Removal of the Mandibular Lesion With Immediate Reconstruction Using Bone Graft

To the Editor: Reconstructing defects after jaw resections presents a challenge for the reconstructive surgeon because of the critical role played by the facial skeleton in function and esthetics.[1] Currently, there are many etiologies that cause loss continuity of maxillary bone as the need for removal of tumors, the effects of gunshot wounds, or the presence of severe infections.[2]

Aggressive odontogenic tumors including mainly ameloblastoma and odontogenic myxoma, although there are other rare lesions that could be classified as such,[3] may require resection because of its expansive and infiltrative behavior. Ameloblastomas are rare epithelial odontogenic tumors that grow slowly but are locally invasive and can be highly destructive of the surrounding dental anatomy.[4,5] This lesion appears more frequently in the mandible (80% of all cases),[6] particularly in the angle and ramus, although it can occur in any mandibular region.[7]

Treatment modalities indicated for ameloblastomas vary from enucleation to resections, which can be followed or not by reconstruction, depending on the histopathological classification. Radiotherapy is not indicated because the lesion is radioresistant.[5] In the scientific environment, some authors indicate cryosurgery,[4] eletrocauterization, and the use of sclerosing agents as treatment options.[8]

The reconstruction approach of the ameloblastomas can be done through free vascularized bone grafts or free bone grafts, with satisfactory results.[6] The authors share the case of a male young patient who presented a large radiolucent lesion of the mandibular body that was resected and immediately reconstructed with free mandible graft harvested from the iliac bone.

CLINICAL REPORT

A 23-year-old male patient was referred to the Oral and Maxillofacial Surgery Department, Presidente Dutra University Hospital for evaluation of a tumor lesion in the mandible. The patient wanted to have his occlusion corrected by orthodontic treatment; the practitioner viewed the lesion in the right mandibular body and immediately referred the patient to our oral and maxillofacial surgery service.

On extraoral clinical examination, we observed no increase in volume in the region compared with the contralateral side, and the patient had good buccal opening; however, there was premature contacts in the posterior region of the right side. The panoramic radiograph revealed a radiolucent image region showing the body of the mandible on the right side with the 48 teeth included in the lesion, with resorption process already begun in the roots of teeth 46 and 47 (Fig. 1A). Computed tomography was requested, and the coronal and axial cuts showed expanding lesion with involvement of the lingual cortex of the mandible (Fig. 1B).

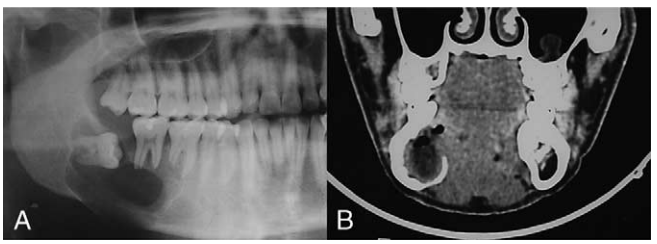

FIGURE 1. A, Panoramic radiograph showing the lesion in the region of the right mandibular angle with associated third molar, resorbing the roots the teeth 46 and 47. B, Coronal CT scan showing the involvement of the lingual cortex.

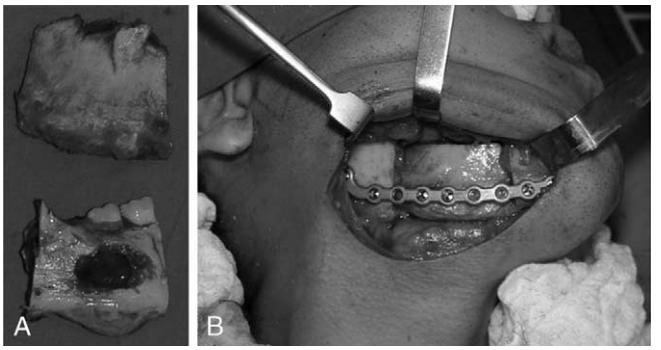

FIGURE 2. A, Graft bone compared to pathological block removed, similar in size. B, Bone graft positioned and fixed to the reconstruction plate system 2.4.

After discussion between team members of the oral and maxillofacial surgery service and the patient on the diagnostic hypotheses and types of treatment, lesion resection and immediate reconstruction using a bone graft were proposed.

The surgery was performed under general anesthesia with tracheal intubation. The patient was placed in maxillomandibular lock through Erich bars to restore the tooth contacts in the postoperative stage similar to the preoperative stage. Submandibular or Risdon incision was made, and after dissection by planes, the lesion was identified and delimited with a safety margin of 3 mm, both anterior and subsequent to the injury. Pathological bone block was removed using a reciprocating saw under copious irrigation with 0.9% saline. At the same time, the orthopedic team removed the free bone graft from the anterior iliac bone on the right side, using chisels and hammers, almost similar in size and thickness of the mandibular block removed (Fig. 2A), optimizing the transoperative time and making it an easier reconstruction. The graft received minor adjustment modeling and was positioned into the defect and fixed with a reconstruction plate system 2.4 (Fig. 2B), which had been prefolded, since the preoperative mandibular contour presented aspects of normality. Suture was performed in layers using polyglactin 910, and the skin was closed with 6-0 nylon suture. The bone block with the lesion was sent for histopathological analysis, which confirmed the result of ameloblastoma.

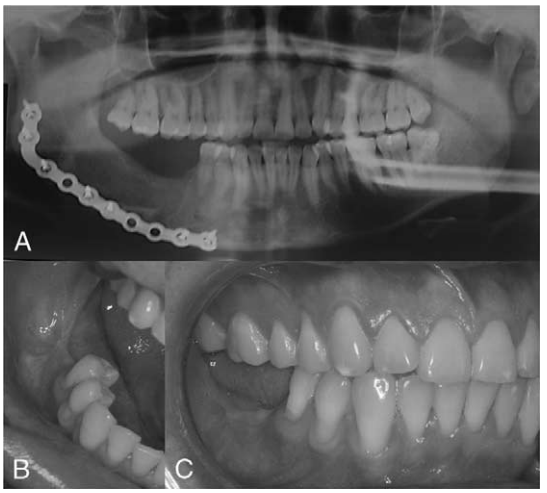

FIGURE 3. A, Panoramic radiograph 1 year postoperatively, showing good consolidation and adaptation of the bone graft. B, Intraoral clinical aspect with excellent tissue healing. C, Intrabuccal aspect of occlusion of the right side.

After a year of follow-up, full adaptation and bone graft union with the bone tissue of the recipient site were noted, with excellent esthetic appearance of the extraoral scar and good intraoral great, and maintenance of preoperative occlusion (Fig. 3). The patient already planned the installation of dental implants for further rehabilitation with prosthesis on implants in the grafted area; however, the patient did not return to the clinic.

DISCUSSION

The mandibular reconstruction after resection of tumors is extremely important in improving quality of life[6] mainly because the cosmetics and functional defects resulting from this loss of tissue will vary according to both the site and the extent of the deficiency.[2]

Ameloblastoma appears more frequently in the mandible (80% of all cases), particularly in the angle and ramus, although it can occur in any mandibular region.[7,8] It is associated with tooth resorption and severe expansion of the cortical bone and may perforate the lingual and buccal plates as what occurred with the clinical presentation of this case. The bone overlying bone, or bone plus soft tissue, infiltrates the soft tissue,[3,5,8] so the forms of treatment can vary greatly; and there are still many controversies.

In this case report, the authors opted for resection of benign tumor with immediate reconstruction by taking into consideration the size that has not yet had facial impact. This form of treatment is permanent with a low rate of recurrence, and the patient is young and with good medical condition; a recurrence can only be attributable to a combination of inadequate surgical treatment and the aggressiveness of the tumor.[9] Marginal resection is the most common treatment approach; however, we have seen reports of 15% recurrences,[8] and this present case had already shown involvement of the mandibular base.

A rate of 20% to 30% of the free nonunion bone graft recipient site has been reported, causing many subsequent surgical procedures to achieve satisfactory results.[4] Fortunately, in this case report, there was healing and adaptation of bone graft amenable to installation of dental implants and subsequent rehabilitation with dentures, restoring esthetics and chewing function.

The iliac graft is the only free flap that enables reconstruction of the initial width and height of the mandible, immediate placement of implants in a position that does not interfere with osteosynthesis material, and, therefore, appropriate rehabilitation with a dental prosthesis.[10] However, in this case, the patient did not receive dental implants because he did not return to the clinic. Most of our service is maxillofacial surgery.

Resection of benign tumor lesions presenting with moderate sizes involving the full thickness of the jaw followed by immediate free reconstruction of the anterior iliac bone grafting is a viable option, nonmutilating, and allows functional restoration to patients.

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Correction of Severe Microstomia Secondary to Gunshot by Using Free Osteocutaneous Radial Forearm Flap

To the Editor: Microstomia is a clinical feature used to describe congenital or acquired reduction in the size of oral aperture. Genetic disorders, burns, traumas, and contractures as a result of perioral surgery are main causes of microstomia. Many procedures have been described for surgical correction of microstomia in literature such as fishtail flap,[1] nasolabial flap,[2] and variations of mucosal flaps and rhomboid flap.[3–5] Using free osteocutaneous radial forearm (FOCRF) flap for facial defects has been described previously.[6]

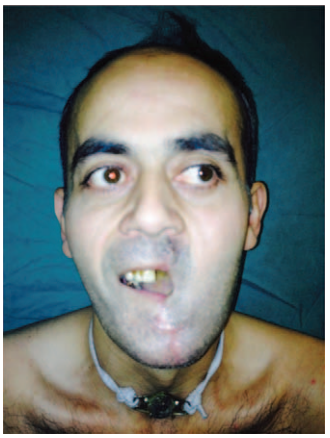

FIGURE 1. Patient's preoperative photograph (frontal view).

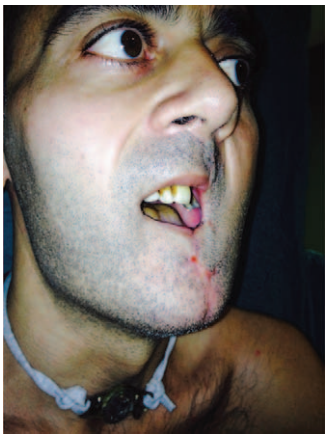

FIGURE 2. Patient's preoperative photograph (oblique view).

A 37-year-old man with severe microstomia secondary to gunshot was admitted to our clinic (Figs. 1 and 2). Bone defect was detected in mandibular parasymphyseal region by using computed tomography (Fig. 3.).

Contractures and scars on the lower lip were excised while the patient was under general anesthesia. Bone fragments in mandibular symphyseal region were removed. An FOCRF flap was elevated on the left forearm. Flap was adapted to mandibular symphyseal region and lower lip. There were no problems on postoperative follow-up. The flap was found to be perfectly adapted on postoperative 3 months (Figs. 4 and 5).

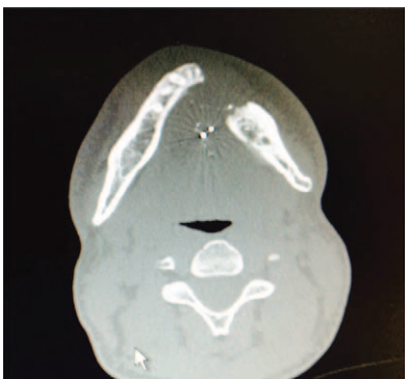

FIGURE 3. Bone defect in mandibular parasymphyseal region.

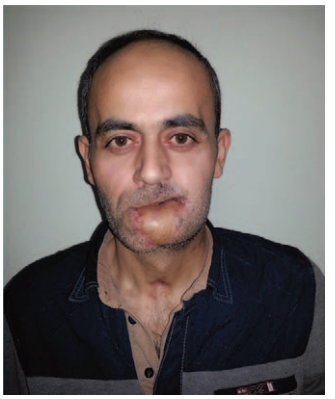

FIGURE 4. Patient's postoperative 3 months' photograph.

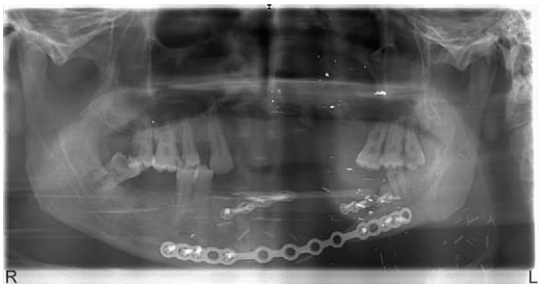

FIGURE 5. Patient's postoperative 3 months' panoramic radiograph.

The FOCRF flap has advantages in maxillofacial reconstruction because of its thin and flexible skin texture and its ease of application to the oral region.[6] The FOCRF flap must be kept in mind as an option for correction of severe microstomia within bone defect.

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How Can We Avoid Distortion in Facial Photographs Using Compact Cameras?

To the Editor: Preoperative and postoperative photographs have enormous importance in plastic surgery both for planning and evaluation of surgical results.[1] For achieving optimal results, the photographs should reflect the object in its true dimensions. This problem is especially seen in facial photographs.[2] Facial photographs which were taken from improper distance with inappropriate objectives may cause distortions such as blunt looking the midface and narrowed upper and lower facial borders.[3]

Digital cameras can mainly be evaluated in 2 major categories: single-lens reflex (SLR) and compact.[4] In SLR cameras, it is possible to see the focus distance during photographing which is not possible in compact cameras.[1] In compact cameras, the focus distance increases with zooming in and decreases with zooming out, which means that if the frame stays constant, the focus distance changes in relation with the distance changes occurring between the object and the camera.[1,4] This problem is not encountered in SLR cameras and a focus distance of 100 mm is considered as standard in facial photographs.[5]

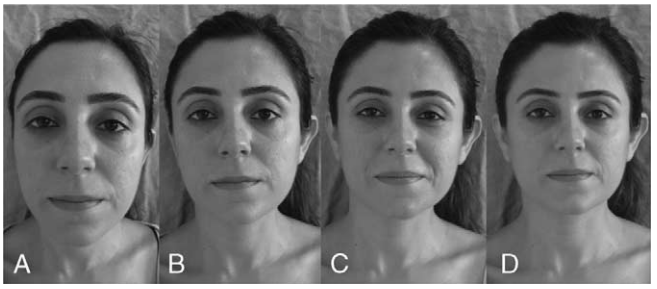

FIGURE 1. The facial marking used for the photo anthropometric evaluation showed on a picture taken from 100 cm distance. The points were marked with a distance of 4 cm in horizontal plane (A-A′ = B-B′ = C-C′ = 4 cm).

