sclerosing osteomyelitis, and ossifying fibroma too.

The first case of an osteoma after a tooth extraction was reported by Rushton[2] in 1951. Peripheral osteoma should be distinguished from jaw exostoses: the main difference is the development of exostoses as reaction, whereas peripheral osteoma is a true neoplasm.

FIGURE 1. Giant vestibular mass that occupies the entire vestibular fornix and protrudes forward: patient referred pain and bleeding during even tooth brushing and mastication.

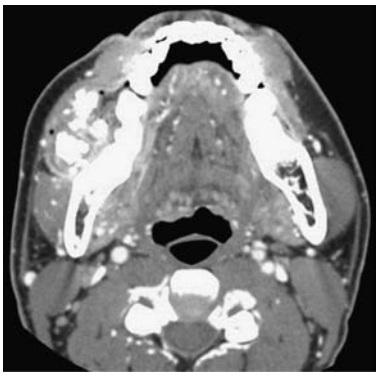

FIGURE 2. Computed tomography scan axial view shows a mass with numerous radiolucency masses in contact with third-molar socket, infiltrating soft tissues, and first and second molars (24 × 10-mm volume).

The peripheral osteoma is a mature bone tumor with a compact part, usually with a sessile base, normal-appearing bone with minimal marrow spaces, and the cancellous part that resembles the bone of origin.[3]

Dimensions play an important role: small osteoma may present no symptoms for many years, and when intervention is needed, usually a minimal surgery is the correct approach. Giant osteoma may disfigure the face and/or become a life thread.[4]

In cases of association with teeth, surgery becomes more invasive, in order to prevent possible recurrency. Recently, Mohammed et al[5] reported an interesting case of osteoid osteoma in association with teeth: in the reported case, Mohammed et al[5] referred nocturnal pain and tenderness as main symptoms that convinced the patient to refer himself to their department. Differently, the patient we reported presented no nocturnal pain, but the main symptom was pain after chewing and bleeding, as the bulky mass interposed between the teeth during function.

Giant osteomas may present a great variety of symptoms: their dimensions and the location appear to be the more important

FIGURE 3. Aspect of the mass after the removal.

FIGURE 4. Postoperative view of the site 1 year after surgery.

variables. Time also plays an important role: our first suspicion was for a malignant mass as the neoformation became bulky in dimensions in few weeks.

The recurrency is another matter of interest: oral osteoma has not been reported to recur after proper surgery, differently with oral osteoblastoma that may recur after complete resection, also many years later.[6]

The interesting presentation of this case should be considered in literature as only few authors reported peripheral osteoma associated with teeth.[5] With regard to the case we reported, the volume (24×10 mm) and the fast growth after the third-molar extraction are noteworthy, too.

Surgical removal of the entire mass along with the teeth of interest should be performed to ensure complete removal and to avoid recurrence. Prolonged follow-up also plays an important role. Removable dental prosthesis should be preferred to dental implants for the first years after surgery.

Aboh Ikenna Valentine, MD, PhD
Chisci Glauco, DDS
Gennaro Paolo, MD, PhD
Department of Maxillofacial Surgery
University of Siena
Siena, Italy
goez@libero.it

Gabriele Guido, MD
Cascino Flavia, MD
Iannetti Giorgio, MD, PhD
Department of Maxillofacial Surgery
"Sapienza" University of Rome
Rome, Italy

REFERENCES

1. Farman AG, Nortje CJ, Grotepass F. Periosteal benign osteoblastoma of the mandible. Report of a case and review of the literature pertaining to benign osteoblastic neoplasm of the jaws. *Br J Oral Surg* 1976;14: 12–22
2. Rushton MA. An osteoid-osteoma of the mandibular alveolus. *Oral Surg Oral Med Oral Pathol* 1951;4:86–88
3. Sayan NB, Uçok C, Karasu HA, et al. Peripheral osteoma of the oral and maxillofacial region: a study of 35 new cases. *J Oral Maxillofac Surg* 2006;60:1299–1301
4. Badauy C, Carrard V, Rados P, et al. Radiopaque mass of the posterior mandible with lingual expansion. *J Oral Maxillofac Surg* 2005;65:2498–2502
5. Mohammed I, Jannan NA, Elrmali A. Osteoid osteoma associated with the teeth: unusual presentation. *Int J Oral Maxillofac Surg* 2013;42:298–302
6. Rahsepar B, Nikgoo A, Fatemidabar SA. Osteoid osteoma of subcondylar region: case report and review of the literature. *J Oral Maxillofac Surg* 2009;67:888–893

Histologic Study of the Articular Eminence in Wilkes IV Temporomandibular Joint Disorder Patients

Temporomandibular joint dysfunction (TMD) is a relatively usual problem, with a 12% to 87% reported incidence in the population. Disorders affecting this complex structure may manifest as musculoskeletal alterations, and close to one third of the adult population develop symptoms connected with articular pathology, which are concurrently related to stress, age, sex, and systemic factors.[1,2]

Numerous investigations and articles have discussed the anatomy and physiology of the temporomandibular joint (TMJ), but only a few of them have analyzed the histological changes of the diseased joint.

The aim of the present research was to determine the microscopic appearance of the articular eminence in patients diagnosed with Wilkes IV TMD who underwent open TMJ surgery with eminectomy.[3]

The sample consisted of 10 TMJ surgery patients in whom 15 articular eminences were removed, processed, stained with hematoxilin-eosin, and analyzed by an oral and maxillofacial pathologist at the Universidad El Bosque School of Dentistry. Patients were operated on from July 2011 to April 2012 at either Hospital Universitario Clínica San Rafael or Hospital Simón Bolívar in Bogotá, Colombia. Eighty percent of patients were women, and 20% were men, with an age average of 29.8 years. Descriptive statistics of the patient sample are shown in Table 1.

The studied eminences showed hyperplastic changes and active chondrocytes showing hypermetabolism on the growing center with hyaline cartilage converting into fibrocartilage. A proliferative zone consisting of calcified bone and cartilaginous matrix, basophilic lines of cementation that indicated bone apposition between mature and hyperplastic bone, was demonstrated (Figs. 1 and 2).

Histopathologic studies of the different anatomical components of the diseased TMJ are not abundant. In 1997, Bernasconi et al[4] described the clinical and histological findings in the synovial tissues on 10 articular discs of patients with TMD, and they found changes such as adhesions between the surface of the disk and the glenoid eminence or the condylar head in all the cases. Histological examination demonstrated hyperplasia of the synovial tissue with formation of prominent protrusions, increased levels of type B cells, and cells with myofibroblastic characteristics. Mineral precipitates and deposits of calcified tissue were found in 3 cases. They

TABLE 1. Descriptive Statistics of Patient Sample (n = 10)

Variables	
Demographics	
Mean age, y	29.8
Male/female	2/8
Health status	
ASA I	8
ASA II	2
Clinical findings	
Chronic TMJ pain	10
Mild	6
Severe	4
Headaches	8
Cervical pain	5
Restricted motion	6
Relapsing condylar luxation	3
Condition	
Right	2
Left	3
Bilateral	5
MRI findings	
Anterior disc displacement without reduction	7
Disc thickening	7
Abnormal bone contours	8

ASA indicates American Society of Anesthesiologists; MRI, magnetic resonance imaging.

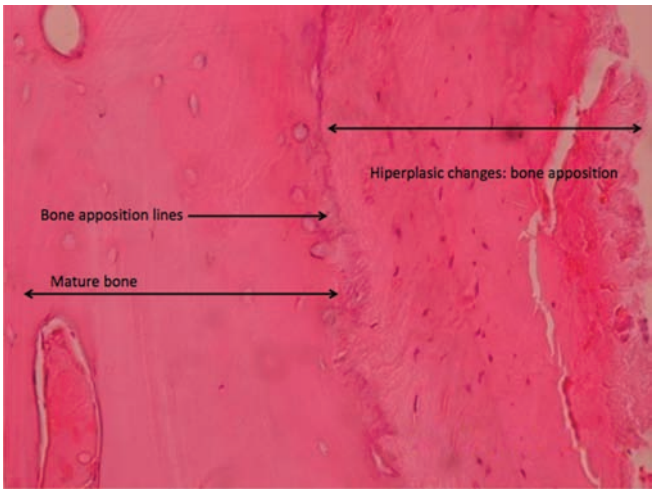

FIGURE 1. Photomicrograph of a left articular eminence of a 32-year-old female patient. It shows bone apposition lines, stained with hematoxylin-eosin, original magnification ×480.

suggested that the morphologic lesions and the functional failure could be caused by excessive tissue stress and functional demands due to arthopathy.

Normal and hyperplastic condyles were studied by Eslami et al[5] in 2003. In their histopathologic comparison, they found a significant difference between the 2 groups in the thickness of the hyperplastic cartilage layer of condylar soft tissue. What they concluded is that the diameter of the abnormal cartilage layer appears significantly increased in condylar hyperplasia.

Similar to what other investigators found in the rest of the anatomical components of the TMJ, we also demonstrated hyperplastic changes in the articular eminence of the diseased joint. These hyperplastic alterations also showed conversion of hyaline cartilage into fibrocartilage, a condition commonly associated to trauma, increased functional demands, chronic inflammatory response, neoplasm, or mucinous degeneration. Other adaptations such as hypermetabolic growth centers, calcified zones of bone and cartilaginous matrix, and basophilic lines of cementation exemplify the proliferative process that takes place when the function of the joint is altered.

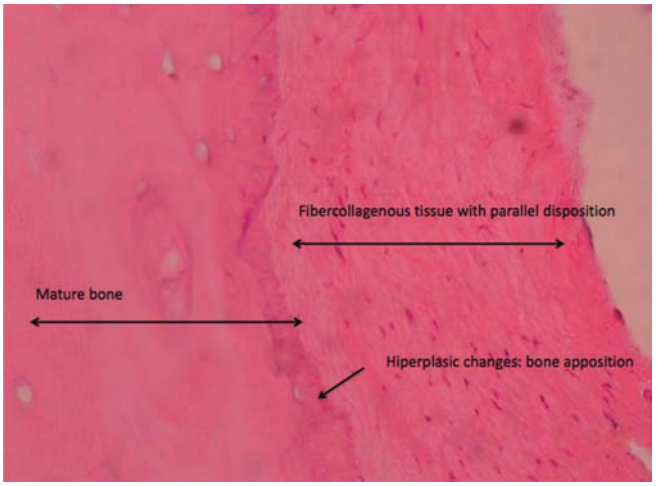

FIGURE 2. Photomicrograph of a right articular eminence of a 52-year-old female patient. The section shows hyperplastic changes from mature bone to a hyperplastic zone, stained with hematoxylin-eosin, original magnification ×480.

In conclusion, histological changes observed on the studied sample suggest that a significant relationship may exist between degenerative alterations of the TMJ and remodeling process of the articular eminence.