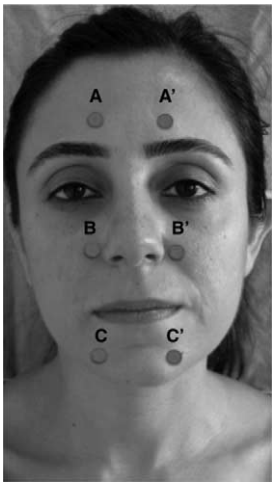

FIGURE 2. The facial photograph of the same patient taken with Canon G12 with different distances: A, 20 cm; B, 70 cm; C, 100 cm; D, 150 cm. Note the obvious distortion obtained in the photograph taken from 20 cm distance.

However, it is not always possible to use a SLR camera and compact cameras are routinely used in many centers and by many plastic surgeons due to their smaller size, easy usage, and portability. For this issue, in this study, the optimal distance preventing distortion by facial photographs when using compact camera is investigated.

Three different cameras were used for the study: Canon G12 (Canon Inc., Tokyo, Japan), Canon PowerShot SX280 HS (Canon Inc.), and Samsung S760 (Samsung Electronics, Seoul, South Korea). Facial markings located at the superior one third (A-A′), middle one third (B-B′), and inferior one third (C-C′) were performed by the patients with equal distances (Fig. 1). After that, facial photographs were taken by creating a distance between the camera and the patient 20 cm, 70 cm, 100 cm, and 150 cm, respectively, with these 3 different compact cameras. The borders of the facial photographs were designed so that the lateral borders were cut next to the ears, the superior borders were cut just above hair, and the inferior border was the clavicle. The distances between the facial markings (A-A′, B-B′, C-C′) were measured using photo anthropometric techniques (pixel counting) with the help of the measure tool of Photoshop Software (Adobe Systems, San Francisco, CA). After measuring pixel counts between the points, the A-A′ distance was chosen as the constant value and all pixel measurement results were divided to A-A′ distance to determine any inconsistency between them. The distance in which all results of the divisions (A-A′/A-A′, B-B′/A-A′, and C-C′/A-A′) are equal to 1.0 is considered as the distance without any distortion.

Table 1 demonstrates the measurement results showing that the worst results were obtained in 20 cm and increased distances

TABLE 1. Photo Anthropometric Measurement Results Obtained by Using the Photoshop Software of the Photographs Taken from 20 cm, 70 cm, 100 cm, and 150 cm Distances Using Each Camera

	20 cm	70 cm	100 cm	150 cm
	x y z	*x y z*	*x y z*	*x y z*
Canon G12	1.0 0.9748 0.9519	1.0 0.9897 0.9849	1.0 1.0 0.9933	1.0 1.0 1.0
Samsung S760	1.0 0.9827 0.9625	1.0 0.9985 0.9915	1.0 1.0 0.9973	1.0 1.0 1.0
Canon PowerShot SX280 HS	1.0 0.9922 0.9847	1.0 0.9913 0.9870	1.0 1.0 0.9927	1.0 1.0 1.0

x indicates the division of A-A′ to A-A′; *y*, the division of B-B′ to A-A′; *z*, the division of C-C′ to A-A′.

provide better results until reaching 150 cm. In 150 cm distance, all pixel-counting measurements achieved by performing the aforementioned divisions are equal resulting as 1.0 which means that the face is displayed in the photograph with its true proportions without any distortion (Table 1 and Fig. 2).

In conclusion, facial photographs should be taken from a distance of 150 cm (1.5 m) by zooming in until the patient's face and neck to the level of the clavicle fits exactly into the frame to achieve natural facial photographs with minimal distortion when using compact cameras.

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Clinical Profile of a Patient With Cleft Lip/Palate With Secondary Skeletal Deformities

To the Editor: All children with clefts who undergo lip and palate repair in infancy and childhood develop midface hypoplasia. It is well accepted that it is only the degree that varies depending on various surgical and patient factors.[1] Although primary lip and palate repair has become available to most of the estimated 27,000 to 33,000 children born with orofacial clefts in India every year,[2] multidisciplinary follow-up and management has not kept pace with this, with most of the centers offering little more than immediate postoperative follow-up. Although there have been efforts by nonprofit organizations as well as government health services to correct this over the last decade,[3,4] it will be evident from the following narrative that several children with cleft lip/palate do not have access to comprehensive follow-up and appropriate interventions from a multidisciplinary team.[5] Most of these children grow up with unaddressed alveolar clefts and untreated surgical complications. In some patients, the problem is further complicated by repeated well-intended but ill-advised interventions and possibly suboptimal surgical techniques, causing excessive fibrosis, which in turn increases the severity of the skeletal midface deformity.[6]

PATIENTS AND METHODS

One hundred consecutive patients with cleft lip/palate who underwent midface correction between the years 2007 and 2013 at a multidisciplinary cleft and craniofacial center in south India were studied. The dates were adjusted so that the cohort studied comprised of exactly 100 patients who underwent skeletal correction for midface hypoplasia, which included orthognathic surgery and distraction osteogenesis. The retrospective study was conducted by reviewing case records and examining photographs, radiographs, and dental models. Only patients who had complete records and were available for review were included. All the patients had skeletal deformities of the maxilla in addition to one or more structural and functional problems including deformities of the lip and nose; unaddressed alveolar clefts; collapsed alveolar arches; contracted palates; crowding, missing, retained, or supernumerary teeth; oronasal fistulae; broken-down palates; and speech disorders. We analyzed the following parameters with which the patients presented to us, viz: age at first presentation, reasons for seeking treatment, distance traveled, number of previous operations, and skeletal and soft tissue deformities at presentation. The treatments the patients underwent at our center including surgical, orthodontic, and speech interventions are enumerated. We have briefly discussed our management protocols and outcomes including prioritization of treatment in dealing with multiple problems and the expedients we adopted to keep travel and costs for the patients and their parents to a minimum. Our decision-making process to deal with complex skeletal midface deformities (Fig. 1) is presented in the form of an algorithm. In addition, we have included case reports of a few typical patients and briefly traced the course of their rehabilitation.

FINDINGS AND DISCUSSION

Age at Presentation and Reasons for Seeking Treatment

Forty-nine male and 51 female patients in the study group reported for treatment for multiple secondary deformities including cleft midface hypoplasia at ages ranging from 11 to 38 years, with the mean age of presentation being 19.41 ± 5.09 (Fig. 2). Of

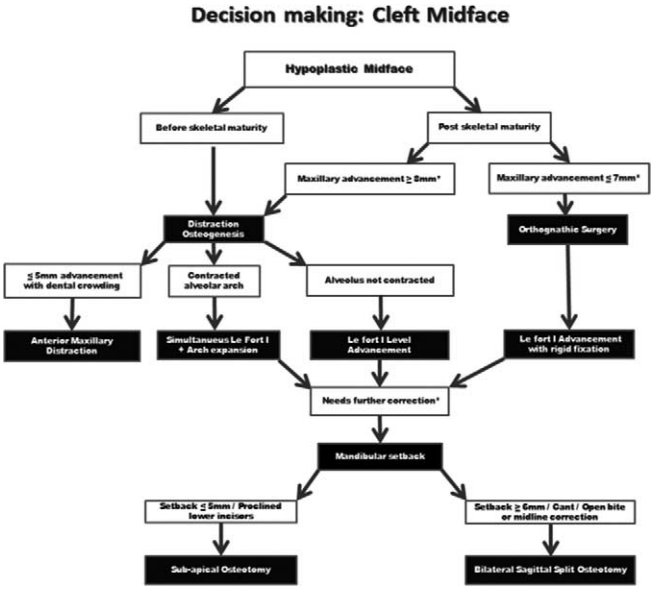

FIGURE 1. Algorithm for decision-making process for management of skeletal midface deformity.

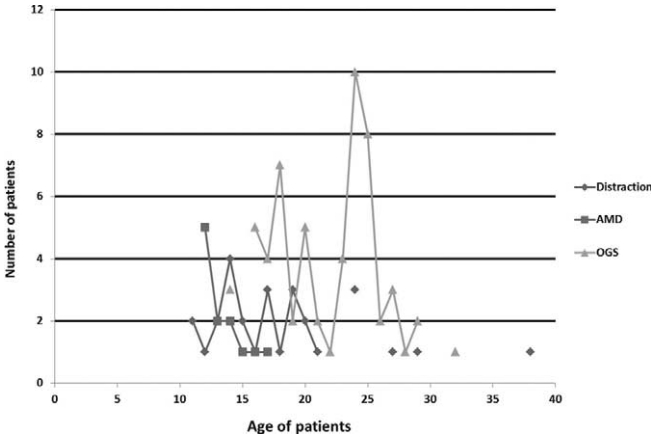

FIGURE 2. Age of surgery for midfacial skeletal deformity correction and procedure performed.

these, only 16 were either referred specifically for midface correction or were seeking treatment for the skeletal deformity of their face. All others were seeking general improvement in form and function. The reasons for coming to the hospital included appearance of the lip and nose, poor speech, palatal fistula, broken-down palate, alveolar gap, dental problems, etc. Although many were aware of the skeletal deformity, they were not aware that their midface skeleton needed correction or that this was possible by surgery till it was explained to them. Interestingly, no patient complained of problems with mastication,[7] whereas nasal regurgitation was a common complaint. This may be attributed to the soft rice-based diet of the predominantly South Indian population. When the ages at first presentation were plotted against the reasons for seeking treatment in the form of a graph (Fig. 3), the following findings were arrived at. The peak in the graph around the age of 12 years was driven by the need for improvement in speech. Most patients or parents in this group admitted to being pressured by school teachers to improve communication. The subsequent peaks around 19 and 23 years of age were driven by requests for improvement in form and function, which may be due to increased self-awareness during the teens and parental concerns about prospects of marriage of their wards.

Distance Traveled

The mean distance the patient traveled to reach the hospital was 279.16 km, with distances ranging from 5 to 1800 km (Fig. 4).

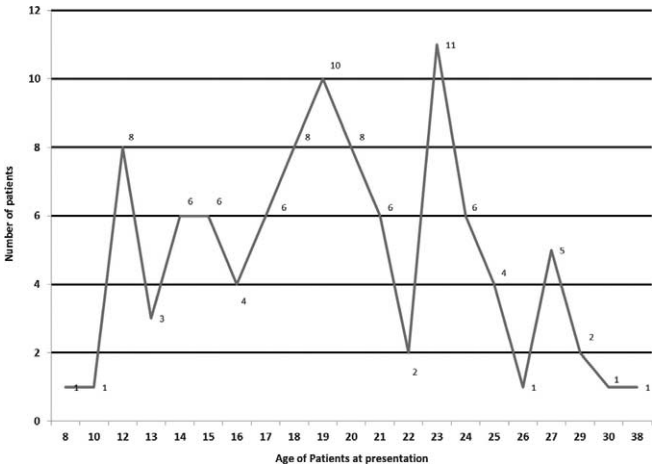

FIGURE 3. Age at first presentation.

FIGURE 4. Representative map showing the distances traveled by patients to reach our multidisciplinary center. (Courtesy: Google Maps).

Although there are more than 200 dedicated cleft centers offering free primary cleft lip and palate operations, there are only a few that offer comprehensive multidisciplinary management of skeletal, soft tissue, orthodontic, and speech problems within the reach of most patients.[5]

Previous Treatment

Of the 100 patients studied, 95 had primary operation elsewhere and reported for secondary correction to our center for the first time. Of these, 56 patients had only primary operations and no follow-up before they came to us. Twenty-eight patients had alveolar bone grafting (ABG), and the rest had undergone one or more additional operations for correction of secondary or residual deformity, for example, attempted fistula closure, palate re-repair, etc. The number of operations each patient had before reporting to us ranged between 1 and 7, with a mean of 2.34 ± 1.14.

Treatment at our Center

All patients underwent evaluation by the multidisciplinary team and a treatment plan was formulated for complete rehabilitation of the patient. Most patients needed orthodontic treatment, speech therapy, and multiple soft tissue operations and ABG in addition to skeletal correction. The operations included repair of oronasal fistulae and broken-down palates, correction of velo-pharyngeal insufficiency (VPI), ABG, and lip nose revision in addition to skeletal correction (Fig. 5). Prioritization of treatment was made, keeping in mind the following guidelines. Skeletal correction always preceded lip-nose revision and VPI correction in all adults

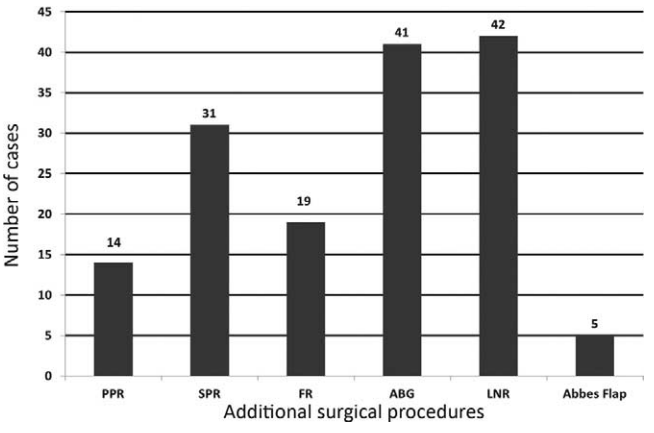

FIGURE 5. Bar graph showing number of additional surgical procedures in addition to skeletal corrective procedures.

and in some children with severe midface hypoplasia necessitating early intervention. Children with mild skeletal midface hypoplasia that did not affect their lifestyle were advised to wait till skeletal maturity for skeletal correction, and priority was given to VPI. Alveolar bone grafting or gingivoperiosteoplasty was done before skeletal correction in all cases of orthognathic surgery. Exceptions were made in cases with severe collapse of the alveolar arch undergoing Le Fort I level distractions, where expansion of the arch was done simultaneously with anteroposterior distraction. In bilateral cases, ABG was done at least on one side before proceeding to distraction. All operations were separated by 6 months, and the interim period was used for orthodontia and speech therapy. This used the interim period effectively and improved compliance. The number of operations each patient underwent at our center, in addition to midface advancement, ranged from 1 to 8, with a mean of 2.92 ± 1.58. Literature search for similar published data from centers other than ours did not yield any results.