Carlos Alberto Ruiz Valero, DDS
Oral and Maxillofacial Surgery Department
Hospital Universitario Clínica San Rafael
and Oral and Maxillofacial Surgery Residency Program
Universidad El Bosque
Bogotá, Colombia
caruizv2000@yahoo.com

Diego Mauricio Barreto Suárez, DDS
Oral and Maxillofacial Surgery Department
Hospital Simón Bolívar
and Oral and Maxillofacial Surgery Department
Universidad El Bosque; and Private Practice
Bogotá, Colombia

María Rosa Buenahora, DDS
Oral and Maxillofacial Pathology Department
Universidad El Bosque
Bogotá, Colombia

Carolina Gamboa Hernández, DDS
Diana Carolina Grajales, DDS
Andrés Gómez-Delgado, DDS
Jaime Castro-Núñez, DMD
Oral and Maxillofacial Surgery Department
Universidad El Bosque
Bogotá, Colombia

REFERENCES

1. Flynn B, Brown DT, Lapp TH, et al. A comparative study of temporomandibular symptoms following mandibular advancement by bilateral sagittal split osteotomies: rigid versus nonrigid fixation. *Oral Surg Oral Med Oral Pathol* 1990;70:372
2. Kitsoulis P, Marini A, Iliou K, et al. Signs and symptoms of temporomandibular joint disorders related to the degree of mouth opening and hearing loss. *BMC Ear Nose Throat Disord* 2011;11:5
3. Wilkes CH. Internal derangements of the temporomandibular joint. Pathological variations. *Arch Otolaryngol Head Neck Surg* 1989;115:469
4. Bernasconi G, Marchetti C, Reguzzoni M, et al. Synovia hyperplasia and calcification in the human TMJ disk. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1997;84:245
5. Eslami B, Behnia H, Javadi H, et al. Histopathologic comparison of normal and hyperplastic condyles. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2003;96:711

Submandibular Ossifying Lipoma

To the Editor: The ossifying lipoma, or osteolipoma, is a tumor characterized by mature, benign bone formation within the neoplastic fatty tissue. With regards to the pathogenesis, two main theories exist: the metaplasia may be related to endochondral ossification of pluripotent mesenchymal cells in the adipose tissue, or may arise by secondary metaplasia of fibroblasts after repetitive trauma, metabolic changes, or ischemia.[1]

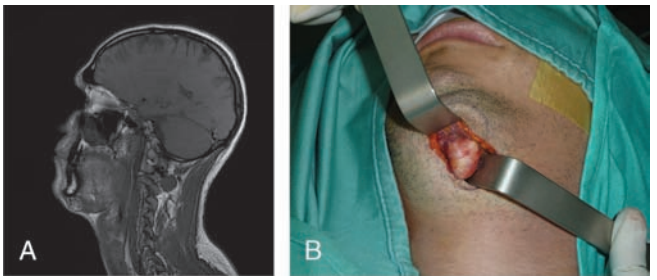

FIGURE 1. MRI showing a compact submandibular mass (A); intraoperative figure showing the excision of the ossifying lipoma with minimally invasive surgery via a minimal skin incision (B).

Symptoms of oral and maxillofacial ossifying lipoma depend on site of their presentation; parapharyngeal or nasopharyngeal mass may cause dysphagia, hearing loss, jaw pain, or paresthesia. We present a case of a 33-year-old man referred to the Department of Maxillofacial Surgery, University of Siena, with a history of painless swelling in the left submandibular region for many years. MRI with contrast revealed the presence of a mass in the left submandibular region. We decided for a minimally invasive surgery with mass removal (Fig. 1). The patient was discharged the day after surgery, and postoperative visits showed reduction of submandibular swelling. Pathological examination revealed for an ossifying lipoma.

Oral and maxillofacial lipoma is very rare. According to the World Health Organization (WHO) 2002 classification of soft tissue tumors, conventional lipoma is composed of lobules of mature adipocytes and can arise within subcutaneous tissue (superficial lipoma) or within deep soft tissues (deep lipoma) or even on the surfaces of bone (parosteal lipoma).[2]

Kavusi et al reported only 3 cases of submandibular ossifying lipoma over 20 cases of oral and maxillofacial osteolipomas.[3]

We treated this tumor with a minimally invasive approach through a small incision in the submental region and accurate excision of the mass, avoiding damage to all surrounding structures. The mass was not attached to the mandibular bone. The presence of dense collagen fibers and osseous trabeculae scattered among adipocytes lend some support to the hypothesis of fibroblastic metaplasia or origination of tumor from a multipotential stem cell. The presence of this tumor in an infrequent traumatic zone excludes the other main cause of lipoma.

Considering the rarity of the variant ossifying lipoma in the submandibular region and the dimensions of the case we reported, we suggest to consider ossifying lipoma in the differential diagnosis of submandibular swelling; minimally invasive surgery is advocated for the management of these masses in the submandibular region, with preservation of surrounding tissues and a minimal scar.

Ikenna V. Aboh, MD, PhD
Glauco Chisci, DDS
Chiara Salini, MD
Paolo Gennaro, MD, PhD
Flavia Cascino, MD
Guido Gabriele, MD
Giorgio Iannetti, MD
Department of Maxillofacial Surgery
University of Siena
Siena, Italy

REFERENCES

1. Fritchie KJ, Renner JB, Rao KW, et al. Osteolipoma: radiological, pathological, and cytogenetic analysis of three cases. *Skeletal Radiol* 2012;41:237–244
2. Fletcher C, Krishnan Unni K, Mertens F, (eds). Pathology and genetics of tumours of soft tissue and bone. In: Kleihues P, Sobin LH, series, eds. *World Health Organization Classification of Tumours*. Lyon: IARC Press, 2002
3. Kavusi S, Farahmand V, Davidson TM, et al. Osteolipoma presenting as a submandibular mass: a rare presentation. *Head Neck Pathol* 2013;7:93–96

Primary Cutis Verticis Gyrata

To the Editor: Cutis verticis gyrata is a rare disease characterized by the formation of furrows and folds on the scalp. As a result of the furrows and folds formed, over time, the scalp gains an appearance resembling that of brain gyri.[1] The disease was first defined by Robert in 1843. It was later classified by Polan and Butterworth[2,3] in 1953, based on disease assessment, as primary (essential or nonessential) or secondary. The primary essential form manifests as an isolated deformation without any associated disease.

A 39-year-old female patient presented to our clinic with folds and thickening on the scalp, and an examination revealed generalized skin folds of various sizes over the entire scalp. The skin folds in the occipital region were observed to be much more intense and evident compared with the folds in other sites (Fig. 1). It was revealed that the complaints of the patient started within a 5×5-cm area in the occipital region when she was 25 years old. With the exception of heat-cold intolerance at the sites of the folds on the scalp, she did not have any physical complaints. The patient was most disturbed about the appearance of the occipital region.

The patient's family had no history of a similar disease. The systemic examination of the patient revealed no metabolic, neurological, ophthalmologic, or developmental pathology. Laboratory results did not reveal any notable findings. In light of the available findings, the patient was considered to have cutis verticis gyrate, and the decision was made to treat her surgically. The most evident skin folds exhibiting transverse and vertical elongation in the occipital region were marked and excised (Fig. 1). To restore the scalp defects that occurred, the caudal and cranial scalp was elevated along the subgaleal plane. It was determined that the galea aponeurotica prevented the skin flaps from drawing forward, and the skin folds were very strictly adhered to the galea aponeurotica; thus, the galea aponeurotica was excised from the bottom of the flaps. After the galea aponeurotica was excised, improvement was observed in the skin folds, with significant laxity in the skin flaps. Next, excess tissue was excised from the skin flaps, and the incision

FIGURE 1. Preoperative view of the patient. Marked folds were excised.

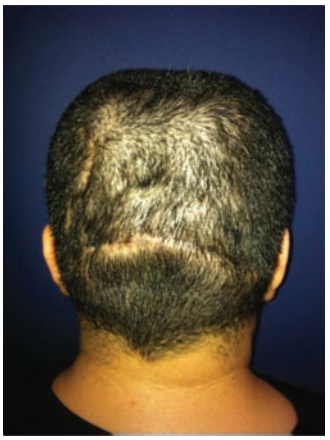

FIGURE 2. Postoperative view after 4 weeks.

lines were sutured to each other. No complications were observed during the operation.

A significant improvement in the physical complaints of the patient, as well as a positive cosmetic outcome to a higher degree, was achieved after surgery (Fig. 2).

Primary essential cutis verticis gyrata is generally characterized by symmetrical folds that can cover the whole scalp but are usually dense in the vertex and occipital regions. It occurs after puberty and is 5 times more common in men than in women. There are no additional diseases associated with the primary essential form; however, the nonessential form is associated with many neurological, psychiatric, and ophthalmologic disorders.[4] The histopathologic findings may vary from a normal skin texture to a thickening of the connective tissue with hypertrophy of adnexal structures.[1]

Secondary cutis verticis gyrata is more common than the primary form, and it occurs as a consequence of a number of local and systemic diseases that are not related to each other.[3] The secondary form may be caused by local inflammatory diseases, such as psoriasis, impetigo, folliculitis, erysipelas, and pemphigus, or space-occupying formations, such as congenital cerebriform intradermal nevus, neurofibromas, fibromas, and hamartomas. Furthermore, the secondary form is also associated with many systemic diseases, mainly including myxedema, acromegaly, amyloidosis, syphilis, Ehlers-Danlos syndrome, insulin resistance syndrome, and primary pachydermoperiostosis.[5–7] Secondary cutis verticis gyrata may occur at any age, and the histopathologic examination is unusual to the extent that it represents the underlying disease.[1]

Cutis verticis gyrata makes patients uncomfortable cosmetically to a serious degree and is also a benign condition. An important exception to this is the cerebriform intradermal nevus that is associated with the secondary form. Cerebriform intradermal nevus may degenerate into malignant melanoma; therefore, the lesion should be completely removed.[7]

For the treatment of cutis verticis gyrata, antihistamines, steroids, thyroid extracts, topical antiseptics, radiotherapy, sleep therapy, and psychotherapy have been attempted; however, all have resulted in failure.[8] Currently, the most effective treatment option is surgery. The lesion size, patient preferences, and the underlying diseases should be considered when planning surgical treatment.[1] Localized lesions may be excised and restored with primary closure or local flaps. For large lesions, serial excision or postexcision grafting may be considered. A tissue expander is a good option for large lesions and has better cosmetic outcomes, especially compared with grafting. The patient presented herein had skin folds over her entire scalp, with the most evident and widest folds in the occipital region. The occipital region was reported because the complaint of the patient was primarily related to this region. During the operation performed on the patient, 2 folds evident in the occipital region were excised, followed by the complete excision of the galea aponeurotica, which is distinct from what has been reported in the literature. After the galea was excised, there was a relaxation in the flaps and a slight flattening in the folds that was noticeable even during the operation. On the other hand, Snyder et al[1] recommended only the scalp excision, whereas Tani et al[9] suggested galea scoring in addition to the scalp excision. However, the presented case clearly shows that the complete excision of the galea will provide better correction of this deformity.

Cem Aslan, MD
Osman Tan, MD
Mubin Hosnuter, MD
Daghan Isik, MD
Department of Plastic
Reconstructive, and Aesthetic Surgery
Katip Celebi University School of Medicine
Ataturk Training and Research Hospital
Izmir, Turkey
cemsln@yahoo.com

REFERENCES

1. Snyder MC, Johnson PJ, Hollins RR. Congenital primary cutis verticis gyrata. *Plast Reconstr Surg* 2002;110:818–821
2. Zhang MJ, Sun ZY, Liu Y, et al. Rare giant secondary cutis verticis gyrata. *J Plast Surg Hand Surg* 2011;45:212–214
3. Polan S, Butterworth T. Cutis verticis gyrata; a review with report of seven new cases. *Am J Ment Defic* 1953;57:613–631
4. Schenato LK, Gil T, Carvalho LA, et al. Essential primary cutis verticis gyrata. *J Pediatr (Rio J)* 2002;78:75–80
5. Diven DG, Tanus T, Raimer SS. Cutis verticis gyrata. *Int J Dermatol* 1991;30:710–712
6. Woollons A, Darley CR, Lee PJ, et al. Cutis verticis gyrata of the scalp in a patient with autosomal dominant insulin resistance syndrome. *Clin Exp Dermatol* 2000;25:125–128
7. Lasser AE. Cerebriform intradermal nevus. *Pediatr Dermatol* 1983;1:42–44
8. Wyatt EH. Cutis verticis gyrata. *Clin Exp Dermatol* 1987;12:293
9. Tani T, Miyamoto Y, Kurozumi N. Surgical treatment of cutis verticis gyrata: a report of 2 cases. *Br J Plast Surg* 1977;30:235–239

Simple Resolution With Maxillary O-Ring Attachment Overdenture

To the Editor: Patients experiencing tooth loss with dentures experience reduced stability and have reduced chewing ability. In these cases, one of the treatment options is the overdenture supported by titanium implants.[1] On the other hand, in the maxillary arch, there is no consensus or treatment concept for overdenture's retained implant, although it is considered a favorable treatment in cases of insufficient bone volume and complaints about the retention and stability of the total prosthesis.[2]

Promoting greater retention and increase of stability of the final prosthesis, the use of a metal bar is commonly accepted in this mode of treatment.[3] In addition to the bars, other fastening devices may be

FIGURE 1. Initial clinical case. Occlusal view of the maxilla; RV observed 2 females regular sizes used in the ERA system distal crowns.

used, such as O-ring system. This setting has the advantage of ease of use, maintenance, and hygiene as well as being low cost and eliminating the possibility of using bar with superestructure.[4] However, the drawbacks include the use of the sealing rings with gradual loss of retention and the need for periodic replacement.[5]

In addition, fracture and loosening of the fastening system are said to be the most common complications in the treatment of maxillary and mandibular overdentures.[6] In addition, they require frequent maintenance, and in some cases, the prostheses have additional volume caused by the presence of the metal bar.[3]

This article describes a case in which a removable denture was transformed into the overdenture financial condition and fears of the patient in relation to the surgical procedure of bone grafting and installation of additional implants in the posterior maxilla.

CLINICAL REPORT

A patient aged 82 years with partially edentulous maxilla and mandible presented herself to private practice complaining about the instability of removable partial denture support after loss of a tooth in the posterior region (Figs. 1–3). On examination, it was found that the patient had good systemic condition and was visually impaired and a nonsmoker. Signs or symptoms of bruxism were not detected. In the initial appointment, several possibilities of rehabilitation with advantages and disadvantages were explained to the patient, opting for the simpler treatment plan.

At the clinical and radiographic examination, it can be observed, in the upper region, the presence of a metal-ceramic fixed partial denture between the jaws with 2 female extracoronal semi-precision dental attachments (ERA-RV attachment) with regular sizes were observed in the distal crowns (Figs. 1, 2, 3A–B). In the lower section, there was a satisfactory removable partial denture and one implant with O-ring system (Figs. 2 and 3C). The upper removable partial denture generated complaints of instability. Note that the absence engaging the left and right side fitting is presented to be damaged. The second upper-right molar tooth holder that was a prosthetic tooth was lost, and an acrylic resin was added in the second prosthesis professional before the initial query (Figs. 3A–B).

The patient was evaluated preoperatively regarding the anatomy of the jaws, bone volume ratio, jaw distance, and occlusion, and the treatment plan, which is as simple as possible, was the installation of

FIGURE 2. Initial panoramic radiograph. Presence of fixed prostheses of the upper canine and canine presence of an implant with lower attachment to a removable partial denture on tooth and implant of this O-ring.

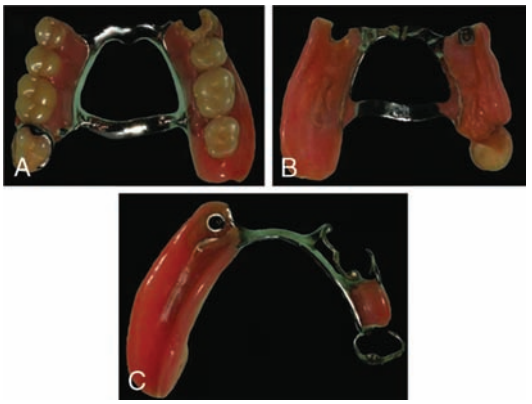

FIGURE 3. A and B, Occlusal view of the initial superior removable partial denture. Note the absence of fitting hand on one side and a snap damaged on the other side. The upper right molar that was supported with prostheses lost a tooth, and acrylic resin was added into the prosthesis. C, Inferior removable partial denture satisfactory with an O-ring system.

2 implants in the maxilla with O-ring system, with subsequent installation of an implant overdenture-type top. The lower denture was maintained by presenting as satisfactory.

The upper and lower models were mounted on the articulator. The diagnostic wax-up of the back teeth of the upper arch was performed to plan the placement of the implants and the surgical guide.

One hour before surgery placement of implants in the jaw, the patient was premedicated with 2-g amoxicillin, 4-mg dexamethasone, and 5-mg diazepam. Local anesthesia (mepivacaine of 2% with epinephrine of 1:100,000) was also administered. The surgical sequence was performed with folding flap and held the installation of a titanium implant in the region of the left upper first molar (AS 313; 3.75×13 mm; SIN - Implant System Nacional Ltda, São Paulo, SP, Brazil) and an implant in the region between the first and second upper right molars (SCOT 513; 5×13 mm; SIN - National implant System Ltda, São Paulo, SP, Brazil). Regions had greater bone density and a good bone quality.

Six months after installing the implant, it was observed that the osseointegrated implants were seen to be offered as stable, apparently immobile, for a functional graft under load without pain, inflammation, or loosening support. Given this observation, transfer molding of the implants was performed with the open tray technique and additional silicone (Futura FGM). In addition, molding of the lower jaw with the lower prosthesis was also performed, and the models were mounted in semiadjustable articulator. Registration of intermaxillary used to assemble models in the articulator was performed with the same polysiloxane used for molding. Thereafter, the molten metal frame cobalt-chromium and artificial teeth were mounted on wax. After approval by the professional and the family, because he/she is a visually impaired patient, the prosthesis was acrilized.

Before installing the prosthesis, the O-rings were installed on the implant, and the prosthesis is positioned at the mouth of the patient connected to the implant to hold O-rings to capture the male regular size RV ERA system, which have a lateral opening increased to fit the RV female that is united and merged with the coping of fixed prosthesis. It was not possible to capture the 2 metal caps of ERA system because of lack of space in the prosthesis (Fig. 4).

The metal capsule has the function to prevent the acrylic prosthesis to be damaged during the replacement of male retention, but the case presented was not possible to put the jackets for not having enough space in the overdenture prosthesis type due to its

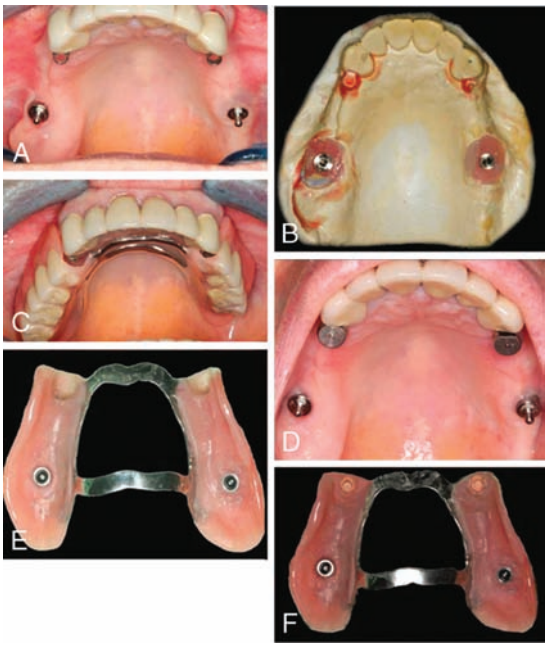

FIGURE 4. A, Occlusal view of the maxilla with 2 implants fitted with O-ring system. B, Model after transfer molding of the 2 implants. C, Overdenture type of prosthesis positioned in the mouth of the patient before capture of male regular-size RV ERA system, which has a greater lateral opening to fit the female RV that is united and merged with the fixed prosthesis coping. D, Two metal jackets positioned on the female RV ERA system. Note that the size of them is impossible to capture. E, View of the overdenture prosthesis type previous to the capture of male RV. F, View of the prosthesis-type overdenture after the capture of male RV.

magnitude. The male chosen for this case was the orange, which ensured good retention of the prosthesis.

The case has 1 year of follow-up, and the patient is satisfied (Fig. 5).

DISCUSSION

The lack of stability and retention of partial and/or complete removable dentures generate trauma to adjacent soft tissues, functional disability, and impairment in phonetics, which may cause inconvenience to social carriers of this type of prosthesis, thus increasing the appeal by dental implants.[7,8] Given the excellent prognosis, these patients can be successfully treated with retained or implant-supported dentures.[9]

In this scenario, the overdentures have become a reliable and popular alternative offering the same chewing efficiency than a fixed prosthesis,[10,11] even in situations that are presented as reported in this clinical case, in which the life expectancy of the population is increasing and patients yearn for fast, simplified, and less costly treatments. Furthermore, this type of treatment has an interesting benefit in the case of patients with advanced age, because installation of a few implants decreases the surgery time, thus reducing morbidity.

The advantages of the prosthesis and overdenture are quite clear: because the overdenture will be supported by 2 or 4 implants, the cost of installing and making the prosthesis will be limited as well as the maintenance of simplified oral hygiene.[1,2] Clinical comfort was generated by greater stability and better retention of the prosthesis with surgical need to install a few implants,[3,6] in addition to improved masticatory function and bite force compared with the conventional full denture.[12] However, because he/she was a patient with visual impairment, some setbacks have been observed as the time until the patient could insert and remove the prosthesis without difficulty, through the adaptation toward a new prosthetic condition.

The overdentures may have other drawbacks, such as the possibility of fracture of the prosthesis, the need for periodic maintenance,[3,12] and loss of fixation devices,[6] because of factors such as use, due to insertion and removal of the prosthesis by the patient; conditions of the oral environment; and masticatory forces.[13] The upper overdentures have more disadvantages relative to more consistency of trabecular bone of the jaw, reducing the ability to support and stabilize implants.[14]

An important aspect that should be pointed out is that the fasteners of type O-ring must be installed parallel to each other to avoid possible fracture at the point of lesser thickness of the system and to facilitate insertion and removal of the prosthesis.[5,15]

As shown in this report, the overdentures constitute themselves into a kind of simplified and faster treatment, because most patients wish to have the return of their masticatory function and esthetics, with compatible costs within the economic reality.

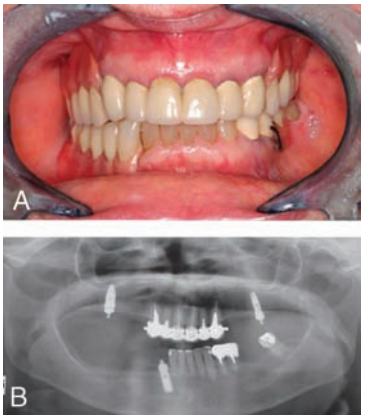

FIGURE 5. A, Front view of the final clinical case. B, Final panoramic radiograph. Observe the two upper implants with O-ring system.