Skeletal Correction

Of the 100 patients studied, 60 underwent orthognathic surgery, 28 patients had Le Fort I level osteotomy and distraction osteogenesis, and 12 had anterior maxillary distraction (AMD) (Fig. 6). The indications and the decision-making process for these procedures are given in the form of an algorithm.

Orthognathic Surgery

Of the 60 patients in this group, 31 patients underwent isolated Le Fort I osteotomy, 13 had Le Fort 1 osteotomy with bilateral sagittal split osteotomy (BSSO), 8 patients had Le Fort I with subapical osteotomy, 3 patients had Le Fort I osteotomy with genioplasty, 3 patients had anterior maxillary osteotomy (AMO), and 2 patients had isolated BSSO (Fig. 6). The advancement of Le Fort I segment ranged from 3 to 10 mm, with a mean of 6.14 ± 2.41 mm. All patients had severe fibrosis and scarring, making Le Fort I level advancement during orthognathic surgery challenging when compared to patients without cleft. Seven patients had pharyngeal flaps of which 4 patients were partially or completely divided to facilitate advancement. The effect of midface advancement on speech is discussed later.

In four of the patients planned for orthognathic surgery, the planned advancement of the Le Fort I segment could not be achieved because of fibrosis or the presence of previous Malek pharyngoplasty.[8] In 2 patients, this was managed by fitting an external distractor temporarily to achieve advancement followed by plate and screw fixation after a week. In the other 2 patients, the plan was changed to distraction osteogenesis.

Distraction Osteogenesis

The patients in the distraction group had either low Le Fort level osteotomies with external frame distractor or AMD. In the Le Fort I group, traction was effected by a transnasal wire in all cases except 2 patients who had tooth-borne distraction device and two of the early cases who had Leipzig plates. The AMD was effected by indigenously fabricated tooth-borne intraoral device. The maxillary advancement ranged from 5 to 18 mm in the Le Fort I group and 5 to 6 mm in the AMD group. All patients who underwent distraction osteogenesis were put on a rigorous protocol of monitored distraction, which was started 5 days after osteotomy with a 3-month consolidation period. This was followed by a further 9-month period of retention with removable devices in addition to 20% to 30% overcorrection to allow for the inevitable relapse.[9,10]

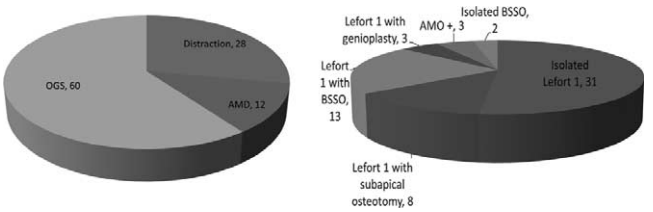

FIGURE 6. Number and various types of skeletal procedures performed.

Speech (n = 50)

The deterioration in speech acceptability was seen in 11 of 50 patients with no significant difference between distraction and orthognathic surgery. The patients whose velopharyngeal function deteriorated either had large advancements (>10 mm) by distraction osteogenesis or had their pharyngeal flap divided during orthognathic surgery. There was no significant change in speech parameters or nasoendoscopic findings preoperatively and postoperatively in patients who had a pharyngeal flap when it was not divided.[11]

Expedients to Improve Patient Compliance Minimize Down Time and Cost

In our situation, even when the treatment is subsidized or rendered completely free, the cost of time, effort, and money for patients and parents due to time away from work, travel, etc, can still be prohibitive. Hence, the possibility of demotivation and exhaustion leading to dropping out of treatment is high. One needs to plan the treatment to keep hospital visits and in-patient stay to a minimum while judiciously prioritizing treatment to get demonstrable results early in the course of treatment to keep the morale high.

Timing of Orthodontic Treatment and Speech Therapy

Since most patients needed multiple operations, these were spaced 6 months apart, and the interim period was used for speech and orthodontia.

Orthodontia

In patients treated by surgery as the first method,[12] the mean time between beginning of orthodontic preparation and surgery was approximately 3 weeks and the postoperative orthodontia lasted for a mean period of 50 weeks. In suitable cases, orthodontic manipulations were aided by the osteotomies already made.[13] When this was compared to the traditional orthodontic/orthognathic methods, there was a considerable saving in time with lesser chances of patient demotivation.

Simultaneous Transverse and Anteroposterior Distraction Osteogenesis

Selected patients had simultaneous palatal expansion and midface advancement,[14] thereby telescoping the time required for both, with the added advantage of even expansion of the palate.[15]

Prosthodontic Rehabilitation

Three bilateral patients with cleft lip/palate with missing premaxillae and one unilateral patient with cleft lip/palate with missing anterior teeth were rehabilitated with advancement of the lateral segments and fixed dental prosthesis. Patients who underwent anterior maxillary distraction osteogenesis needed osteointegrated implants as spacers to prevent relapse.

FIGURE 7. Ms. N, 24 years old at first presentation, complete unilateral cleft of lip and palate. Previous operations: 2. Problems: poor lip and nose, anterior palatal fistula, broken-down soft palate with scarring and tissue loss, VPI, poor speech intelligibility (5/7), midface retrusion, collapsed alveolar arch, malocclusion, and malaligned and missing teeth. Management: palate re-repair with Malek pharyngeal flap, anterior fistula repair with tongue flap, Le Fort I distraction of 12 mm, lip-nose revision, speech therapy, orthodontia, and prosthetics. Summary: 6 operations over 4 years. Speech improved from 5/7 in 2004 to 2/7 in 2009.

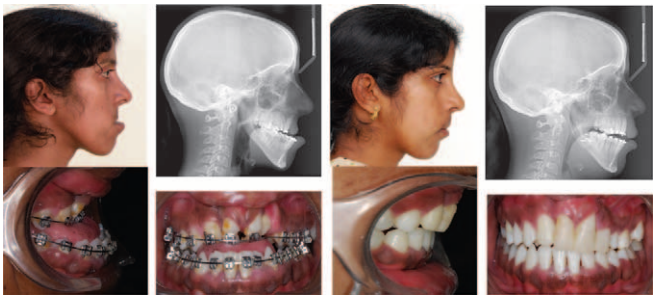

FIGURE 8. Ms. S, 22 years old at first presentation, complete bilateral cleft of lip and palate. Previous operations: 6. Problems: class III malocclusion, absolute prognathism, broken-down palate, VPI, bilateral alveolar cleft, and anterior fistula with a failed tongue flap. Management: 20-mm correction with Le Fort I distraction and BSSO, palate re-repair with Malek pharyngoplasty, anterior palate repair with remnants of the tongue flap, ABG on one side and gingivoperiosteoplasty on the opposite side. Summary: 4 operations over 2 years. Logistics problem for orthodontia overcome by surgery first method. Full closure of velopharyngeal port on endoscopy. Difficulty with speech therapy because of logistics.

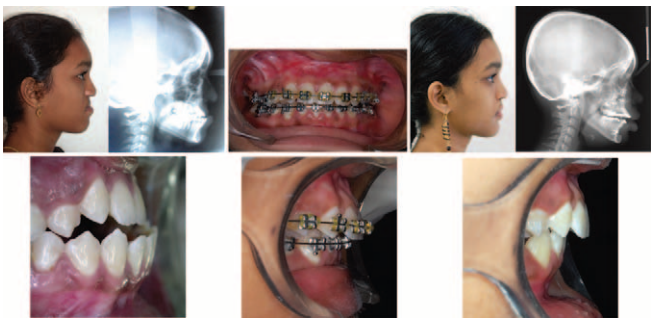

FIGURE 9. Ms. N, 8 years old at first presentation, complete unilateral cleft of lip and palate. Previous operations: 2. Problems: poor lip and nose, anterior palatal fistula, poor speech intelligibility (4/7) with VPI. Developed concave profile with class III malocclusion, reverse overjet with anterior open bite in the teen years. Management: palate re-repair with fistula closure and Malek pharyngoplasty at 8 years, ABG at 9 years, lip nose revision at 9 years, orthognathic surgery at 18 years (maxilla: Le Fort I advancement by 6 mm, cant correction by impaction on left side by 3 mm; mandible: subapical osteotomy with setback to the width of first premolar), orthodontia, and speech therapy. Summary: 5 operations over 10 years. Speech intelligibility of 2/7 at the end of treatment.

COMPLICATIONS

Distraction Osteogenesis

Minor complications are common and need to be identified and corrected as they occur.[9] Of the 40 patients who underwent distraction osteogenesis, failure of distraction needing reosteotomy was seen in 2 patients, loss of tooth anchorage was seen in another 2 patients, breakage of steel wire was seen in one patient, and loosening of the halo frame distractor was seen in 2 patients. One patient had asymmetric distraction. Perioperative bleeding necessitating interventional radiological embolization of a branch of the ascending pharyngeal artery was seen in one patient. Significant relapse necessitating further procedures was seen in 4 patients. Two of these patients had defaulted during the postdistraction phase and came back to us with severe relapse. All of them had been operated on for severe retrusion in their early teens and had skeletal growth left in them. To date, one each could be managed with repeat maxillary distraction and BSSO and BSSO alone, respectively.

Orthognathic Surgery

Of the 60 patients who underwent orthognathic surgery, gingival regression was seen in 3 patients after Le Fort I osteotomy. Significantly, more morbidity was seen with mandibular surgery. Four patients who had BSSO had anterior migration of mandible in the immediate postoperative period. However, all of them responded to postoperative elastics and settled in 4–6 weeks. All patients had hypoesthesia or anesthesia of the lower lip and chin in the postoperative period. However, only 2 patients continued to have hypoesthesia beyond 6 months. However, none of the patients had a demonstrable division of the inferior alveolar nerve during surgery. None had a complete unfavorable split (Figs. 7–9).

CONCLUSIONS

Patients with cleft lip and palate in India as in many parts of the less developed world do not always have access to multidisciplinary care and follow-up after primary lip and palate repair. Hence, it is common for them to seek treatment in their teens or later for a hypoplastic midface with multiple secondary cleft deformities and unaddressed complications of primary surgeries. These patients need a comprehensive protocol for management of their multiple soft tissue and functional problems in addition to skeletal midface correction. Since the rehabilitation process is prolonged, it needs to be done by judiciously combining procedures, keeping hospital visits to a minimum and costs low to minimize patient dropout due to demotivation or exhaustion. In the long term, there is a need for more multidisciplinary units that can provide comprehensive follow-up and treatment after primary surgery so that secondary problems and complications when they occur are addressed early at the appropriate age.

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Infantile Myofibromatosis of the Soft Palate

To the Editor: A 6-year-old white girl came to our attention in 2004; her parents referred a 1-year history of breathing difficulties, especially during sleeping, with recurrent rhinopharyngitis. Clinical examination revealed an intraoral painless mass deforming posterior soft palate and pushing it down; no mucosal ulcerations were detected (Fig. 1A). Contrast-enhanced computed tomographic (CT) scan performed at another center showed a solid lesion associated to homogeneous and intense contrast enhancement involving soft palate (Fig. 1B). No lymphadenopathy was detected, and a vascular malformation was suspected. Digital subtraction angiography excluded this hypothesis.

On December 2004, when she came to our attention, a magnetic resonance (MR) examination with gadolinium was performed. It confirmed an expansive mass involving soft palate with rhinopharyngeal subtotal obstruction. The pathological tissue was characterized by mild hyperintense signal on T1-weighted images compared to tongue signal (Figs. 1D). An inhomogeneous hyperintensity on T2-weighted, ''salt and pepper'' like, (Figs. 1C-F), and homogeneous enhancement after contrast injection was evident (Figs. 1E-G); the mass lesion appeared well distinguishable with respect to the surrounding anatomical structures. Because of MR findings and the increasing clinical manifestation, a surgical resection was performed without a previous biopsy because of presumed benignity of the lesion documented in the MR image, and to avoid risk of hemorrhage. Transoral excision of the mass was performed under general anesthesia. Intraoperative pathological examination revealed the presence of a benign mesenchymal tumor. The postoperative course was uneventful. Definitive histopathological examination confirmed the presence of a $2.6 \times 2.2 \times 2.1$ infantile myofibroma. Microscopically, it was composed of fusiform cells with eosinophilic and leptochromatic cytoplasms positioned in bundled fibrous tissue. Mitotic index was less than 1×10 high. Immunohistochemical stain showed positive reaction to smooth muscle actin and desmin but was negative for S-100 protein. At 8 years' follow-up, no local relapse can be observed.

Infantile myofibromatosis is a benign condition first described by Stout in 1954,[1] most frequently affecting soft tissues of the head and neck with a predilection for males between neonatal and infantile period.[2,3] It can manifest as solitary lesion or in a multicentric form; in most cases, it is a self-limiting disease, without spreading out, but sometimes it may have an aggressive behavior.