Júlio César Silva de Oliveira, DDS, MSc
Department of Surgery and Integrated Clinic
Araçatuba Dental School
Univ Estadual Paulista Júlio de Mesquita Filho
Araçatuba - UNESP, São Paulo, Brazil

Eloá Rodrigues Luvizuto, DDS, MSc
Department of Surgery and Integrated Clinic
Araçatuba Dental School
Univ Estadual Paulista Júlio de Mesquita Filho
Araçatuba - UNESP, São Paulo, Brazil
eloaluvizuto@hotmail.com or eloa@foa.unesp.br

Thallita P. Queiroz, DDS, MSc
Department of Health Sciences
Dental School
University Center of Araraquara - UNIARA
São Paulo, Brazil

Walter Betoni-Júnior, DDS, MSc
Department of Oral Surgery
Dental School
University of Cuiabá UNIC
Mato Grosso, Brazil

Allan Costa e Silva, DDS
Especialist of Implantology
School of Dentistry of Cuiabá
Mato Grosso, Brazil

REFERENCES

1. Batenburg RHK, Raghoebar GM, Van Oort RP, et al. Mandibular overdentures supported by two or four endosteal implants: a prospective, comparative study. *Int J Oral Maxillofac Surg* 1998;27:435–439
2. Slot W, Raghoebar GM, Vissink A, et al. A systematic review of implant-supported maxillary overdentures after a mean observation period of at least 1 year. *J Clin Periodontol* 2010;37:98–110
3. Uludag B. Fabrication of a fractured mandibular bar-retained implant overdenture without implant-level impression making: a clinical report. *J Prosthodont* 2012;21:622–625
4. Baker PS, Ivanhoe JR. Fabrication of occlusal device for protection of implant overdenture abutments with O-ring attachments. *J Prosthet Dent* 2003;90:605–607
5. Winkler S, Piermatti J, Rothman A, et al. An overview of the o-ring implant overdenture attachment: clinical reports. *J Oral Implantol* 2002;28:82–86
6. Slot W, Raghoebar GM, van Dijk G, et al. Attachment of clips in a bar-retained maxillary implant overdenture: a clinical report. *J Prosthet Dent* 2012;107:353–357
7. Patras M, Kourtis S, Sykaras N. Creating natural-looking removable prostheses: combining art and science to imitate nature. *J Esthet Restor Dent* 2012;24:160–168
8. Scherer MD, McGlumphy EA, Seghi RR, et al. Comparison of retention and stability of two implant-retained overdentures based on implant location. *J Prosthet Dent* 2014;112:515–521
9. Andreiotelli M, Att W, Strub JR. Prosthodontic complications with implant overdentures: a systematic literature review. *Int J Prosthodont* 2010;23:195–203
10. Fueki K, Kimoto K, Ogawa T, et al. Effect of implant-supported or retained dentures on masticatory performance: a systematic review. *J Prosthet Dent* 2007;98:470–477
11. de Grandmont P, Feine JS, Tache R, et al. Within-subject comparisons of implant-supported mandibular prostheses: psychometric evaluation. *J Dent Res* 1994;73:1096–1104
12. Alsabeeha NH. Elders with implant overdentures: a 22-year clinical report. *Gerodontology* 2012;29:239–242
13. Saavedra G, Barbosa SH, Landin KT, et al. Influência do ângulo de inserção na degradação da retenção do O-ring em overdenture. *Implant News* 2007;4:249–254
14. Sadowsky SJ. Mandibular implant-retained overdentures: a literature review. *J Prosthet Dent* 2001;86:468–473
15. Saavedra GSFA, Zogheib LV, Kimpara ET, et al. Conceitos atuais do sistema de encaixe overdenture sobreimplante. *Implant News* 2008;5:611–618

Repair of the Complicated Orocutaneous Fistula Using Atelocollagen Sponge

To the Editor: When the orocutaneous fistula is not surgically removed, it may invade or erode underlying alveolar bone, resulting in bone infection, erosion, and bony deformity. A 14-year-old girl presented to a local dermatology clinic 3 months prior with a chief complaint of recurrent wound over the chin. The patient reported that the lesion would partially resolve over the course of several months but recur intermittently. The lesion was initially diagnosed as a pyogenic granuloma. The patient subsequently underwent treatment with a pulse dye laser on 2 separate occasions, with only partial resolution. Reevaluation 4 months later demonstrated full recurrence of the lesion, with intermittent bleeding. This patient was referred to my department. The cystic erythematous papule-like skin tumor had grown gradually to 20×20 mm. Preoperative contrast-enhanced computed tomography findings revealed that a fistula existed just above the bone and that the underlying bone was eroded. Intraoral finding showed dental caries. This time the lesion had wider and deeper margins, and a friable tract of tissue extending into the region of the left mandibular body was found. Small artificial dermis sponge was inserted into the extracted site of the left molar alveolar socket to improve bony healing, seal the gap of the mandibular bone, and divide the intraoral and extraoral cavity. And then, the exposed bony lesion was covered by buccal mucosal flap. The lesion healed quickly and uneventfully over 2 weeks with only minor scarring of the cutaneous lesions. Three months after the operation, the texture of the implanted region is almost the same as the contralateral side and is almost symmetrical. Cutaneous draining sinus tracts were typically formed by periapical abscesses, which resulted from inflammatory degeneration of the pulp and periodontal membrane of the tooth.[1] Synthetic materials, fibrillar collagen-like type I collagen, are the major constituent of the extracellular matrix and structural protein of the bone.[2] The results of this case suggest that absorbable atelocollagen sponge is relatively a favorable bone void filler that prevents tissue collapse and food packing and enhances periodontal healing.[2] When the dermal sponge was placed into the defect, it becomes soft and reduces in size due to infiltration of exudative fluid from the surrounding tissue. The artificial dermis sponge (Teruplug; Terumo Co Ltd, Tokyo, Japan) used in this study is composed of a thick atelocollagen sponge (small, 8×25 mm; medium, 15×25 mm). Despite the fact that chronic dental infections are the most common cause of draining cutaneous sinus tracts in the face and neck, the condition is still commonly misdiagnosed, and misdiagnosis often leads to prolonged and ineffective therapy. Because of the nature of these cutaneous lesions and the lack of associated dental symptoms in the majority of cases, patients often initially consult primary care providers and dermatologists. In conclusion, the author believes that artificial dermis sponge may be a useful implantation material to correct alveolus depression and make a barrier between the intraoral and extraoral cavity in the patient with infected and complicated odontogenic fistula (Fig. 1).

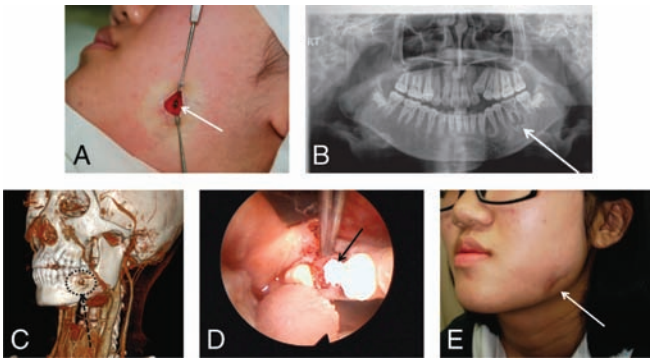

FIGURE 1. Case presentation. A, An elliptical skin incision was made. Arrow indicates the fistula between skin and first molar. B, Panoramic view shows radiolucency of the apical space of the first molar. The indicator shows the first molar lesion. C, Three-dimensional contrast enhanced computed tomography scan shows bony erosion of the outer cortex of the mandible. D, Intraoral endoscopic finding reveals bony destruction after first molar extraction. Black arrows show small atelocollagen sponge in the exposed socket and outer bony opening. E, Three-month postoperative finding shows well-healed opening of the orocutaneous fistula and surgical scar.

Hwan Jun Choi, MD, PhD
Department of Plastic and Reconstructive Surgery
College of Medicine, Soonchunhyang University
Cheonan, Korea
medi619@hanmail.net

ACKNOWLEDGEMENT

This work was supported by Soonchunhyang University Research Fund.

REFERENCES

1. Baumgartner JC, Picket AB, Muller JT. Microscopic examination of oral sinus tracts and their associated periapical lesions. *J Endod* 1984;10:146–152
2. Oh HY, Choi HJ, Kwon JS, et al. Usefulness of atelo-collagen sponge (Teruplug) for treatment of mandibular angle fractures with third molar extraction. *J Korean Soc Plast Reconstr Surg* 2011;38:1550–1560

Kuttner Tumor

Characterized as a chronic inflammatory lesion, Kuttner tumor (KT) or chronic sclerosing sialoadenitis is a rare glandular disorder. It manifests as a glandular tumefaction with a firm consistency, generally unilateral, which may be asymptomatic or associated with pain while eating.[1–3] Sialolithiasis is its etiological factor most commonly reported.

A 57-year-old black man complained of increased volume in the right submandibular region, diminished salivation, and great difficulty with swallowing associated with pain, with a 2-month history of evolution. A tumefaction of firm consistency to palpation (Figs. 1A–C). His medical history included cardiomyopathy, with a report of infarction 7 years ago, alcoholism and cigarette smoking for more than 30 years. On initial radiographic examination, it was possible to visualize a circular, well-delimited radiopaque image, suggestive of calcification (Fig. 2A). In the computed tomograph, there was an increase in volume of the right submandibular gland, presence of a circular, hyper-attenuated image within the gland measuring approximately 1.3 cm and upstream ductal ectasia, suggestive of salivary calculus (Fig. 2B). Ultrasonography showed the submandibular gland with altered dimensions, irregular contours, a diffusely heterogeneous echotexture, and exhibiting a hyperechogenic image associated with a posterior acoustic shadow measuring approximately 2 cm at its largest axis in the projection of the duct, determining upstream dilatation.

The treatment proposed was right submandibular salivary gland resection (Fig. 3A). Histopathological analysis of the surgically removed tissue showed evidence of a chronic inflammatory process, lymphoplasmacytic infiltrate, atrophy of the glandular parenchyma, and dilated excretor duct containing a sialolith within it (Fig. 3B). The diagnosis was concluded as KT, associated with the presence of salivary calculus, configuring a sialolithiasis.

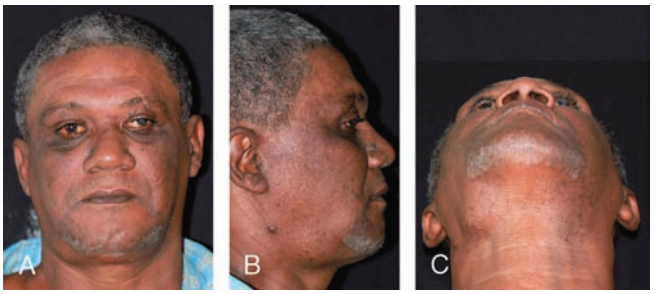

FIGURE 1. Extraoral clinical images highlighting the submandibular swelling on the right side: front (A), profile (B), and inferior-superior (C).

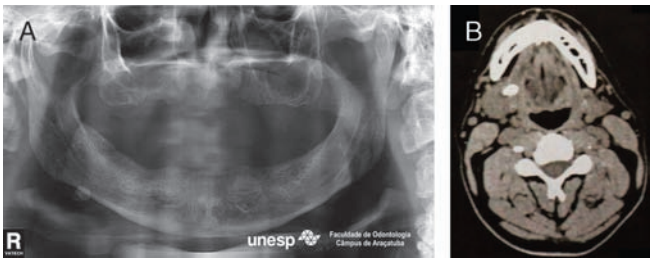

FIGURE 2. Initial panoramic radiograph (A) and CT (B) showing circular hyperattenuating images within the right submandibular gland.

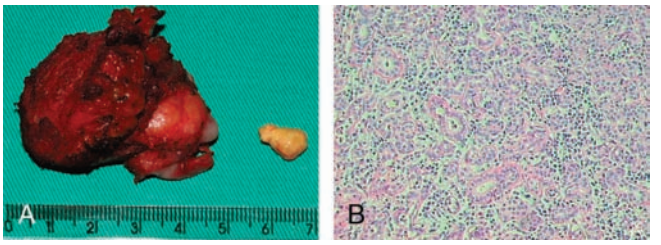

FIGURE 3. Images of the gland surgically removed for histopathological analysis (A) and the result of the analysis (B).

This case was similar to the cases of Kuttner.[4] In this case, there was no resolution of the swelling during the fasting period; therefore, it is most unlikely that the clinical progression of the disease was related to chewing. The literature has shown that removal only of the sialolith may resolve the submandibular edema; however, in 70% of the cases, the swelling persists.[5] Therefore, the treatment proposed was submandibular gland resection. To sum up, KT is easily distinguishable from a neoplasia, and it has no clinical and histological characteristics of malignancy. Although denominated as ''tumor,'' it is not a neoplasia but a benign process that clinically confuses and perturbs professionals by the extension and consistency of its tumefaction. In this case, clinical and histopathological characteristics are compatible with KT, and the presence of sialolith suggests sialolithiasis as its etiological factor.

Sabrina Ferreira, DDS, MS
Francisley Ávila Souza, DDS, MS
Leonardo Perez Faverani, DDS, PhD
Bianca Bravim Bomfim, DDS, MS
Idelmo Rangel Garcia Júnior, DDS, MS
Division of Surgery and Traumatology Oral Maxillofacial
Department of Surgery and General Clinic
Araçatuba Dental School
Univ Estadual Paulista Júlio de Mesquita Filho—UNESP
Brazil
sabrife@bol.com.br

Renata Callestini Felipini, DDS, PhD
Division of Pathology
Araçatuba Dental School
Univ Estadual Paulista Júlio de Mesquita Filho—UNESP
Brazil

Everton Pontes Martins, MD, PhD
Santa Casa de Misericórdia
Araçatuba, São Paulo, Brazil

REFERENCES

1. Williams HK, Connor R, Edmondson H. Chronic sclerosing sialadenitis of the submandibular and parotid glands: a report of a case and review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2000;89:720–723
2. Markou K, Karasmanis I, Vlachtsis K, et al. Chronic sclerosing sialadenitis mimicking tumors of the salivary glands. *Eur J Surg Oncol* 2007;33:1207–1208
3. Adachi M, Fujita Y, Murata T, et al. A case of Kuttner tumor of the submandibular gland. *Auris Nasus Larynx* 2004;31:309–312
4. Kuttner H. Uber entzundiche Tumoren der Submaaxillar-speicheldruse. *Bruns Beitr Klin Chir* 1896;15:815–834
5. Ahuja AT, Richards PS, Wong KT, et al. Kuttner tumour (chronic sclerosing sialadenitis) of the submandibular gland: sonographic appearances. *Ultrasound Med Biol* 2003;29:913–919

Response To "Mean Platelet Volume May Not Be Beneficial for the Management of Patients With Adenoid Hypertrophy"

Dear Editor: We are pleased with the interest in our article and appreciate the comments that provide significant information.[1] First of all, we agree with Dr Beyan regarding the methodological concern of mean platelet volume (MPV) assessments. Blood samples must be studied within 2 hours, as in our study, to avoid falsely elevated results due to EDTA.[2] In addition, when commenting on MPV values in different studies, one must be cautious about the analyzer device. Different analyzer devices might produce different MPV results,[3] and this fact should be taken into account before generalization of the results. Therefore, we used the same analyzer device in our study.

Second, MPV is considered to be a marker and determinant of platelet function because larger platelets are hemostatically more reactive than platelets of normal size. Although it is obvious that MPV is not a platelet function test, several studies have shown that elevated MPV is associated with increased platelet reactivity.[4,5] Thus, platelet volume has been proposed as an indirect marker of increased platelet reactivity. Moreover, our main driving force behind this study was to find a useful marker for severe nasal airway resistance due to adenoid hypertrophy.

Cüneyt Kucur, MD
Fatih Oghan, MD
Department of Otorhinolaryngology
Head Neck Surgery
Dumlupinar University Faculty of Medicine
Kutahya, Turkey
ckucur@mynet.com

REFERENCES

1. Beyan C, Beyan E. Mean platelet volume may not be beneficial for the management of patients with adenoid hypertrophy. *J Craniofac Surg* 2014;25:1580–1581
2. Dastjerdi MS, Emami T, Najafian A, et al. Mean platelet volume measurement, EDTA or citrate? *Hematology* 2006;11:317–319
3. Lancé MD, van Oerle R, Henskens YM, et al. Do we need time adjusted mean platelet volume measurements? *Lab Hematol* 2010;16:28–31
4. Leader A, Pereg D, Lishner M. Are platelet volume indices of clinical use? A multidisciplinary review. *Ann Med* 2012;44 (8):805–816
5. Sansanayudh N, Anothaisintawee T, Muntham D, et al. Mean platelet volume and coronary artery disease: a systematic review and meta-analysis. *Int J Cardiol* 2014;175:433–440

Treatment of a Central Incisor Crown-Root Fracture With Buccal Wall Reconstruction and Immediately Loaded Titanium Implant

To the Editor: Dental traumas are considered an increasing public health problem, unlike dental caries that have been declining over the past decades. Most dental traumas occur in the maxillary anterior teeth. The main causes of such traumas are sport injuries, violence, falls, and motor vehicle crashes; other causes may also exist depending on a country's development and local habits.

Crown-root fractures are defined as those that involve cement, enamel, and dentin and may or may not involve the pulp. The relative frequency increases across age groups (children, 1.8%; adolescents, 6.3%; and adults, 9.2%).[1] Diagnosis is made through clinical and radiographic examinations, the latter frequently being limited by the position and extension of the fracture. Treatment options for such cases include the following: (1) orthodontic or surgical extrusion, (2) gingivectomy and osteotomy/osteoplasty, (3) intentional replantation, and (4) extraction.

When tooth extraction is the only treatment option in cases of crown-root fractures of anterior teeth, tooth loss indicates a substantial increase in the number of aesthetic difficulties. This is caused primarily by the association of alveolar bone loss.[2] Filling an anterior space with an implant-supported porcelain crown is a major challenge from both aesthetic and functional aspects. Clinical success depends not only on persistent osseointegration but also on harmonious integration of the crown in the dental arch.[3]

Buccal wall defects after tooth extraction especially in cases of crown-root fractures are frequent in the anterior portion of maxilla. Common reasons for such defects include the impact of the trauma on a thin buccal bone. Regeneration of the postextraction defect with vital, well-vascularized, dense bone is critical to a successful implant-supported restoration.[4] The aim of this study was to relate a clinical case of a crown-root fractured of an upper left central incisor in which the buccal bone wall was lost. The clinical case was solved with the extraction of the tooth, immediate implant placement, and reconstruction of the buccal bone wall with autogenous bone graft removed from the maxillary tuber previous to immediately loaded titanium implant.

CASE REPORT

The patient, a 24-year-old man with good medical health, sought attendance at the Clinic of the Implantology Course for treatment of a dental traumatism after being victim of a stone against the face. The patient has complained about discomfort and inability to smile due to a fractured left maxillary central incisor. Clinical examination has shown a tooth fractured with some fragments of fractured enamel and/or dentin lost and others attached only by soft tissue (Fig. 1A). Radiographic examination of the same tooth has shown root canal treatment with an intracanal post and an extensive crown-root fracture (Fig. 1B).

Owing to extension of the crown-root fracture, the treatment plan proposed was to install and load an implant immediately after tooth

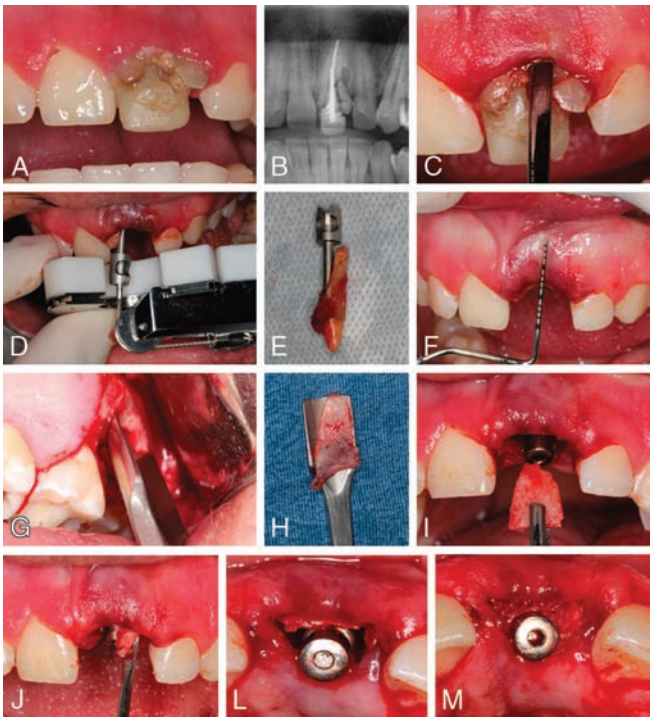

FIGURE 1. Initial clinical case (A) and initial panoramic radiograph (B). Observe the left central incisor with crown-root fracture; extraction of the left central incisor: sindesmotomy (C); extraction device (Neodent, Curitiba, Paraná, Brazil) inserted into the root canal to extract the tooth (D); tooth extracted (E); 7 mm of buccal wall defect (F); blade of bone been removed with the aid of a gouge chisel (G); blade of bone removed (H); blade of bone of the size of the buccal wall defect has been inserted on the buccal side of the socket after implant placement (I); slice of bone inserted (J, L); particulate autologous bone graft inserted inside the gaps between the implant and the slice of bone (M).

extraction followed by buccal bone wall reconstruction with a blade of autogenous bone graft to exert mechanical barrier function, and bone particles, both removed from maxillary tuber. After local anesthesia (2% mepivacaine/adrenaline, 1:100,000), a less traumatic extraction was performed using an extraction device (Neodent, Curitiba, Paraná, Brazil). Initially, root preparation with a 1.3-mm helical cutter was done to install a 1.6-mm tractor using the digital key and delicate sindesmotomy, without making incisions. Then the steel cable was installed in the tractor perpendicular to the tooth, and the whole extractor was set up. To conduct root extraction, the lever is rotated clockwise, causing the steel cable attached to the tractor to remove the root while preserving the surrounding structures (Figs. 1C–E).