In 1981, Chung proposed a distinction of 3 clinical forms: solitary, multicentric, and multicentric with visceral involvement.

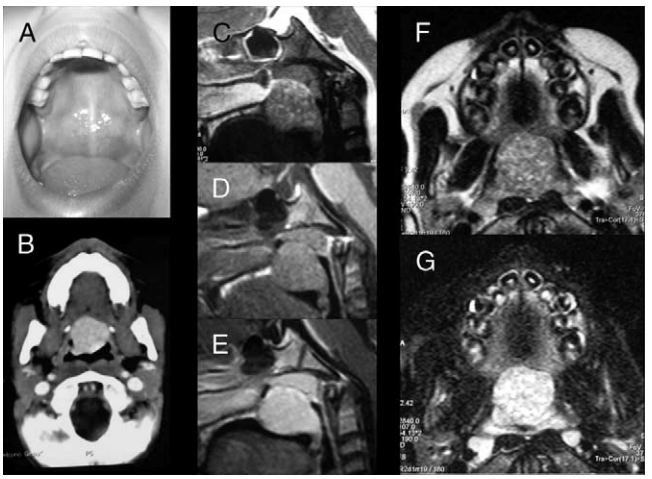

FIGURE 1. Preoperative findings. A, An intraoral mass deforming posterior soft palate without mucosal ulcerations. B, Contrast-enhanced CT scan showed a solid lesion associated to homogeneous and intense contrast enhancement. D, On MRI examination, the pathological tissue was characterized by mild hyperintense signal on T1-weighted images compared to tongue signal. An inhomogeneous hyperintensity on T2-weighted, "salt and pepper" like (C, F), and homogeneous enhancement after contrast injection was evident (E, G). The mass lesion appeared well distinguishable with respect to the surrounding anatomical structures.

He introduced the term *infantile myofibromatosis*, referring to the fact that this solitary form most affected children 2 years old or younger (89% of 60 subjects).[3] He also documented the high frequency of the solitary form (75%) and noticed that most involved anatomical sites are head and neck area, (in particular skull, eye socket, and parotid gland) with a recurrence of 35%.[3] Other cases of oropharyngeal solitary myofibromatosis have been previously described, but it is the first case of soft palate involvement[4–9]. Diagnosis is challenging, since this tumor can be misdiagnosed with other lesions affecting oropharyngeal area[2,10–12] such as the following: (*a*) fibrosarcomas that present low-contrast enhancement after in CT scan, whereas in MR, they show hypoisointensity of signal in T1- and T2-weighted images for collagen producing a few enhancement, irregular borders, and bone erosion signs; (*b*) rhabdomyosarcomas that present similar features of signal intensity to squamocellular neoplasms[13] with hypointensity of signal in T1-weighted, which can vary the regular hyperintensity of signal linked to the physiologic presence of mucous glands; (*c*) minor salivary gland adenomas in the soft and hard palatal region have been reported too; and in this case, some neoplasms have demonstrated intermediate intensity of signal in T1 compared with muscle and fatty tissues, and a high intensity in T2 with irregular borders depending on the neoplasms histotype. Whereas malignant neoplasms show irregular borders, the MR shows the internal structure of minor salivary gland tumors in a multidirectional view, with the ability of finding out neoplasm. In our experience, we have noticed that many radiologic features of an initial CT examination had lead to a wrong diagnosis. It is known that this kind of lesions shows intense enhancement after contrast material injection, but it is not a specific finding. Reports about MR examinations in infantile myofibromatosis are rare and with different pathways.[13] The signals range from low-intensity foci on T1-weighted images and bright signal on T2-weighted images to a ''target'' appearance before and after gadolinium described by other authors.[13] The MR examination leads to obtain a reliable diagnosis. The inhomogeneity of signal in the involved tissue in T2 and the high cellularity associated to lower contents of collagen result in another benign parameter as well.[13] Benign nature of the lesion was suspected because of the absence of perilesional edema, distinct limits, and the homogeneous enhancement after contrast injection. The hypothesis based on MR examination result was confirmed by complete excision during the surgical procedure and the absence of relapse 8 years after surgery. Excision in free margin represents the only strategy of treatment[3,12] in similar conditions.

In conclusion, even if infrequent, infantile myofibromatosis has to be suspected in case of soft tissue mass affecting the oropharyngeal area in young patients. Correct clinical and radiological preoperative assessments represent the key points to perform the right treatment. Magnetic resonance imaging seems to be the best diagnostic option, allowing for exclusion of the malignancy of the lesion. Resection in free margins is the treatment of choice to avoid recurrence.

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Rhinolith Induced by a Foreign Body

To the Editor: In this report, we present an unusual and interesting case of a huge rhinolith formation caused by an exogenous foreign body (unidentified plastic object) lodged in the nasal cavity. In addition, we focus on the imaging findings, particularly the contribution of computed tomography (CT), and emphasize that preoperative radiologic evaluation is necessary to diagnose and evaluate rhinolithiasis.

A 34-year-old healthy man who complained of nasal obstruction, purulent discharge, and severe halitosis on the left side for 3 years came to our hospital. An irregular, yellowish, and stony

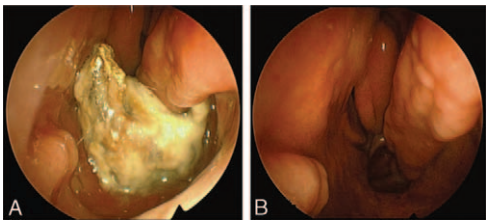

FIGURE 1. A, Nasal endoscopy revealed a huge rhinolith located in between the nasal septum and the inferior turbinate of the left nasal cavity. B, Two weeks later after removal of the rhinolith, the left nasal cavity is clear on nasal endoscopy.

mass covered with purulent secretions was visible in the left nasal cavity on nasal endoscopy. It was adherent to the septum, the turbinates, and the nasal floor. In addition, nasal mucosa around the rhinolith was edematous and congested, and this mucosal change induced pain whenever the rhinolith moved (Fig. 1A). Although our provisional diagnosis was rhinolithiasis on the basis of nasal endoscopy, paranasal sinus CT was performed to evaluate the involvement of paranasal sinuses and the relationship with surrounding tissues. It showed a mass measuring approximately $2.3 \times 1.9 \times 2.1$ cm with irregular calcification and air trapping in the central portion and no evidence of invasion of paranasal sinuses (Fig. 2). We achieved to remove the rhinolith by using various forceps and piecemeal via nasal endoscopy under general anesthesia to minimize mucosal injuries. During this procedure, we found a plastic cap in the central portion of the rhinolith (Fig. 1B). He has been completely relieved of his symptoms after the operation. On endoscopy, we confirmed that the mucosa in his nasal cavity is clear when following up 2 weeks after the operation (Fig. 3).

Rhinoliths are most typically located in the floor of the nasal cavity, either between the maxillary sinus wall and the inferior turbinate or between the inferior turbinate and the nasal septum, halfway between the anterior and posterior nares.[1] Foreign bodies are usually introduced during childhood, being almost in the nasal floor. It causes an inflammatory reaction, leading to deposits of carbonate and calcium phosphate, magnesium, iron, and aluminum, besides organic substances such as glutamic acid and glycin, leading to a slow and progressive increase in size.[2] In this case, we find an unrecognized foreign body in the center of the rhinolith. We think that this exogenous foreign body acts as the nidus of the rhinolith.

Calcified nasal mass on CT images should raise the suspicion of other pathologic entities such as calcified polyp, ossifying fibroma, odontogenic tumor, osteoma, osteosarcoma, osteomyelitis, and carcinoma.[3] Although a definite diagnosis may be possible only by endoscopy such as in our case, CT is the most valuable tool in evaluating the patient with the rhinolith and can enable to identify the location and the size of the rhinoliths and evaluate the involvement of paranasal sinuses requiring treatment. On CT, rhinoliths commonly appear as a homogenous, high-density (metallic) lesion with smooth mineralization, but this is not a pathognomonic finding.

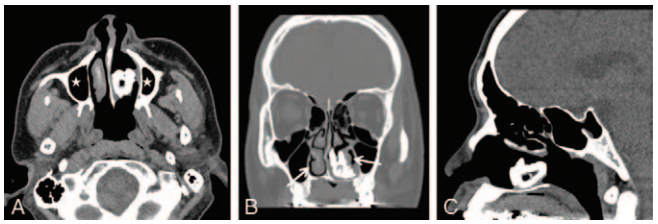

FIGURE 2. CT scans show a metallic high-density mass with irregular calcification and air trapping but without any involvement of paranasal sinuses. In addition, nasal septal deviation to the right side, poor development of the inferior turbinate (white arrow), and hypopneumatization of the maxillary sinus (asterisk) in comparison with the contralateral side are observed. A, Axial view; B, coronal view; C, sagittal view.

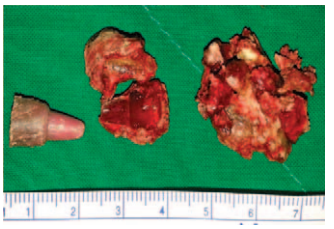

FIGURE 3. The specimen is an unidentified plastic cap in the central portion of the rhinolith after totally endoscopic removal.

Another finding of CT scan is that the central portion of the rhinolith, which may contain organic material, may be of somewhat lower density, or a foreign body nidus may be evident.[1] In the current case, CT images showed irregular calcification with air trapping in the central portion of the calcified mass, which is helpful to assume that the exogenous foreign body acts as the nidus of the rhinolith. Other notable findings are nasal septal deviation to the right side and poor development of the inferior turbinate and the maxillary sinus in comparison with the contralateral side. We suppose that the slow-growing rhinolith induced by an inserted and unrecognized nasal foreign body during childhood may lead to these findings. In other words, the long-term direct pressure effect induced by the huge rhinolith may cause nasal septal deviation, hypopneumatization of the maxillary sinus, and poor development of the inferior turbinate, such as in the current case. In conclusion, we emphasize that CT may be helpful to evaluate the nidus of the rhinolith and predict the shape of the exogenous foreign body acting as a nidus.

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Compound Odontoma in a Pediatric Patient With Aspects Similar to Complex Odontoma

To the Editor: According to the World Health Organization, odontomas are classified in 2 categories: compounds or complexes. In compound odontomas, all the dental tissues have a homogeneous distribution with a toothlike lesion and prevalence in the maxillary anterior region. On the contrary, in cases that the calcified dental tissues do not have toothlike contours, they are denominated

complex odontomas with prevalence in the mandibular posterior region.[1–7]

In a 9-year-old male patient complaining of a gradual and painless mass in the mandibular posterior region involving teeth 75 and 36 with 6 months of evolution, intraoral examination revealed a lesion with no definite contours on the distal of tooth 43 to the mesial of tooth 36, involving tooth 75. Laterally, the mass extended to the vestibular-buccal plane. The panoramic radiographs and computed tomography (CT) revealed a unilocular lesion with defined contours located in the left mandibular molar extending from the posterior region of tooth 33 to the mesial of tooth 16. The lesion was circumscribed with a nondefined radiotranslucent contour in the superior portion (Fig. 1A). The expansion of the lingual and buccal cortex was clearly visualized on CT images (axial slices) (Figs. 1B–E). A central complex odontoma was proposed as a differential diagnosis based on the radiographic images.

The surgical removal of the lesion with an intraoral access was performed with general anesthesia (Figs. 2A–C). After the surgical procedure, the surgical handpiece was referred for histopathologic examination. Macroscopically, the surgical handpiece had fragments composed of bone tissue with soft tissues, and the dimensions of the conjunct were $4 \times 4 \times 0.8$ cm. The histopathologic analysis revealed areas of disorganized dental tissues and several structures composed of mineralized tissues similar to small teeth (Fig. 2D). The histopathologic diagnosis was compound odontoma.

The histopathologic aspect of the complex odontoma showed small spaces with pulp tissue, enamel matrix, and epithelial remains. The latter was observed inside a mineralized and calcified dentin mass. A fibrotic thin capsule or a cystic contour is observed around the lesion.[8,9] However, in the current study, the microscope analysis of the histopathologic examination revealed several mineralized structures, similar to small teeth. Thus, the definitive diagnosis was compound odontoma. Image examinations postoperatively at 1 day (Fig. 2E) as well as at 2 and 6 months (Figs. 2F, G) showed increased radiographic bone density, compatible with normal bone healing.

Therefore, the early diagnosis and treatment of odontomas are extremely important to avoid complications, that is, the retention of primary teeth and the noneruption of permanent teeth.

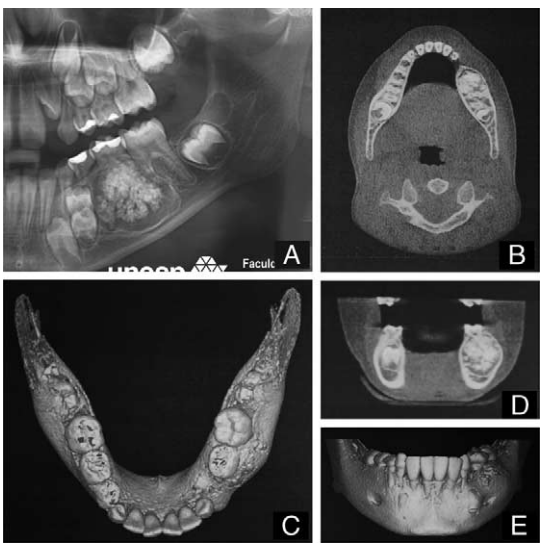

FIGURE 1. Radiographic image (A), CT scan (B and D), and three-dimensional reconstruction (C and E).

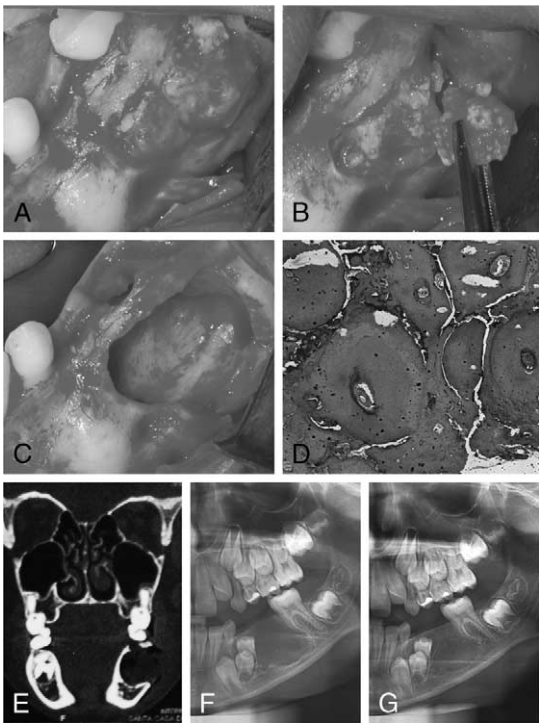

FIGURE 2. Lesion exposition (A), exeresis (B and C), and histopathologic characteristic of compound odontoma (D). Image examinations postoperatively at 1 day (E) as well as at 2 and 6 months (F and G) showing increased radiographic bone density, compatible with normal bone healing.

Histopathologic examination is essential to the correct diagnosis even if the macroscopic characteristics are well defined.

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The Use of a Rubber Seal to Prevent Mucosal Injury in the Treatment of Mandibular Fractures With Intermaxillary Fixation

To the Editor: The treatment of maxillofacial fractures involves different methods such as bandages and splinting, open reduction and internal fixation, and intermaxillary fixation (IMF).[1,2] Although arch bars provide an effective and versatile means for IMF, their use includes such consequences as risk of penetration injury to the surgeon or oral buccal mucosa, increased surgical time for both placement and removal, trauma to the periodontium, and compromised oral hygiene.[2] Also, interradicular miniscrews may be associated with damage to the buccal mucosa.[3]

We present a modification using a rubber seal to prevent buccal mucosal injury caused by screws when using IMF for mandibular fractures. In the technique, after placing bilateral acrylic posterior bite blocks, intermaxillary fixation was made using screw and running wire. Screw position was superior and medial to the upper canine root tips as well as inferior and medial to the lower canine root tips. To prevent mucosal injury during the treatment, a rubber seal was taken from a 5-cc syringe and placed between the screw and mucosa (Figs. 1, 2, 3). The patients were followed by maintaining the fixation of the maxilla and mandible for 4 weeks. After stable union was achieved, the wires, screws, and bite blocks were removed. Then, soft diet was maintained for 2 weeks.

In conclusion, the rubber seal was able to prevent mucosal injury during the treatment. Any patient complaint was not seen and postoperative period was uneventful. We recommend the use of the technique as a safe, easy, and effective fixation method when using intermaxillary transmucosal fixation screw in the treatment of mandibular and/or maxillary fractures.

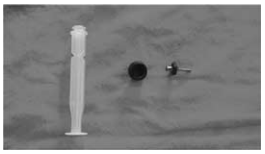

FIGURE 1. Rubber seal was taken from a 5-cc syringe.

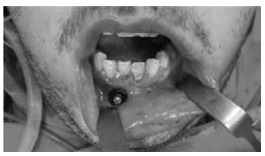

FIGURE 2. Rubber seal was placed to keep intact the buccal mucosa between the screw and mucosa.

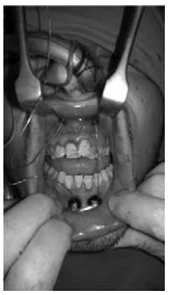

FIGURE 3. Wire was used to provide IMF.

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Hypopharyngeal Carcinoma With Pancreatic Metastasis

To the Editor: Hypopharyngeal squamous cell carcinoma (HSCC) is mainly a locoregional disease. The overall incidence of distant metastases in HSCC is 9% to 11%.[1,2] Although distant metastasis tends to occur late in the course of the disease, it is a major determinant of management and prognosis. Approximately 25% of hypopharyngeal cancer cases develop distant metastases within 12 months after treatment. The most frequent site for distant metastasis is lung, accounting for approximately 70% of the cases. It is followed by bone, liver, and skin.[3] Solid organ metastasis other than these sites is very rare.[4] We present a patient with a solitary pancreatic metastasis from a treated squamous cell carcinoma of the pyriform sinus.

A 62-year-old Caucasian male patient was admitted to our clinic for a biopsy-proven squamous cell carcinoma of the hypopharynx. On physical examination, a 3.5 × 1.5 cm soft tissue mass was noted in the left pyriform sinus extending to the posterior pharyngeal wall. Vocal cords were intact and mobile bilaterally. There was a palpable right-sided level II lymph node (1.8 × 1.5 cm). Whole-body PET/CT scan showed a large hypermetabolic hypopharyngeal mass, predominantly in the left pyriform sinus extending to the posterior hypopharynx (Figs. 1A, B). There were hypermetabolic left level III, right level II, III, and IV lymph nodes. There was no evidence of metastatic disease. Disease was clinically and radiologically classified as T2N2M0 (stage IVa). Our treatment of choice was organ preservation therapy. He underwent chemotherapy with high-dose cisplatin and concurrent 70 Gy radiation therapy to the neck in 35 fractions.

After chemoradiation therapy, control PET/CT demonstrated visible decrease in tumor volume and improvement in cervical lymphadenopathy. However, a new hypermetabolic mass was noticed in the celiac axis region (Fig. 1D). On abdominal CT scan, a heterogeneous large ill-defined pancreatic mass (8.1 × 5.6 cm) was noted along the superior aspect of the body of the pancreas encasing the celiac axis, splenic artery, and vein. There was heterogeneity and mild ductal dilatation in distal body and tail of the pancreas (Fig. 1C). CT-guided biopsy of the pancreatic lesion revealed poorly differentiated squamous cell carcinoma stained positive with p63 and CK5-6 (Fig. 2). Tumor cells were negative for TTF-1, CK7, CK20, and mucicarmine staining. Three additional cycles of cetuximab, paclitaxel, and carboplatin treatments were administered for metastatic disease.

Clinically, hypopharyngeal cancers are known to demonstrate an aggressive nature, which is characterized by diffuse local spread and an early and relatively high rate of distant metastases. Approximately 17% of head and neck cancers have distant metastases when clinically diagnosed.[4] The most common site of metastasis for advanced head and neck cancer is lung. In a study by Alvi et al, lung (67%) was identified as the most frequent site for distant metastases, followed by bone (17%), skin (10%), and liver (7%).[3] However, there has been no report in literature with pancreatic metastasis from a primary head and neck cancer. In this patient, pancreatic lesion was identified incidentally during control PET/CT. Vague abdominal pain, fatigue, and malaise were noticed only in retrospective questioning. Abdominal CT displayed the location and extent of disease with contrast enhancement. CT-guided biopsy and pathologic evaluation of the tissue specimens revealed the origin of the pancreatic tumor.

To obtain higher cure rates, it is important to diagnose delayed regional and distant metastasis at an earlier stage. This is especially true for hypopharyngeal carcinomas. Distant metastases of hypopharyngeal cancers are common especially in advanced primary lesions and those with nodal involvement.[4,5] Therefore, patients with advanced-stage HSSC should be monitored closely for prognostication.

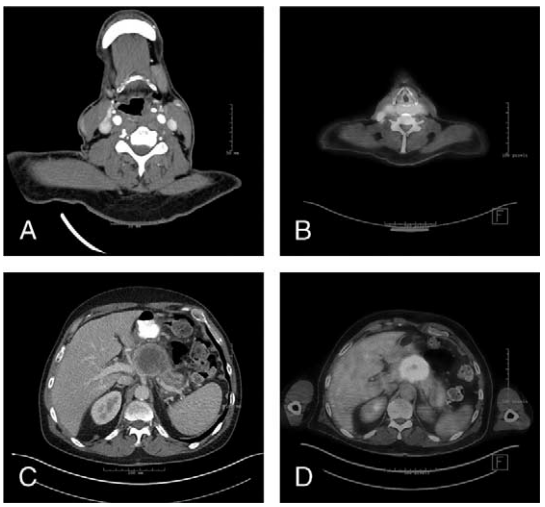

FIGURE 1. Neck CT (A) and PET/CT (B) imaging demonstrates an asymmetric, hypermetabolic soft tissue mass in the left pyriform sinus, extending toward the post cricoid region. C, A heterogeneous large mass in the upper abdomen with distal pancreatic atrophy and mild ductal dilatation is shown in abdominal CT. D, Abdominal PET/CT reveals a hypermetabolic mass in the celiac-axis region concerning for neoplastic process.

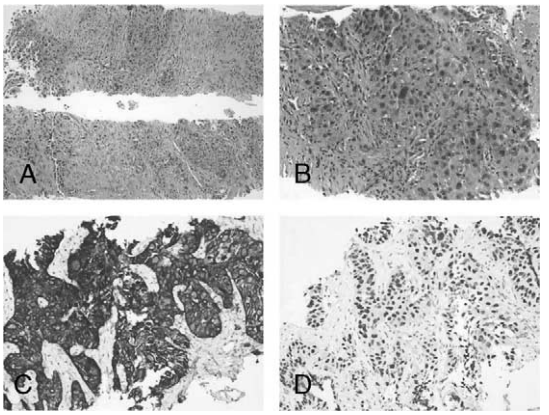

FIGURE 2. A, Sections from the pancreatic biopsy show nests of neoplastic epithelial cells surrounded by desmoplastic stroma (hematoxylin and eosin stain, original magnification ×10). B, The tumor cells show syncytial arrangement, pleomorphic nuclei with prominent nucleoli (hematoxylin and eosin stain, original magnification ×20). The tumor cells show (C) cytoplasmic and membranous positivity for CK5/CK6 and (D) nuclear staining for p63 (immunohistochemical stain, original magnification ×20).

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Unexpected Findings in Neck Dissection for Laryngeal Squamous Cell Carcinoma: Duplication of Left Internal Jugular Vein

To the Editor: Internal jugular vein (IJV) is the major vein of the head and neck and an important landmark in neck dissection. Duplication of internal jugular vein is an unusual anatomical variation with an estimated incidence of 0.4% among the general population.[1] The anatomical course of the jugular veins are usually very consistent, but variations such as duplication may cause iatrogenic injuries.[2] We present a case with duplication of left IJV. The goal of the present report was to discuss the clinical and radiological importance of these rare anatomical variation.

CLINICAL REPORT

Our patient was a 52-year-old man with T4N2aM0 transglottic squamous cell carcinoma of the larynx. Total laryngectomy and bilateral functional neck dissection were planned. During the dissection of the left side of the neck, we observed duplications of the jugular vein. Duplicated vein separated at the facial and jugular vein junction and poured into the left brachiocephalic vein at the level of jugular and subclavian vein junction (Fig. 1). Computed tomography angiography (CTA) scans showed the separation and draining levels exactly in the postoperative period (Fig. 2).

DISCUSSION

Duplication of the IJV may be unilateral or bilateral. Unilateral duplications have been reported more.[1] Previous reports of IJV variations have used the terms "duplicated" and "fenestrated". Downie[2] et al emphasized the main difference between these 2 conditions. Characteristics of duplicated IJV is 2 separate branches along the normal pathway and separately draining into the subclavian vein, whereas in fenestrated IJV, it divides into 2 separate veins and join together to form a single vein before draining into the subclavian vein.[2] We see incorrect usage of "duplication" term for many cases with fenestration in the literature.

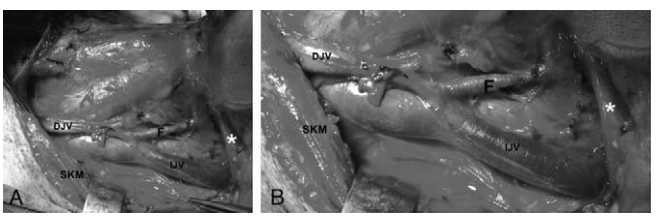

FIGURE 1. A and B, Intraoperative view of the left internal jugular vein duplication. F indicates facial vein; IJV, internal jugular vein; white asterisks, digastric muscle; SKM, sternocleidomastoid muscle; DJV, duplication of the left internal jugular vein.

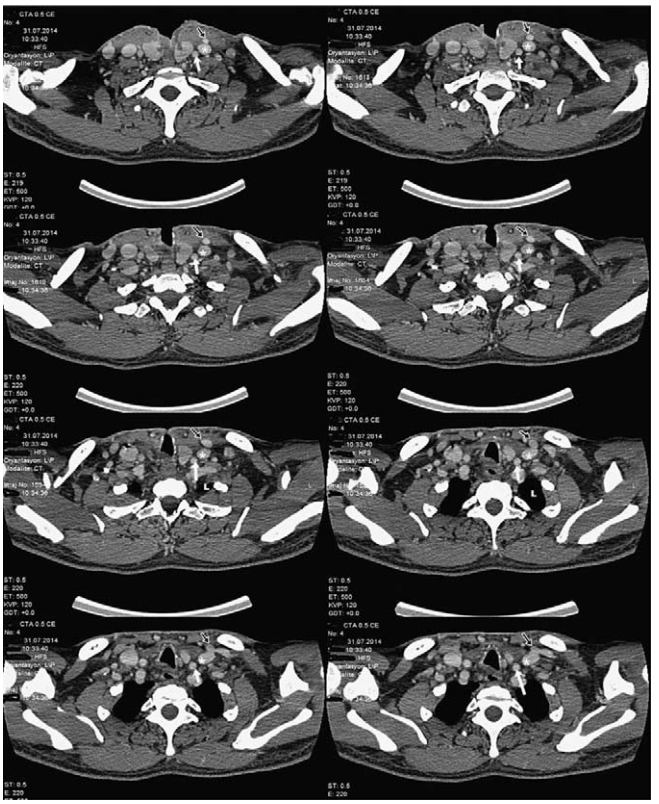

FIGURE 2. Axial postoperatively taken computed tomography angiography scans. Black arrows indicate left duplication of the jugular vein; white arrows, left common carotid; white asterisks, internal jugular vein.