Right after tooth extraction, the implant (Conect AR, 3.75 × 13 mm; Conexão Sistemas de Prótese, Arujá, São Paulo, Brazil) was installed with 50 N·cm performing an approach in the palatine wall. As you can see in Figure 1F, buccal bone wall has been lost in 7 mm of depth. To reconstruct the large postextraction buccal wall defect, we performed a surgical access in the region of the right maxillary tuberosity for removal of a bone blade. A supernumerary tooth was observed in this region during the sindesmotomy, which was then extracted, and the alveolar buccal wall of this element was used to remove the bone blade (Fig. 1G) with the aid of an 8-mm gouge chisel (Fig. 1H). Subsequently, the removed blade of cortical bone wedge shape was prepared with scissors and adapted between the buccal mucosa and the installed implant (Figs. 1I–L). Particulate autologous bone graft, also removed from the right tuber, was inserted inside the gaps between the implant and the blade of bone to fill the empty space and to give support to the blade of bone (Fig. 1M).

As medication therapy, antibiotic (amoxicillin, 500 mg, every 8 hours for 7 days) and analgesic (paracetamol, 100 mg, every 8 hours while pain persisted) were prescribed. Transfer molding was performed, and the implant was restored with a temporary restoration, which was adjusted and installed on the same day of the implant placement applying 20 N·cm of torque. Soft tissue was sutured with silk threat 5-0 (Figs. 2A, B).

After 6 months of follow-up, a new transfer molding was performed to customize the permanent all-ceramic implant restoration (CAD/CAM, Precision, Conexão Sistemas de Prótese, Sao Paolo, Brazil). The achieved gingival contour at this period can be observed on Figure 2C. Satisfactory results have been shown with regard to soft and hard tissues wound healing and aesthetic prosthetic restoration. At 2 years after loading, the implant-supported restoration remained completely functional with no evidence of complication. Computed tomography revealed excellent bone-to-implant contact along the entire implant body (Figs. 2D–E).

DISCUSSION

Restoration of a failing single tooth is currently probably the most common indication for implant insertion. Frequently at the time of implant insertion, bone augmentation is needed to restore the buccal wall integrity, to enhance implant survival and success, or to increase soft tissue support in demanding esthetic cases. A number of therapeutic approaches to restore the buccal bone wall integrity have been used, including different grafting materials such as beta-tricalcium phosphate matrix,[5] hydroxyapatite ceramic matrix, and bovine bone mineral matrix, among others. In addition, the association of biomaterials with growth factors like bone morphogenetic proteins,[6] a platelet-derived growth factor,[7] is becoming popular. However, when the amount of bone required is limited, autogenous bone graft removed from the maxillary tuber after supranumerary extraction is a viable alternative and is costless.

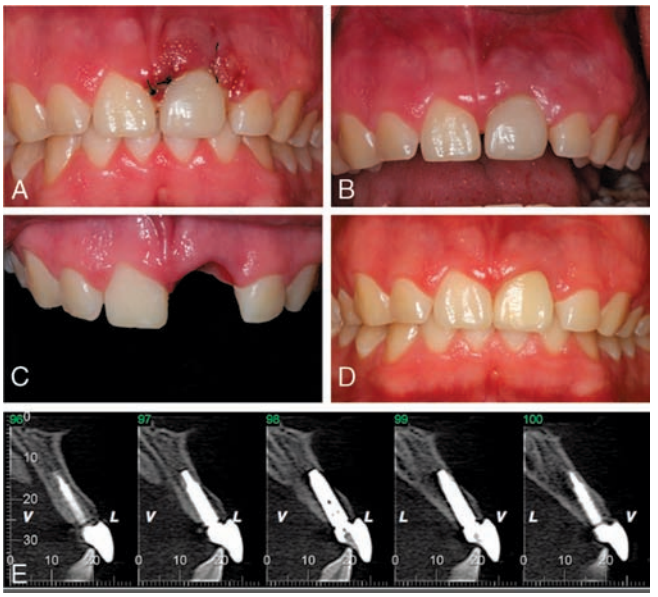

FIGURE 2. Provisional crown adjusted and installed on the same day of the implant installation (A); clinical case at 7 days postoperative (B); gingival contour achieved after 6 months postoperative (C); clinical case at 2 years postoperative with the final ceramic metal-free implant crown (CAD/CAM) (D); final computed tomography slices at 2 years postoperative: transaxial slices (E).

Regeneration of well-vascularized vital bone is critical to long-term positive outcomes when managing large postextraction buccal wall defects.[4] In this case presented, the autogenous bone graft was used as an osteogenetic, osteoinductive, and osteoconductive material. This method has been shown to be effective for bone regeneration in the treatment of atrophic edentulous ridges.

Because of the highly esthetic nature of this area, proper management of this site was critical. Considering the need for exceptional results, it was decided that the crown-root fractured maxillary left central incisor would be removed and treated with flapless immediately loaded titanium implant placement associated with the reconstruction of the buccal bone wall with autogenous bone graft.

Sufficient initial implant stability, one of the main prerequisites for immediate loading, was achieved in this clinical case. Provisional restoration was placed after implant surgery to create soft tissue contours that resemble normal gingival topography before placement of the definitive prosthesis and to avoid additional surgical operation or other soft tissue manipulation.[8] For aesthetic reasons, the authors have opted for all-ceramic restoration.

As described in literature, trimodal approach protocol (immediate postextraction placement, flapless, and immediate provisional restoration) is a reliable and simple protocol to place and restore immediate implants in the aesthetic zone giving back to the patient good aesthetic outcome with minimal complications.[9] The case reported herein added an autologous bone graft procedure removed from the tuber to this protocol, giving the opportunity to think about establishment of a new protocol called quadrimodal approach protocol. However, studies are needed to verify long-term aesthetic results with this approach and to define and quantify biotypes.

In the case presented herein, the authors can conclude that the placement of implant immediately after tooth extraction combined with immediate prosthetic load placement and buccal wall reconstruction with autogenous bone graft removed from the maxillary tuber, enables the maintenance of alveolar bone height and architecture of the gingival tissues, providing excellent, natural-appearing restorations.

Thallita P. Queiroz, DDS, MS
Rogério Margonar, DDS, MS
Department of Health Sciences
Implantology Post Graduation Course, Dental School
University Center of Araraquara—UNIARA
São Paulo, Brazil

Eloá R. Luvizuto, DDS, MS
Jonatas Caldeira Steves, DDS, MS
Department of Surgery and Integrated Clinic
Araçatuba Dental School
UNESP—Univ Estadual Paulista
São Paulo, Brazil
eloaluvizuto@hotmail.com

Walter Betoni-Júnior, DDS, MS
Department of Oral Surgery
Dental School
University of Cuiabá—UNIC
Mato Grosso, Brazil

Celso Koogi Sonoda, DDS, MS
Department of Surgery and Integrated Clinic
Araçatuba Dental School
UNESP—Univ Estadual Paulista
São Paulo, Brazil

REFERENCES

1. Lauridsen E, Hermann NV, Gerds TA, et al. Pattern of traumatic dental injuries in the permanent dentition among children, adolescents, and adults. *Dent Traumatol* 2012;28:358–363
2. Margonar R, Queiroz TP, Luvizuto ER, et al. Anterior tooth rehabilitation with frozen homogenous bone graft and immediately loaded titanium implant using computer-guided surgery. *J Craniofac Surg* 2012;23: e470–e472
3. Jung RE, Pjetursson BE, Glauser R, et al. A systematic review of the 5-year survival and complication rates of implant-supported single crowns. *Clin Oral Implants Res* 2008;19:119–130
4. Snyder MB. Treatment of a large postextraction buccal wall defect with mineralized allograft, beta-TCP, and rhPDGF-BB: a growth factor-mediated bone regenerative approach. *Int J Periodontics Restorative Dent* 2012;32:705–711
5. Luvizuto ER, Queiroz TP, Margonar R, et al. Osteoconductive properties of beta-tricalcium phosphate matrix, polylactic and polyglycolic acid gel, and calcium phosphate cement in bone defects. *J Craniofac Surg* 2012;23:e430–e433
6. Luvizuto ER, Tangl S, Zanoni G, et al. The effect of BMP-2 on the osteoconductive properties of beta-tricalcium phosphate in rat calvaria defects. *Biomaterials* 2011;32:3855–3861
7. Nevins M, Camelo M, Nevins ML, et al. Growth factor–mediated combination therapy to treat large local human alveolar ridge defects. *Int J Periodontics Restorative Dent* 2012;32:263–271
8. Biggs WF, Litvak AL Jr. Immediate provisional restorations to aid in gingival healing and optimal contours for implant patients. *J Prosthet Dent* 2001;86:177–180
9. Cabello G, Rioboo M, Fabrega JG. Immediate placement and restoration of implants in the aesthetic zone with a trimodal approach: soft tissue alterations and its relation to gingival biotype. *Clin Oral Implants Res* 2013;24:1094–1100

Tracheal Metastasis

To the Editor: Endobronchial and endotracheal metastases are extremely rare and they have been associated with various thoracic and extrathoracic malignancies.[1–4] Whereas extrapulmonary endobronchial metastases are infrequent, endotracheal metastases are even more rare, occurring in only 5% of patients with extrapulmonary endobronchial metastases.[3,4]

The most common tumors that can determine endobronchial metastases are breast, colorectal, and kidney carcinomas.[1–4]

Endobronchial metastasis is usually a late finding in the course of a patient's disease.[1–4] In particular, the exact mechanism for the production of airway metastases is unclear but most likely involves migration of malignant cells along lymphatics into submucosal tissue.[3,4]

A 64-year-old man with a history of myocardial infarction was hospitalized in July 2005 for sudden intestinal occlusion. A neoplastic stenosis 10 cm from the splenic flexure was detected. Computed tomography (CT) of the chest showed 3 lung metastases localized in the right lung and a 1-mm lesion in the left lung. A left hemicolectomy was performed, and the pathological examination showed a medium-low differentiated adenocarcinoma invading the wall of the large bowel up to the serosal surface. Of 21 nodes examined, 11 were positive for neoplastic invasion (pathological staging pT3 pN2). Chemotherapy with irinotecan, bolus fluorouracil, and leucovorin was administered, using the IFL regimen. After 12 courses, a partial remission of the lung nodules was demonstrated. In an 18-Fluoro-deoxyglucose positron emission tomography (FDG-PET) scan performed at the end of the treatment, the pulmonary nodules were positive and no extrapulmonary lesions were evident. Surgical exploration and metastasectomy of 4 nodules

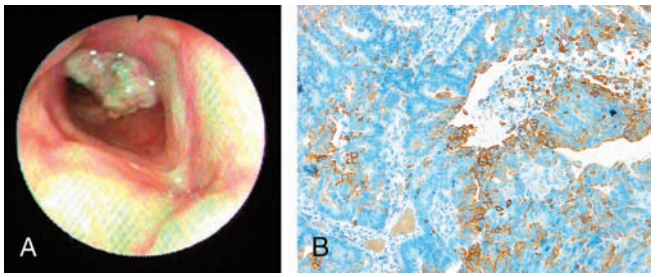

FIGURE 1. A, Endoscopic image showing a nodule bulging from the posterior side of the trachea. B, Histological image consistent with a typical large-bowel adenocarcinoma.

through thoracotomy was performed in February 2006. Histologically, 3 lesions were confirmed to be colon cancer metastases, and one was a hamartochondroma. During the following 9 months, a 5-mm left lung nodule was observed by CT scan, and it slowly increased in size. A new FDG-PET scan performed at 1 year confirmed the presence of the left lung lesion and also revealed a glucose uptake in the tracheal wall between glottic plan and aortic arch. A fiberoptic endoscopy showed a nodule bulging from the posterior side of the trachea without erosion of the adjacent cartilage (Fig. 1A). The patient did not complain of any symptom. Laser excision was performed after a preventive tracheotomy, and postoperative radiation was added. Histological examination showed a typical large-bowel adenocarcinoma with immunohistochemical features consistent with the known primary tumor (Fig. 1B). A follow-up CT scan showed a complete disappearance of the tracheal node but a further increase of the left lung lesion, so that a videothoracoscopic resection was performed. Owing to patient's refusal, no additional chemotherapy was administered, and he is free of disease at 15 months from surgery.