Etiology of IJV duplication is unclear. Three main hypothetical explanations have been proposed to describe the etiology of duplicated IJV: vascular, neural, and bony hypothesis. The vascular hypothesis notices the paucity or absence of muscular layer in the vein that might be responsible for the duplication; neural theories are based on the relation between the vein and the spinal accessory nerve and therefore relate only to the division of the vessel in the upper third of the neck. The bony hypothesis relates to anomalies of the vein at the level of the jugular foramen.[1,3] None of the theories explain the lower duplications and the possible role of omohyoideus muscle. Previously, Bachoo and Evans[4] noticed the importance of omohyoideus muscle on the lower duplications of IJV.[4]

CTA demonstrates the whole tract of duplicated vein and adjacent vascular structures exactly.[5] In our opinion, it is useful for the distinction between fenestration and duplication.

The surgeon should know the exact position and possible variations to prevent complications. Intraoperative discovery of duplicated IJV makes neck dissection more time consuming and complicated. IJV is one of the mostly preferred vessel for central venous catheterization. It is important for any surgeon, physician, or intensive care practitioner to be aware of this anatomical variation because cannulation of a duplicated IJV may lead to iatrogenic injuries and life-threatening situations. In addition, radiologists have to be aware of such anomalies when reporting imaging of the head and neck. They may be missed because they are uncommon cases. These kinds of anomalies should be clearly documented in the medical records.[6]

CONCLUSIONS

The major outcomes from this report are as follows:

- Duplication of IJV is a rare condition that is usually detected incidentally.
- Failure to recognize this variation may result in iatrogenic damage during neck surgery or central venous catheterization.
- The clinician or surgeon has to be aware of these kinds of anatomical variations, and medical records should be assessed before invasive procedures.
- CTA is useful for the diagnosis and distinction of these variations from fenestration.

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Hyaline Ring Granulomas in a Dentigerous Cyst

To the Editor: Hyaline ring granulomas (HRGs), which were first described in 1971 by Lewars,[1] are characterized by hyaline rings or ovoid homogenous/fibrillar hyaline masses embedded in fibrous connective tissue permeated by variable numbers of inflammatory and multinucleated giant cells.[2,3] The etiopathogenesis of these uncommon histopathologic findings remains a matter of discussion, and 2 opposing theories have been proposed. The exogenous theory suggests that HRGs are derived from the implantation of foreign material (vegetable food particles, therapeutic agents).[1,2,4] In contrast, the endogenous theory proposes that these structures are the result of hyaline degeneration of the walls of blood vessels.[5]

Hyaline ring granulomas have been observed in lesions of the lung, skin, gallbladder, fallopian tubes, and oral cavity.[4,6] In the latter case, HRGs have been identified in both extraosseous[6] and intraosseous lesions, particularly radicular cysts, residual radicular cysts, and paradental cysts.[2,3] Reports of the occurrence of HRGs in developmental odontogenic cysts are rare.[7,8] Here, we report the third case of HRGs identified in a dentigerous cyst and discuss the etiopathogenesis and clinical-pathologic features of this entity.

A 7-year-old boy was referred for evaluation of a painless, bluish swelling located in the left anterior region of the mandible. Palpation of the lesion showed a soft consistency, a finding compatible with the breakdown of cortical bone. The deciduous canine in the affected region showed loss of pulp vitality, a small extrusion, and tooth mobility. Panoramic radiography revealed a unilocular, well-delimited, radiolucent lesion associated with the crown of the unerupted permanent canine (Fig. 1A).

The diagnostic hypothesis was a dentigerous cyst, and an incisional biopsy was performed. Histopathologic examination showed a

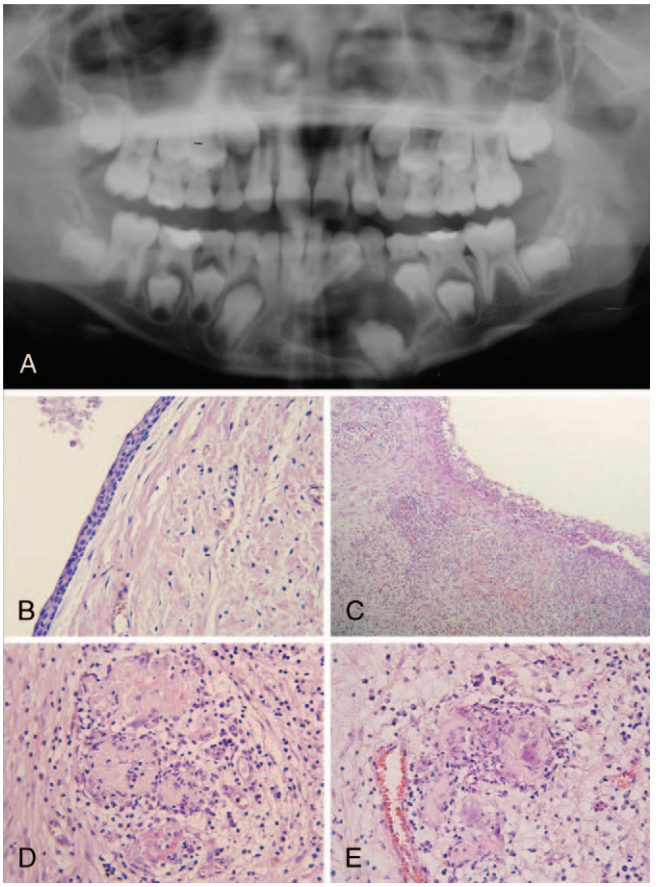

FIGURE 1. A, Panoramic radiograph showing a well-demarcated, unilocular radiolucency area involving the crown of the unerupted lower left permanent canine. B, Cystic cavity lined with thin nonkeratinized stratified squamous epithelium without rete ridges (hematoxylin-eosin [H/E], original magnification ×400). C, Epithelial lining showing extensive hydropic degeneration, spongiosis, and exocytosis. Fibrous capsule exhibited intense inflammatory infiltrate (H/E, original magnification ×100). D, Hyaline ring granulomas in the form of roughly circular homogeneous or fibrillar masses exhibiting a corrugated border lying in fibrous connective tissue infiltrated by inflammatory cells (H/E, original magnification ×400). E, Multinucleated giant cells inside and adjacent to hyaline material (H/E, original magnification ×400).

pathologic cavity lined with nonkeratinized stratified squamous epithelium consisting of few layers with a flat interface, which exhibited areas of hyperplasia, hydropic degeneration, spongiosis, and exocytosis (Figs. 1B, C). Foci of intense mononuclear inflammatory infiltrate and areas of hemorrhage were observed in the surrounding fibrous capsule. In addition, small homogenous/fibrillar ovoid hyaline masses associated with multinucleated giants cells were seen in the capsule. These findings were compatible with HRGs (Figs. 1D, E). Marsupialization of the lesion was performed on the basis of the definitive diagnosis of an inflamed dentigerous cyst. Panoramic radiography performed 18 months after marsupialization showed a marked reduction of lesion size and the progression of rhizogenesis of the associated tooth. The patient remains under follow-up.

Hyaline ring granulomas are uncommon histopathologic findings. In the oral cavity, HRGs were usually identified in inflammatory odontogenic cysts and approximately 58 cases have been reported in the English literature.[3] On the other hand, there are only 2 reports of HRGs in developmental odontogenic cysts (Table 1).[7,8] The current study is the third report of HRGs occurring in a dentigerous cyst (PubMed Database).

In a retrospective study of 661 cases of inflammatory odontogenic cysts, Henriques et al[3] reported a higher relative frequency of HRGs in residual radicular cysts (6.1%), followed by paradental cysts (5.6%) and radicular cysts (3.0%). These authors suggested that their findings support the concept of an exogenous origin of HRGs in inflammatory odontogenic cysts because sites of previous tooth extraction and pericoronitis in lower third molars, associated with food stagnation in the affected area, are possible routes of implantation of food particles. The results of histopathologic,[3,6] immunohistochemical,[3] and ultrastructural[6] studies agree with the suggestion that the implantation of exogenous material, particularly leguminous food, is responsible for the formation of oral HRGs.

The presence of HRGs has also been associated with the implantation of exogenous material in developmental odontogenic cysts. In this respect, in the case reported by Ide et al,[8] HRGs were observed in the capsule of an infected dentigerous cyst in which a communication was established between the cyst lumen and oral cavity through a fistula. In the current study, there was no evidence of an obvious portal of entry for an exogenous particle into the cystic lesion. However, exogenous material may have entered through the periodontal ligament as a consequence of the breakdown of buccal cortical mandibular bone associated with extrusion and mobility of the deciduous canine in the affected area. In agreement with this suggestion, the periodontal pocket has been reported to be a possible entry site of exogenous material, which could give origin to oral HRGs.[8]

In contrast to the findings reported earlier, Chen et al[7] described a case of HRGs in the fibrous capsule of a dentigerous cyst without a history of previous trauma or surgical intervention in the area. According to the authors, HRGs arise from a pool of extravasated serum proteins that undergo coagulation. Supporting a possible endogenous origin of these structures, Dunlap and Barker[5] suggested that HRGs are the result of hyaline degeneration of the walls of blood vessels induced by localized acute vasculitis. Moreover, it has been reported that these microscopic findings may be formed by degenerated collagen fibers.[3,6] In view of the small number of reports of HRGs in developmental odontogenic cysts, clinical, histopathologic, immunohistochemical, and ultrastructural studies involving large case series are needed to elucidate the etiopathogenesis of these microscopic findings in these lesions.

TABLE 1. Reported Cases of Developmental Odontogenic Cysts With Hyaline Ring Granulomas According to Cyst Type, Age, Sex, and Anatomic Location

Authors	Cyst Type	Age, y	Sex	Anatomic Location
Chen et al[7]	Dentigerous cyst	20	Male	Maxilla
Ide et al[8]	Dentigerous cyst	21	Female	Mandible
Current study	Dentigerous cyst	7	Male	Mandible

In the retrospective study of Henriques et al,[3] only 22 (3.3%) of the 661 cases of inflammatory odontogenic cysts exhibited HRGs, a fact suggesting a low frequency of these microscopic findings in these lesions. In addition, HRGs generally appeared as small and inconspicuous structures in the inflammatory odontogenic cysts studied. Similarly, in the current study, HRGs were only identified after careful histopathologic examination of the lesion. Taken together, these findings suggest that the small number of reports of HRGs in developmental odontogenic cysts[7,8] might be related to careless microscopic evaluation of these lesions during routine laboratory diagnosis. On the other hand, detailed retrospective histopathologic studies involving large case series of developmental odontogenic cysts are needed to establish the true frequency of these microscopic findings in these lesions.

In summary, the current study highlights the importance of thorough histopathologic assessment of odontogenic cysts to establish the true frequency of HRGs in these lesions and to elucidate their etiopathogenesis.

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Emergency in Temporomandibular Joint Ankylosis

To the Editor: Aspiration pneumonia, which is common in patients with anatomic abnormalities of the upper aerodigestive tract or elders with neurologic dysphagia and disruption of gastroesophageal junction, develops owing to inhalation of colonized secretions from the oropharynx with entry into the lungs.[1] We report a rare case of emergency and definite management of combination of aspiration pneumonia, obstructive sleep apnea syndrome (OSAS), and temporomandibular joint (TMJ) ankylosis in a neglected girl aged 7 years.

CLINICAL REPORT

A 7-year-old, malnourished girl known to have TMJ ankylosis was referred to our emergency department from a primary health center for further management of breathlessness for 2 days with pyrexia, cough, and cold for 8 days. Her parents gave history of forcep's delivery, recurrent chest infection, snoring and sudden awakening during sleep, daytime sleepiness, as well as progressive limitation of jaw opening. On general examination, she was drowsy, with bilateral crypts and wheeze with labored breathing as well as decreased air entry bilaterally with severe intercostal recession and supraclavicular recession with classic features of bilateral TMJ ankylosis (Figs. 1A, C). She was diagnosed with respiratory distress with bilateral pneumonia secondary to silent aspiration with TMJ ankylosis and OSAS. On the second day, the patient developed sudden bradycardia with drop in Spo_2 warranting emergency tracheostomy. Cone beam computed tomographic scans showed bilateral TMJ ankylosis with severe narrowing of airway, elongated coronoid, and retropositioned chin. (Figs. 1B, D). On resolution of pneumonitis and systemic improvement, a 2-stage management of TMJ ankylosis and OSAS was undertaken. Bilateral mandibular distraction with horizontal flip pedicled (HFP) genioplasty was done in the first stage (Fig. 1F), and release of bilateral TMJ ankylosis, coronoidectomy, as well as temporalis muscle interposition with costochondral reconstruction of ramus-condylar unit with distractor removal were done in the second stage 3 months later. At the 6-month postoperative follow-up, the patient had a 25-mm interincisal opening and showed complete elimination of OSAS with widening of airway (Figs. 1E, G-J).

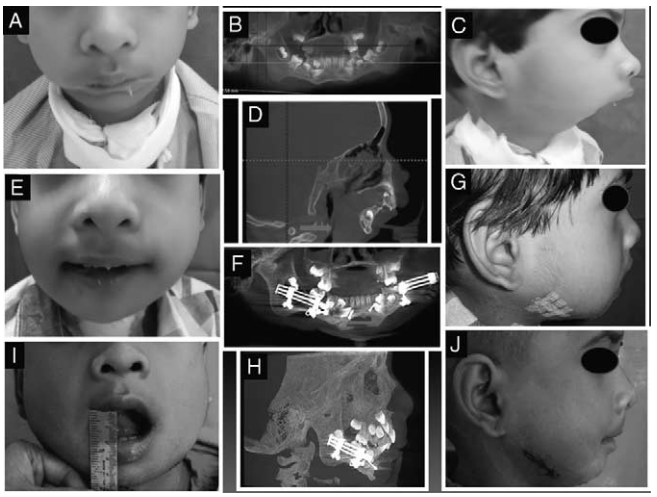

FIGURE 1. A to C, Predistraction clinical and radiologic images showing receded chin and bilateral TMJ ankylosis. D, Radiographic airway assessment showing narrowing of airway; E to G, Postdistraction clinical and radiologic images with lag screw (arrow) in situ used to fix genioplasty. H, Postdistraction widening of airway with distractors in situ (arrow). I to J, Postankylosis release.

DISCUSSION

Temporomandibular joint ankylosis, which is a serious and disabling condition, presents with multiple problems. Its signs and symptoms are proportional to the degree of ankylosis, nature of ankylosis, age of onset of ankylosis, and age at which treatment is initiated.[2] In our case, tracheostomy was performed to secure airway on emergency basis and aid management of pneumonitis. Bilateral osteodistraction of the mandible was done to achieve a long-term increase in oropharyngeal airway and improvement in facial profile. One disadvantage of distraction osteogenesis is that it fails to address the deficient chin, which, in our case, was achieved through HFP genioplasty, which also relives OSAS immediately by advancing the tongue and its musculature.[2] Reconstruction of ramus condyle unit with costochondral graft was thought to be the best option at the age of 7 years to establish functional joint and facial appearance.[3]

CONCLUSIONS

1. In TMJ ankylosis, treatment must be instituted at the earliest, to avoid an emergency situation such as that in our patient.
2. We recommend distraction osteogenesis, followed by release of ankylosis, for better control of distraction vector.
3. In cases of TMJ ankylosis with OSAS, we recommend HFP genioplasty during distraction to achieve immediate advancement of suprahyoid group of muscles and increase in airway space.

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Efficient Utility of WhatsApp: From Computer Screen to the Surgeon's Hand to Determine Maxillofacial Traumas

To the Editor: Advances in communication technologies and mobile phones have been improving rapidly for 10 years. These advances and developed applications (Apps) compatible with smartphones have led them to be a crucial device for their users, especially that apps let people use smartphones like a computer. It is obvious that the smartphones are much more than just a phone.[1] On the other hand, the smartphones are used commonly among doctors too, and there are lots of medical apps. WhatsApp (WhatsApp, Inc., Mountain View, CA) is the most popular instant messaging application for smartphones. The users may send and receive location information, images, video, audio, and text messages in real time to individuals and groups of friends at no cost.[2]

We used WhatsApp application as an image and video transfer program to send computerized tomography (CT) sequences between emergency service and plastic surgeons in the night-time consultations on the patients having maxillofacial traumas.

For sending and receiving images and videos, the smartphones having both WhatsApp and high-definition camera were used. The physician at the emergency service took the view of CT images from a computer screen and sent the images (Fig. 1A), and serial images were recorded as videos (Fig. 1B) on app. Due to high-definition cameras on the smartphones, it is possible to narrow and magnify the images. Moreover, sent images or videos did not need any specific programs to run them. It provided us to see the images immediately and to easily determine whether there was fracture.

Plastic surgeons get benefits from technological advancements,[3] and it is easy to adopt these advancements to their professional lives. Tele-consultation is a well-known and established way of evaluating patients.[4] It is utilized in the examination of soft tissue injuries and burns at emergency services.[5] For craniofacial traumas, telemedicine was used too.[6] They needed personal computers, remote access to the server, and specific requirements. Although radiographic images such as CT scans may be viewed on smartphones, they need specific interfaces and apps that allow surgeons to view CT scans over a secure network from a remote location.[7]

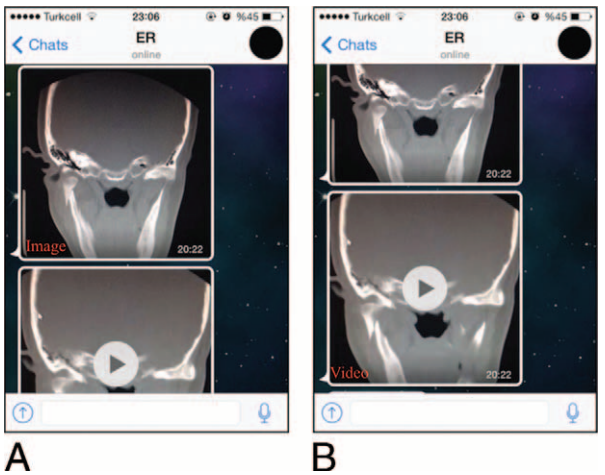

FIGURE 1. Screenshot view of a smartphone: sent CT sequence as image (A) and sent CT sequences as video (B) on WhatsApp chat screen. ER indicates emergency room.

For considering our needs, we thought that during tele-consultation, the fastest, easiest, and most common way should have been used. In our method, the smartphones which have a good quality camera and WhatsApp were used. They are very common recently and are also found in almost all doctors' pockets.[1] Capturing scan images and recording serial views of scans as a video are easy and do not need any specifications. Moreover, the receiver surgeon does not have to be in front of the computer or at home. Wherever he is, he may get and evaluate the views. It allows us to conclude the consultation rapidly.

In conclusion, sending and receiving images and videos on WhatsApp is an easy, rapid way of evaluating of maxillofacial CT scans in the night-time tele-consultation.

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Intramasseteric Arteriovenous Malformation

To the Editor: Less than 1% of all vascular malformations are found in the skeletal muscles. The masseter muscle is the most common location for intramuscular vascular malformations in the head and neck region. Most of the vascular malformations involving the masseter muscle are venous malformations. Arteriovenous malformations within the masseter muscle are extremely rare.[1,2]

A 27-year-old woman presented to our clinic with a complaint of swelling and pain in her right cheek, which she had first noticed 1.5 years ago, after her first pregnancy. Physical examination findings revealed a 4 × 2-cm right preauricular mass that extended to the mandibular border (Fig. 1A). The mass was palpable in the intraoral examination. Magnetic resonance imaging (MRI) with intravenous contrast agent demonstrated a bulging mass within the right masseter muscle, with heterogenous enhancement on T2-weighted images (Fig. 1B). Doppler ultrasonography, along with MRI, confirmed the diagnosis of arteriovenous malformation. Conventional angiography of the external carotid artery, performed by an interventional radiologist, revealed a 3 × 2-cm mass that was fed by the branches of the ipsilateral facial and lingual arteries. Superselective embolization of the feeding vessels with polyvinyl alcohol microparticles had been performed. At the next day of the embolization, the mass was removed surgically. The incision was made along the mandibular border starting from the ear lobule. Skin flaps including the platysma muscle were elevated. Marginal mandibular and buccal branches of the facial nerve were identified and preserved. After the facial artery and vein were ligated, the dissection went up superiorly in the subfascial plane. The mass was removed through a vertical incision made through the masseter muscle (Fig. 1C). There had been no complications in the postoperative period. No recurrence has been detected on physical examination or MRI 2 years after surgery (Fig. 1D).

Intramasseteric vascular malformations commonly present as a swelling in the buccal or preauricular region. Some patients also may have pain with chewing. Owing to their intramuscular localization, intramasseteric vascular malformations do not give skin a bluish hue and are frequently mistaken for parotid tumors in the initial evaluation.[3]

Arteriovenous malformations are unlikely to regress spontaneously and tend to be locally aggressive. The current recommended treatment for symptomatic arteriovenous malformations is superselective embolization, followed by surgical removal in the next 48 hours where appropriate.[1,3] It should also be kept in mind that the embolization is not a procedure without risks. Stroke, cranial nerve palsy, skin necrosis, bleeding, blindness, acute hemodynamic changes, and pulmonary embolism are among the reported complications of the embolization.[1]

An incision starting preauricularly, extending to the mandibular angle inferiorly and to the parasymphysis of the mandibula, if necessary, provides enough exposure for surgical removal of intramasseteric arteriovenous malformations. Smith et al[4] proposed adding superficial parotidectomy to the operation to detect the branches of the facial nerve and to have a better bleeding control. However, the superficial parotidectomy may result in transient facial nerve palsy, contour deformities, and Frey syndrome in many patients who had undergone this surgery. By using the previously mentioned incision, we could identify and preserve the buccal and marginal mandibular branches of the facial nerve without performing superficial parotidectomy. Another approach to reach the lesion within the masseter muscle is using an intraoral incision. Although this approach has an advantage of no visible external scar, the higher risk for facial nerve damage and limited ability to control the hemorrhage outweigh its benefit.

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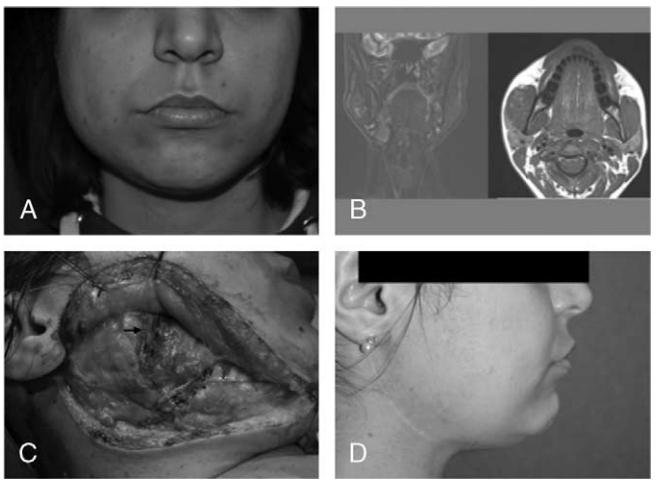

FIGURE 1. A, Preoperative photograph of a 27-year-old woman showing swelling and asymmetry on the lower right side of the face. B, Magnetic resonance image of the neck. A coronal section (left) demonstrates a contrast-enhanced lesion that causes bulging of the right masseter muscle. An axial section (right) of the MRI demonstrates a lesion with ill-defined borders within the right masseter muscle. C, The intraoperative picture shows the site of incision, the plane of the skin flap, and the vertical incision through the masseter muscle (arrow). D, Postoperative second-year photograph of the patient shows a well-healed incision scar along the mandibular border.

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Mandibular Fracture in Bone Graft Harvesting: Is It Possible to Occur?

To the Editor: Dental implant success rehabilitation is related to adequate bone volume. Reconstruction of the posterior maxilla region is required in patients with long time of tooth loss, because of alveolar bone resorption and sinus pneumatization. Autogenous bone graft from the mandible is the most predictable and favorable graft for maxillary sinus augmentation, because of the vital cells transfer that gives the osteogenic property.[1] The main donor sites from the mandible are ramus and symphysis. Mandibular ramus graft volume could be 5 to 10 mL with low reabsorption rates and less complication than mandibular symphysis.[2] Nevertheless, bone fracture and bleeding can occur during graft harvesting.[3] This article aimed to report an incomplete mandibular body fracture during mandibular ramus block graft harvesting for maxillary sinus bone augmentation.

CASE REPORT

A healthy 50-year-old man was referred to the implantology team of Aracatuba Dental School, Univ Estadual Paulista, for dental implant rehabilitation. After clinical examination and image diagnostic analysis was planned, a right maxillary sinus augmentation with autogenous bone graft harvest from the mandible ramus was performed. Surgical procedures underwent under local anesthesia with lidocaine 2% with adrenaline 1:100,000 (DFL, Taquara, Rio de Janeiro, Brazil). The autogenous bone graft was harvested from the right mandible ramus with 701 burr under continuous 0.9% saline solution irrigation. Osteotomies were delimited anteriorly, posteriorly, and superiorly with a burr and inferiorly with a steel disk (Fig. 1A). Afterward, a chisel was used to harvest the bone block; at that time, a lateral mandibular displacement was noticed, which makes the diagnosis possible by direct visualization of the mandibular body fracture, without superior-inferior displacement. This clinical condition was managed by bone block division, to facilitate block removal and avoid mandibular line fracture expansion. The surgical procedure for maxillary sinus augmentation was carried on without any other complication.

A postoperative computed tomography (CT) scan showed an incomplete fracture at the right mandible body (Figs. 1B–D). Because of this, a conservative treatment was performed besides soft-diet indication during 8 weeks. Bone healing occurred without any complication but neurosensory disturbance, which improved spontaneously after 6-month follow-up. Dental implants were placed 6 months after maxillary sinus augmentation.

DISCUSSION

Mandibular autogenous bone graft represents several advantages over extraoral sites for maxillary sinus augmentation such as less resorption rate, excellent incorporation, short healing time, procedures under local anesthesia, and less morbidity. Surgical procedure of bone block harvest from ramus region has minimal discomfort compared with the chin area; however, complications can occur. There are few articles reporting mandibular fractures after bone harvesting[4]; nevertheless, this complication is rare, and the surgical team must be prepared to face it. Treatment options depend on the type of fracture; complete mandibular fractures after bone graft harvesting must be reduced and fixed with 2.4-mm load-bearing plate; on the other hand, an incomplete fracture can be treated conservatively, as was decided to treat this case.

Neurosensory disturbance of the inferior alveolar nerve is another complication of bone harvest procedures, which occurred on this case. It was probably a consequence of bone block displacement for depolarization of myelin sheath. Patient-related spontaneous regression of this condition after 6 months of follow-up.

Mandibular fractures can occur, and they are rare complications after mandibular ramus bone harvesting. Treatment options are related to the type of fracture. In this case, unfortunately, it was diagnosed as favorable fracture that allowed conservative treatment. Neurosensory disturbances are also complications related to graft procedures, and in this case, it was solved spontaneously within 6 months.