Generally, tracheal metastases appear in a widespread disease from solid tumors. Frequent imaging with CT and, more recently, use of PET scan can detect unusual metastatic sites preceding the appearance of symptoms. The symptoms associated with endotracheal and endobronchial metastases usually consist in hemoptysis and cough.[1,2] At least, palliative treatment is warranted to prevent such a worrisome condition.[3] Radiotherapy and chemotherapy are the treatments of choice to achieve palliation. In our patient, presence of resectable lung nodule with absence of widespread disease prompted the adoption of mini-invasive surgical approach followed by radiation. Resection of metastasis located in the main tracheabronchial tree is comparable to parenchymal disease.[4] The surgical approach to tracheal lesions can be particularly demanding and seems justifiable in case of primary lesions. Laser resection, although potentially nonradical, is an optimal palliative method. In our patient, resection followed by radiotherapy was successful in achieving long-term survival. Last generation chemotherapy, used as induction therapy before surgery or as adjuvant treatment, demonstrated clear survival benefits as far as liver resection is concerned.[1–4] Such results probably apply to the less frequent condition of isolated lung metastases. Our patient had been treated with active chemotherapy after colorectal resection and refused any other systemic treatment after detection of metastasis.

In conclusion, we reported the diagnostic process and management of a rare condition of colorectal cancer with related pulmonary and tracheal metastasis, and we suggest that aggressive local treatment results both in successful palliation and long-term survival.

Carlo Droz Blanc, MD
Giovanni Donati, MD
Emanuela Carbone, MD

Division of Thoracic Surgery
Aosta Hospital, Aosta, Italy

Stefano Righi, MD
Paolo Boffano, MD
Divisions of Otolaryngology,
Maxillofacial Surgery and Dentistry
Aosta Hospital, Aosta, Italy
paolo.boffano@gmail.com

Anna Stella, MD
Gianmauro Numico, MD
Division of Medical Oncology
Aosta, Italy

Pierpaolo Carassai, MD
Division of Pathology
Aosta Hospital, Aosta, Italy

REFERENCES

1. Watanabe] S, Oda M, Ohta Y, et al. Endotracheal metastasis of rectal cancer. *Eur J Cardiothorac Surg* 2002;21:924
2. Salud A, Porcel JM, Rovirosa A, et al. Endobronchial metastatic disease: analysis of 32 cases. *J Surg Oncol* 1996;62:249–252
3. Petru E, Friedrich G, Pickel H, et al. Life-threatening tracheal metastasis complicating ovarian cancer—a case report. *Gynecol Oncol* 1999;74:141–142
4. Kiryu T, Hoshi H, Matsui E, et al. Endotracheal/endobronchial metastases: clinicopathologic study with special reference to developmental modes. *Chest* 2001;119:768–775

Nasal Septum Perforation Repair Using a Split Septal Cartilage Graft

To the Editor: There are many options for the treatment of septal perforation, both surgical or nonsurgical. Treatment may be conservative, surgical, or prosthetic. The literature describes a number of techniques for surgically closing a nasal septal perforation.[1] Surgical techniques have not been standardized. Grafts and flaps used for the treatment may cause donor site morbidity. Alloplastic materials may cause foreign body reactions and recurrence.

Although there is no standard classification system for the perforation size, the upper limit of small size differs between authors, up to 5 mm or 1 cm.[1] Small to moderate perforations can be repaired with local advancement flaps alone or combined with interposition grafts.[2,3] Although there is still a controversy over interpositional graft necessity for nasal septal perforation repair, it seems that they reduce reperforation.[4] Although high success rates with interposition of grafts have been repatedly reported, cartilaginous remnants inside the nose are usually insufficient and low quality. Autologous auricular cartilage are mostly required in such cases. We used a simple method for the treatment of one of our patients. A 52-year old man presented with a 1-cm septal perforation in the posteroinferior membraneous septum causing epistaxis and whistling sound for 10 years. He had a history of long-term use of topical nasal decongestants. He underwent a nasal surgery under general anesthesia at Gazi University Hospital. Exposure of the perforation site was achieved via external rhinoplasty technique. Septal cartilage was removed completely (Fig. 1)

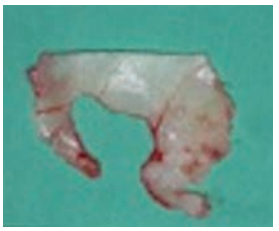

FIGURE 1. Completely removed cartilaginous septum.

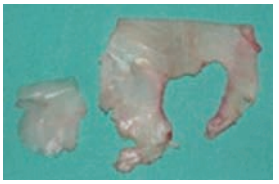

FIGURE 2. Split septal cartilage.

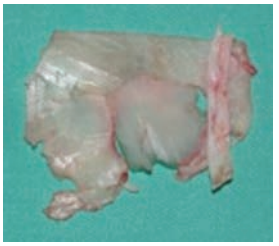

FIGURE 3. Repair of cartilagenous septum using split cartilage.

FIGURE 4. Mucosal closure.

and the posterior part of that was split (Fig. 2). We achieved a new cartilage graft and used it for the closure of the perforation (Fig. 3). The new graft was attached to the edge of the perforation site with absorbable suture material. Next, bilateral mucosal advancement flaps were used for the mucosal repair (Fig. 4). Postoperatively, there has been no early complication and no recurrence after 6 months of follow-up. We believe that small perforations can be treated with split septal cartilagenous graft and bilateral mucosal advancement flap in patients who have thick and strong septal cartilage without a necessity for another graft. This method has the following advantages: (1) decrease donor site morbidity and (2) no foregin body reactions.

Safak Uygur, MD
Department of Plastic Surgery
Koc University Hospital
Istanbul, Turkey
safakuygur@gmail.com

Mubin Aral, MD
Department of Plastic Surgery
Yenimahalle Government Hospital
Ankara, Turkey

Sebahattin Kandal, MD
Piuplast Aesthetic Center
Istanbul, Turkey

Suhan Ayhan, MD
Department of Plastic Surgery
Gazi University Medical School
Ankara, Turkey

REFERENCES

1. Kim SW, Rhee CS. Nasal septal perforation repair: predictive factors and systematic review of the literature. *Curr Opin Otolaryngol Head Neck Surg* 2012;20:58–65
2. Andre RF, Lohuis PJ, Vuyk HD. Nasal septum perforation repair using differently designed, bilateralintranasal flaps, with nonopposing suture lines. *J Plast Reconstr Aesthet Surg* 2006;59:829–834
3. Li F, Liu Q, Yu H, et al. Pedicled local mucosal flap and autogenous graft for the closure of nasoseptal perforations. *Acta Otolaryngol* 2011;131:983–988
4. Goh AY, Hussain SSM. Different surgical treatments for nasal septal perforation and their outcomes. *J Laryngol Otol* 2007;121:419–426

Fat-Removal Orbital Decompressing for Severe Bilateral Proptosis due to Weber-Christian Disease

To the Editor: Weber-Christian disease (WCD) was first described by Pfeifer in 1892 and depicted as a clinical presence by Weber and Christian in the 1920s.[1] It is also known as idiopathic nodular nonsuppurative panniculitis and is characterized by relapsing subcutaneous inflammatory painful nodules and frequent constitutional symptoms such as fever, malaise, general fatigue, polyarthralgia, and polymyalgia.[1,2] Weber-Christian disease presents in the extremities and trunk mainly.[1,2] It is commonly seen in women aged 30 to 60 years.[1,2] Lobular panniculitis is the main characteristic in biopsy materials from the nodules.[1,2] Anterior and posterior uveitis,[3] macular hemorrhage,[4] as well as episcleral and conjunctival nodules[5] are ocular manifestations of WCD. Proptosis, which is caused by panniculitis of orbital fat tissue, is a rare manifestation of WCD.[6] In our case, we have described fat-removal orbital decompressing for severe bilateral proptosis due to WCD.

A 59-year-old woman with bilateral proptosis presented to our clinic. The patient had been diagnosed with WCD before. She was being followed up in a dermatology clinic. She had bilateral eyelid ptosis. Bilateral eyelid ptosis and proptosis were determined on physical examination (Fig. 1). Computed tomographic scan was performed (Fig. 2). The patient was prepared for surgery.

The skin incision in the lateral (temporal) half of the upper eyelid was designed in a curvilinear fashion with the medial portion being convex superiorly and the lateral portion in line with the lateral canthus. Most appropriately, the incision was placed into a natural supratarsal skin crease. Local anesthetics were infiltrated to the surgical area. The upper-eyelid incision was made in a medial to lateral direction under digital traction and light pressure of the skin while the patient was under general anesthesia. Fifteen milliliters of bilateral orbital inflammatory fat pads were dissected and excised (Fig. 3). The subcutaneous tissue was sutured by using 5.0 Vicryl (Ethicon) and the skin was sutured running subcuticular by using

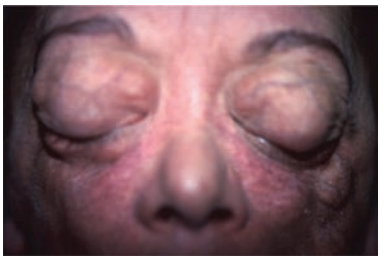

FIGURE 1. Bilateral eyelid ptosis and proptosis on the patient's preoperative picture.

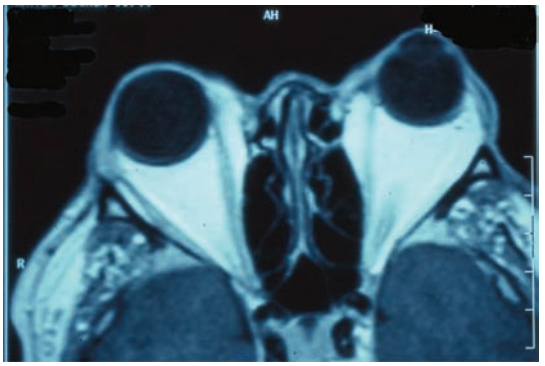

FIGURE 2. Computed tomographic scan demonstrating diffuse thickening of the bilateral upper eyelids with enophthalmos. The bilateral optic nerve and extraocular muscles are grossly normal.

FIGURE 3. The patient's preoperative picture.

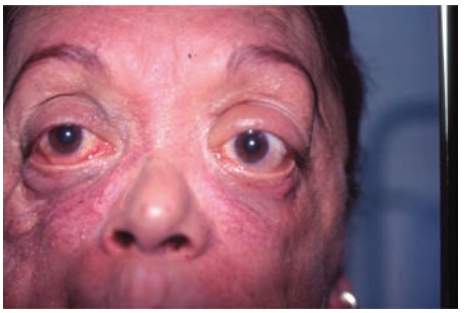

FIGURE 4. The patient's postoperative 2-month picture.

6.0 propilen and closed with light dressing. The sutures were removed on postoperative day 5, and the suture line was intact. The patient was reassessed at 2 months after the surgery (Fig. 4).