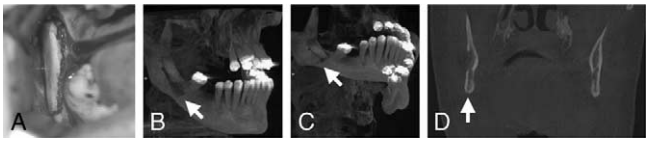

FIGURE 1. A, Clinical image showing bone graft osteotomized in mandibular ramus. B, Volume CT image showing mandibular bone after autogenous bone harvest. C, Basilar incomplete fracture line. D, Coronal CT image indicating mandibular fracture.

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The Speech Language Pathologist's Role in the Cleft Lip and Palate Team

The current essay is a position statement concerning the role of the speech language pathologist (SLP) in the Cleft Lip and Palate Team in Jordan. The past and current medical practices at the level of University Hospitals, Ministry of Health (MOH) Hospitals, Royal Medical Services (RMS), and Private Sector showed that the concept of teamwork started in the mid-1980s at the RMS. Medical centers affiliated to universities have similar protocol of referral of patients with cleft lip and palate (CLP) to that of the RMS. Members of the team usually refer patients for consultations rather than having face-to-face conferencing throughout the management process.

The international practice patterns pertaining to the theme of Cleft Lip and Palate Team were reviewed; the current practice in Jordan was shown to be noncompliant. The barriers to noncompliance and to the inclusion of the speech pathologist in the Cleft Lip and Palate Team and the potential recommendations to improve the current practice patterns in Jordan were stated.

Multidisciplinary teamwork is rapidly growing in the field of medical practice, although there were many discrepancies from country-to-country and region-to-region. It had been generally observed that patients who received team care gained higher benefits in terms of the efficacy of assessment, diagnosis, treatment outcomes, and the cost of management.[1] Studies concerning the management of cleft palate indicated that it was most effective and efficient when the various aspects of treatment were integrative rather administered singly.[2]

The teamwork concept for diagnosis and treatment of patients with cleft lip (CL), cleft palate (CP), or CLP was first pronounced during the 1930s.[3] The presence of the CP team among the medical services in Jordan was not established until mid-1980s by the RMS. The team included members from plastic surgery, audiology, speech pathology, orthodontics, and ENT disciplines. The past and current practice patterns of medical centers affiliated to universities have similar protocol of referral to that of the RMS; the team usually refers patients to its members for consultations rather than having face-to-face management. Although Jordan has a capable reputation in the region for providing high-quality medical services, the role of interdisciplinary team approach was not fully recognized.

The Operation Smile Jordan chapter (OSJ) is currently operating on children from Jordan and the surrounding countries. OSJ was founded in collaboration with Operation Smile, Inc and the MOH in 2000. The team includes Jordanian and international surgeons and other health care professionals. The team provided 153 operations for children who had CLP between 2000 and 2004. The OSJ had promoted the scope of action of the medical missions that provided medical and surgical care for more than 1700 children with CLP since 2004. At the time of this writing, the OSJ is providing postoperative programs including medical and surgical follow-ups and speech therapy services.

Current research showed that the SLP services provided for children with CLP seem to be lacking in most of the developing countries, although teamwork and speech services are well established in almost all of the industrial countries.[4,5] The concept of CLP team is not well established at the level of medical practices in Jordan. In addition, the SLP has limited role among the other members of the CLP team. Despite the fact that the SLP is probably the most knowledgeable team member of the underlying factors of velopharyngeal inadequacy (VPI), and thus, can provide the best recommendation for treatment of VPI.[6]

GOALS

The main goals of the current study were as follows:

- To stress the significance of the interdisciplinary team approach in the management of CLP and provide a model of the ideal team members.
- To emphasize the role and responsibilities of the SLP in the team through the following:
  (a) Emphasizing the significance of the SLP interventions.
  (b) Clarifying the qualifications of the SLP in the CLP team.
  (c) Identifying the barriers facing SLP services in Jordan.
- To provide recommendations for the current practice patterns of the SLP in Jordan particularly provided for children with CLP.

SIGNIFICANCE OF THE CLEFT LIP AND PALATE TEAM ESTABLISHMENT

Because of the unique nature of the congenital malformation in patients with CLP that affects structures involved in many functions, it seems plausible that multidisciplinary specialists should be involved in the treatment of those patients.[2,7] In the United States and Canada, there were known 296 CP and craniofacial anomalies teams with clearly established standards of care and organization.[8] Factors affecting the establishment of CLP teams and proper delivery of speech services depend primarily on individual income, health insurance, available resources in the country, and the availability of the qualified SLP. Those established and functioning CLP teams provided care to patients and may serve as data banks of the possible causes and epidemiology of CLP and craniofacial anomalies.[9,10]

Early primary surgery to repair the hard and soft palate is preferred for speech purposes and feeding. Even following primary palatal surgery, approximately 20% to 30% of individuals born with CP develop speech that is hypernasal.[11] Ideally, every child born with CP should be evaluated for speech and language development by at least 8 months of age or preferably sooner.[12] According to the Royal College of Surgeons of England, these patients are preferred to be seen by at least 1 CLP team member within the first 24 hours.[1] The team should at least consist of the following specialists: SLP, surgeon, and orthodontist.[13] All of the teams need a leader or an identifiable person to direct and coordinate the team activities, chair the conferences, seek information, facilitate and summarize discussion of the team conference, and communicate findings to the parents, patients, and the professional community.[14]

In 1974 and 1975 consecutively, the University of Iowa and the American Cleft Palate Association, each held a conference designed to consider the standards of CLP management. Results stressed the possible effects of the CLP on the patient; that included general health, facial appearance, speech and language skills, maxillofacial and dental-occlusal development, otologic and audiologic health, and psychosocial status. Recommendations stressed that management shall be planned, conducted, and supervised by the team that consists of representatives from plastic surgery, dentistry, speech pathology, pediatrics, otolaryngology, social service, psychology, psychiatry, nursing, audiology, genetics, and radiology.

THE ROLE OF THE SPEECH LANGUAGE PATHOLOGIST IN THE TEAM

The SLP is the team member with the best understanding of the VP function for speech and probably the most knowledgeable in defining the factors underlying the VPI. Therefore, the SLP's recommendation for treatment to eliminate VPI, even if surgical or prosthetic, should be given primary consideration.[6] The role of the SLP on the team has evolved from being primarily a consultant to that of being responsible for the overall management plans as they affect the speech of the patient with craniofacial malformations.[15] The SLP role must be expanded to include input regarding VP function relative to decision making before the surgery consideration.[6]

The SLP has professional training for the assessment of speech characteristics of patients with CP by either perceptual or objective measures. Phonation, articulation, resonance, and overall speech intelligibility are other areas of assessment. Perceptual assessment refers to the judgment of the quality of speech, voice, and resonance of the patient by listening to the patient's speech; usually the therapist must be well trained for this purpose. Many different instruments achieve objective evaluation such as the nasometer that measures the degree of nasal and oral resonance, Visi-pitch (KayPENTAX, Montvale, NJ) that measures pitch registers, and other computerized programs for voice quality. These instrumental evaluations supplement the clinician's perceptual assessment but do not replace it. Articulation evaluation is done by application of tests and phonetically controlled sound passages.

Perceptual and objective assessment of the CLP made by the SLP alone will not provide complete understanding of the anatomic and physiologic variables of the VP mechanism, so multiview video fluoroscopy or nasopharyngoscopy is needed to image the defects directly. Measures must be obtained while the patient is producing samples of connected speech because it has been shown that VP function for the production of simple sounds is not necessarily predictive.[16–18] The SLP is the professional who is

responsible for providing such speech samples and helping the radiologist, ENT, or surgeon interpret the results. In previous years, video fluoroscopic examination was the method of investigation for the VP mechanism, but with the recent advances in technology, nasopharyngoscopy examination is preferred in most centers. It provides the examiner with direct view and requires no radiation. Nasopharyngoscopy results are complementary to radiographic studies but in most cases, they are superior to those obtained through videofluoroscopy. This is because of the excellent clarity of the images and the maneuverability of the scope. Nasopharyngoscopy allows the examiner to see the configuration of the adenoid tissue and observe any fissures in the adenoid pad that affects the firmness of the closure.

Nasopharyngoscopy also allows the examiner to view different VP patterns of closure such as coronal, sagittal, and circular. Defects in the nasal surface of the velum or the presence of an oral-nasal fistula can be identified. Even very small gaps can be visualized with this technique.[19] The SLP who uses standardized measures of articulation and resonance along with nasopharyngoscopy or video fluoroscopy should be able to quantify the component(s) of velopharyngeal dysfunction (VPD). With this information, the SLP assumes the primary responsibility for identifying the specific physiologic conditions, and is therefore able to recommend specific treatment based on this data.[6] The University of Florida Craniofacial Center emphasized the role of the SLP as the team member providing information relative to the functioning of the VP port for speech based on results of multiview videofluoroscopy or nasopharyngoscopy. With this information, the SLP recommends what he or she believes to be the optimal treatment procedure to achieve maximal closure of the VP port during speech. As the actual treatment that some patients received was sometimes different from that recommended, Dixon-Wood et al[6] conducted a retrospective study on 100 patients to identify the frequency of success in eliminating VPI relative to treatment procedures recommended by the SLP versus treatment procedures other than that recommended by SLP. Results showed that 78 patients were treated according to recommendations of the SLP. For these patients, only 8 (10%) continued to demonstrate VPI; however, for the 22 patients who received treatment other than that recommended by the SLP, 7 (32%) continued to demonstrate significant speech problems associated with VPI.

In his study, Skolnick[20] intended to plea the radiologists to include an SLP during the radiologic examination of the VP port. Skolnick explained that most radiologists are usually accustomed with radiologic data only and do not possess adequate understanding of different speech tasks that produce specific VP movement; therefore, radiologic examination of VP port should be a cooperative effort between the radiologist and the SLP. The responsibility of the SLP would be first to explain the procedure to the patient, to rehearse the speech tasks to be performed with the patient before the radiologic examination, and to be completely familiar with patient's speech difficulties. Second, the SLP must be present during the performance of the radiologic examination. The SLP must be monitoring the radiologic images and, if necessary, modify the speech tasks to emphasize abnormalities suggested by the images. The SLP and other members of the CLP team should attempt to educate the radiologist on VP port function under conditions of both normal and abnormal speech.[20] The SLP and ENT may apply these same principles to joint evaluation of nasopharyngoscopy.

According to Strauss,[8] the SLP who intends to work in the CLP team should meet the following criteria:

- The CLP team has a SLP who attends team meetings and whose education, training, and experience have adequately prepared him/her for the diagnosis and treatment of patients with CLP.
- At least 1 SLP on the CP team provided speech therapy and/or a complete speech and language evaluation to a minimum of 10 patients with CLP in the past year.
- The CP team SLP performs a structured speech assessment during team evaluations.
- The CP team uses clinical speech instrumentation (such as endoscopy, aerodynamics, videofluoroscopy, and so on.) to assess VP function when indicated.

Additionally, Moller and Starr[14] suggested that personal characteristics such as interest in the patient and the disorder, willingness to share openly, mutual respect, participation as an equal partners, and motivation to learn were essential.

BARRIERS TO THE SPEECH LANGUAGE PATHOLOGIST IN THE CLEFT LIP AND PALATE TEAM IN JORDAN

- Most SLPs prefer to work in private clinics and schools than medical settings; therefore, no mutual interaction with medical practitioners may occur that may help in team building.
- The curricula of speech therapy programs do not have sufficient medical orientation.
- CLP teams are not well established in terms of the standards of work and prospective team members; in addition, they do not meet under 1 roof.
- SLP services are still in its infancy and barely known to other medical practitioners; this fact will affect the referral and consultations of SLPs.

RECOMMENDATIONS TO IMPROVE THE CURRENT PRACTICE PATTERN IN JORDAN

The following recommendations may enhance establishing CLP teams at the various medical sectors in Jordan:

- Inclusion of the basic knowledge and adequate training on CLP in the curriculum in academic programs provided for SLPs.
- Express the need of a trained SLP in CLP management to the speech committee at the MOH to focus on the area of CLP in the licensure examination.
- Recommendations to the minister of health, director of RMS, and the doctor's union to create new vacancies for medical SLPs in different medical settings.
- Recommendations to the directors of private hospitals to offer posts for SLP services to be represented as other medical disciplines.
- Empower the SLP services to be covered by health insurance companies.
- Creation of awareness via the media to spread the knowledge regarding CLP ideal management and stressing the SLP role among other practitioners.
- The SLPs should develop materials for information, education, and communication that may include simple sensitization and awareness materials for medical/allied and community level health care workers, preparation of books, audio/video material on step-by-step therapy procedures, and handouts for parents.
- The SLPs should plan part of their services for outreach programs and seek either paid or volunteer work with international CLP teams to have a chance to develop their expertise.
- The SLPs need to continue their effort hand-to-hand to establish the Speech Association in Jordan that will improve the concept of the team work for CLP.

CONCLUSIONS

The vocal tract serves both biologic and speech functions; when a given patient is born with such a CLP defect, it is assumed that this patient will experience feeding, speech, and cosmetic effects. Hence, a number of health practitioners who have specialized training and experience in the diagnosis and management of the designated defects being observed should be involved in the CLP team.[5] Review of the related literature that deals with the significance of team dynamics showed that the SLP is an essential CLP team member. The primary role of the SLP includes feeding and swallowing disorders, speech and language impairments, as well as critical decisions in the selection of the surgical, prosthetic, or behavioral management. The SLP also provides the interpretations of the VP images obtained by nasopharyngoscopy or videofluoroscopy. The SLP also provides patient and family counseling regarding coping with the resultant communication impairments associated with the CLP condition. Unfortunately, the CLP team concept is under established, efforts of health care professionals must be integrated to establish these teams at the level of major medical centers providing care for children with CLP.

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