Ocular manifestations are annoying problems for patients with WCD. In our study, we have described fat-removal orbital decompressing for severe bilateral proptosis due to WCD.

Orhan Cizmeci, MD
Samet Vasfi Kuvat, MD
Department of Plastic
Reconstructive and Aesthetic Surgery
Istanbul University Faculty of Medicine
Istanbul, Turkey
sametkuvat@yahoo.com

Fatih Ceran, MD
Department of Plastic
Reconstructive Aesthetic Surgery
Bagcilar Training Research Hospital
Istanbul, Turkey

REFERENCES

1. Panush RS, Yonker RA, Dlesk A, et al. Weber-Christian disease. Analysis of 15 cases and review of the literature. *Medicine (Baltimore)* 1985;64:181–191
2. White JW Jr, Winkelmann RK. Weber-Christian panniculitis: a review of 30 cases with this diagnosis. *J Am Acad Dermatol* 1998;39:56–62
3. Klien BA. Nodular non-suppurative panniculitis (Weber-Christian syndrome) with relapsing uveitis. *Am J Ophthalmol* 1959;48:730–734
4. Freedman J. Ocular pathology associated with Weber-Christian syndrome. *Br J Ophthalmol* 1972;56:896–898
5. Frayer WC, Wise RT, Tsaltas TT. Ocular and adnexal changes associated with relapsing febrile non-suppurative panniculitis (Weber-Christian disease). *Trans Am Ophthalmol Soc* 1968;66:233–242
6. Cook JN, Kikkawa DO. Proptosis as the manifesting sign of Weber-Christian disease. *Am J Ophthalmol* 1997;124:125–126

Foreign Body Removal by Inferior Meatal Antrostomy

To the Editor: Removal of foreign bodies in the maxillary sinus, irrespective of the type of material, is the recommended treatment, especially to avoid potential complications such as acute and/or chronic recurrent sinusitis, rhinolith formation, aspergillosis, cutaneous fistula, facial neuralgic pain, and even malignancies that could develop over the long term.[1–4] However, although there is a consensus concerning the need for removing the foreign bodies, there is no international consensus on what is the best surgical approach to use.[4]

A 57-year-old woman presented with a 2-month history of heavy feeling on the right cheek and cacosmia. She had a history of dental treatment 4 months ago. Cone beam CT scans performed in dental clinic revealed the homogeneous soft tissue density in the maxillary and ethmoid sinus of right side and the small-sized metallic density, suggestive of a foreign body on the floor of maxillary sinus (Fig. 1A). After antibiotic therapy with Augmentin (amoxicillin + clavulanate) for about 2 weeks, she visited our hospital for surgical treatment by endoscopic sinus surgery. Because her symptoms were much improved, CT scans were performed to check the interval change of sinus inflammation and to decide the best surgical

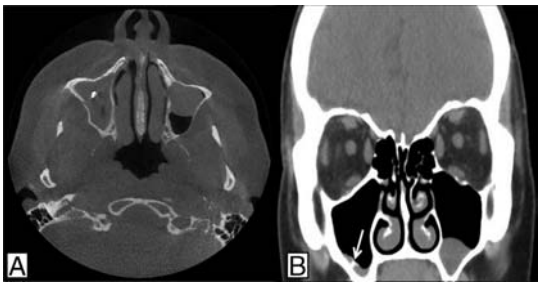

FIGURE 1. A, On cone beam axial CT, the right maxillary sinus was totally filled with soft tissue density and the high-density material suggesting the small size of the foreign body was observed. B, On preoperative PNS CT, the small size of the high-density foreign material (white arrow) was observed in the anteroinferior portion of the maxillary sinus, but the maxillary sinus was relatively clear with normal ostiomeatal complex.

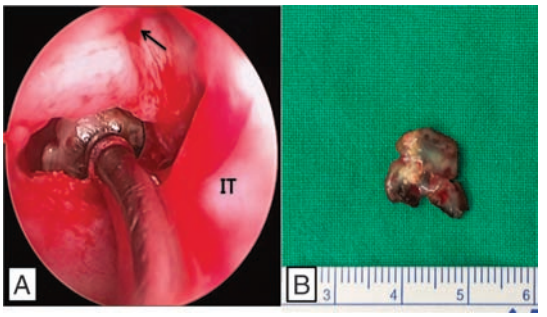

FIGURE 2. A, A curved suction was introduced into the antrostomy and the foreign body was easily removed. The antral mucosa was not markedly thickened, nor was there any bleeding after extraction of the foreign body. B, The foreign body was identified as alloplastic material, bone wax (black arrow, Hasner valve; IT, inferior turbinate).

TABLE 1. Factors Determining the Surgical Approach for Removal of Maxillary Sinus Foreign Bodies

		Indication						
Type	**Approach**	**Paranasal Sinus Involvement**	**Ostiomeatal Unit**	**Size of Foreign Body**	**Location Limitation**	**Advantages**	**Disadvantages**	**Anesthesia**
Intraoral	Classic Caldwell-Luc	Only maxillary sinus	Not affected	Independent	No limitation	Excellent access and visibility, easy removal	Wide bone defect, retraction of the soft tissue of the cheek, paresthesia, and pain; in case of involvement of infraorbital nerve branches, make a sinus lift procedure difficult	Local/ general
	Modified Caldwell-Luc approach							
	Bone flap technique, bone lip approach	Only maxillary sinus	Not affected	Considerable, >1 cm, metallic	No limitation	Avoid scarring retraction of soft tissues including infraorbital nerve branches	High rate of infection due to replacement of the bony window, acted as a non-revascularized bone graft with insufficient blood supply, delay of healing and ossification, total resorption of the bony window	Local
	Bone window technique	Only maxillary sinus	Not affected	Independent	No limitation	Vascularization provided by the sinus mucosa ensures the survival of the bony window (no significant resorption), low ossification of the margins of the bony window, relatively short time (30 min or less), very favorable postoperative recovery, limited pain and swelling	Long-term follow-up may be needed	Local
	Canine fossa puncture	Only maxillary sinus	Not affected	Small size, the largest cross-section: 6 mm	Displaced in the posterior and/or upper part of the maxillary sinus	Limited surgical access, favorable postoperative recovery	Limited control of the surgical field, postoperative facial numbness and pain	Local

TABLE 1. (*Continued*)

Type	Approach	Indication				Advantages	Disadvantages	Anesthesia
		Paranasal Sinus Involvement	Ostiomeatal Unit	Size of Foreign Body	Location Limitation			
Transnasal	Middle meatal antrostomy, functional endoscopic sinus surgery (FESS)	Independent	Affected	Difficult, if the foreign body is large	Difficult whenever foreign bodies are placed in the lower recess of the sinus floor, mainly both the maxillary anterior wall and the lacrimal recess	Higher retrieval rate, minimally invasive, better visualization and access to sinus, lower postoperative morbidity, reduced operation time, earlier discharge, preservation of the mucociliary integrity and function, possibility of treating the affected paranasal sinuses, preserve the integrity of the maxillary sinus walls and the sinus mucosa	Difficult in case of concomitant nasal anatomic variations reducing the nasal space, when the foreign body is large or accompanied by a dental cyst	General or deep sedation

approach. CT scans revealed that the patency of ostiomeatal unit was well maintained without any sinus mucosal lesions, but the metallic density of the foreign body was observed in the right maxillary sinus (Fig. 1B). We determined to remove the foreign body through the inferior meatal antrostomy under local anesthesia. After the inferior turbinate is carefully fractured in the medial and upper direction, a trocar is used to perforate the lateral nasal wall approximately 1 cm posterior to the Hasner valve. This opening is then enlarged with various forceps and a microdebrider. A curved suction is introduced into the antrostomy and the maxillary sinus and a foreign body are easily visualized and removed under a 30-degree rigid endoscope (Fig. 2A). The foreign body is identified as bone wax (Fig. 2B).

There are several important factors in determining the surgical approach for removal of maxillary sinus foreign bodies: the size and location of the foreign body, the functional status of the ostiomeatal complex, and the presence of other paranasal sinus involvement (Table 1).[4–6] After a complete diagnosis through diagnostic endoscopy and imaging studies, the most appropriate surgical approach for every case would be decided. We emphasize through this case that the use of antibiotics before surgery may play an important role in determining the surgical approach.

It is widely recognized that prompt and conservative surgical intervention is desirable to remove the foreign bodies from the maxillary sinus and also to treat acute or chronic maxillary sinusitis. However, because sinusitis due to dental foreign bodies is not an emergency situation, we think it is important to evaluate the responsiveness of antibiotics before determining the surgical approach. After the use of appropriate antibiotics, additional imaging studies are necessary for checking the functional status of ostiomeatal complex and the interval change of sinusitis. However, there is no basic information about the duration of antibiotics use and the drug of choice.

In the present case, we chose the transnasal approach via wide inferior meatal antrostomy because the size of the foreign body is small and both the ostiomeatal complex and maxillary sinus are clear after 2 weeks of antibiotic medication. We think the inferior meatal antrostomy has 2 advantages. First, although it has been shown that inferior meatal antrostomy is unfavorable because the mucociliary drainage of the sinus mucosa continues to target the middle meatus, normal in vivo mucociliary clearance has been demonstrated in patients after inferior meatal antrostomy.[7] Second, the sufficient antrostomy window with various angled endoscopes is helpful for removing the foreign bodies easily, even if they are present in any location, especially the floor and anterior wall of the maxillary sinus.[8]

We emphasize through this case that the use of antibiotics before surgery may play an important role in determining the surgical approach. Also, we suggest that the endoscopic approach from the inferior meatal window may be an ideal method for extracting the foreign body of the maxillary sinus.

Kyung Soo Kim, MD, PhD
Department of Otorhinolaryngology–Head and Neck Surgery
Chung-Ang University College of Medicine
224-1, Heukseok-dong, Dongjak-gu
Seoul, Korea
99-21045@hanmail.net

REFERENCES

1. Birnmeyer G. On late sequelae of metallic foreign bodies in the region of the paranasal sinuses. *J Laryngol Rhinol Otol* 1963;42:778–785
2. Brinson GM, Senior BA, Yarbrough WG. Endoscopic management of retained airgun projectiles in the paranasal sinuses. *Otolaryngol Head Neck Surg* 2004;130:25–30
3. Scolozzi P, Martinez A, Lombardi T, et al. Lateral antrotomy as a surgical approach for maxillary sinus: a modified technique with free bone flap repositioning and fixation with a titanium plate. *J Oral Maxillofac Surg* 2009;67:689–692
4. Scolozzi P, Momjian A, Lombardi T. Removal of unusual, large high-velocity metallic maxillary sinus foreign bodies by a modified free bone flap technique. *Eur Arch Otorhinolaryngol* 2010;267:317–320
5. Biglioli F, Chiapasco M. An easy access to retrieve dental implants displaced into the maxillary sinus: the bone window technique. *Clin Oral Impl Res* 2014;25:1344–1351
6. González-García A, González-García J, Diniz-Freitas M, et al. Accidental displacement and migration of endosseous implants into adjacent craniofacial structures: a review and update. *Med Oral Patol Oral Cir Bucal* 2012;17:e769–e774
7. Yanagisawa E, Joe J. Inferior meatal antrostomy: is it still indicated? *Ear Nose Throat Journal* 1997;76:368–370
8. Moeller CW, Stankiewicz JA. Endoscopic inferior meatal antrostomy. *Operat Tech Otolaryngol Head Neck Surg* 2010;21: 156–159