Hepatocellular Carcinoma Metastatic to the Scalp

Charlotte M. Walter, BS, Emily J. Kirby, MD, Henry C. Vasconez, MD, Brian D. Rinker, MD

Abstract: A case of scalp metastasis from hepatocellular carcinoma (HCC) is reported that was initially diagnosed as a soft-tissue tumor. Attempted excision of the lesion resulted in an open wound requiring soft-tissue reconstruction of the scalp. Results of pathologic examination showed metastatic HCC. The patient returned postoperatively with bleeding, which was unable to be controlled, resulting in his death. Scalp metastases from HCC are very rare but must be considered when treating a patient with known cirrhosis, hepatitis, or HCC.

Key Words: Hepatocellular carcinoma, extrahepatic metastasis, cranial metastasis, massive bleeding

Hepatocellular carcinoma (HCC) is a common type of visceral carcinoma. It is the fifth most common cancer in the world and the third most common cause of cancer death worldwide.[1] Although not as prevalent in the United States, it remains the fastest-growing cause of cancer death in men.[1] With increasing rates of hepatitis C virus and cirrhosis in the United States, diagnoses of HCC are expected to increase correspondingly.

Metastatic lesions represent 1 type of extrahepatic manifestation of HCC that includes splenomegaly, gastrointestinal tract bleeding, ascites, pruritus, malaise, fatigue, weakness, pyrexia, anorexia, and nausea and vomiting.[2] The lung, bone, and lymph nodes are frequent sites of metastasis of HCC.[3] The most common sites for distant skin metastases of internal carcinomas are the chest, back, and abdomen.

Prolonged liver injury caused by chronic hepatitis C infection leads to fibrosis of the liver parenchyma and cirrhosis and predisposes the liver to HCC.[4] These changes cause a decrease in overall hepatocyte function including a decrease in clotting factors produced by the hepatocytes, leading to a coagulopathic state.[4] Hepatocellular carcinoma lesions tend to be hypervascular.[2] Because of the underlying coagulopathic state and the hypervascularity of the tumor, hemorrhagic events in HCC at both primary and metastatic sites occur frequently.

CLINICAL REPORT

A 58-year-old white man presented to a community general surgeon with a mass on his left forehead, above his eyebrow, which was thought to be a lipoma on physical examination. The patient had a known history of hepatitis C virus and cirrhosis. He was taken to the operating room by the general surgeon for excision of the mass. The surgeon noted the lesion to be grayish and rubbery, with extension to the outer table of the skull. The surgeon excised the soft-tissue mass in its entirety, but was unable to achieve primary closure. A pathology report identified the lesion as metastatic HCC. A bolster dressing was secured over the wound, and the patient was referred to plastic surgery for reconstruction of the defect. He was taken to the operating room, where he was noted to have a frontotemporal soft-tissue and calvarial defect approximately 8 × 5 cm with exposed dura and lytic lesions surrounding the calvarial defect. The open wound was reconstructed with a fasciocutaneous flap of the scalp and split-thickness skin grafting of the donor site. The patient was referred to oncology for a full metastatic workup. He met with a physician for initial consultation, but failed to keep any further appointments.

During staple removal in the plastic surgery clinic 3 weeks later, he began actively bleeding. Hemostasis could not be achieved in the clinic using manual pressure, silver nitrate, or handheld Bovie electrocautery, so he was taken emergently to the operating room. Neurosurgery was consulted because of the location of the metastatic tumor with possible involvement of the dura. Bleeding continued. Hemostasis could not be achieved intraoperatively using manual pressure, Bovie electrocautery, bipolar electrocautery, gelfoam, Surgicel, argon laser coagulation, or topical thrombin. Coagulation studies at this time revealed a platelet count of 149,000/μL, international normalized ratio of 1.5, prothrombin time of 14.6 seconds, activated partial thromboplastin time of 28 seconds, and a clottable fibrinogen of 234 mg/dL. Evaluation of bleeding time was not performed. He had a serum hemoglobin level of 8.6 g/dL and a hematocrit of 25.1%. Resuscitation of the patient continued intraoperatively with 4 U of packed red blood cells, 4 U of fresh frozen plasma, and 1 U of cryoprecipitate. He also received 4500 mL of crystalloid during this operation. Recombinant activated factor VII (NovoSeven; Novo Nordisk Pharmaceuticals, Princeton, NJ) was given in 2 doses of 3 mg each. The decision was made by the neurosurgery and plastic surgery teams to temporize the wound overnight while resuscitation continued. A pressure dressing was placed on the wound, and resuscitation continued overnight. The following morning, the patient had an international normalized ratio of 1.2, activated partial thromboplastin time of 29 seconds, fibrinogen level 202 mg/dL, and platelet count 105,000/μL. The patient was taken back to the operating room by neurosurgery and plastic surgery. Neurosurgery removed the remainder of the hypervascular tumor in attempts to stop the hemorrhage, but the patient continued to bleed. Intraoperatively, the patient received one 6-pack of platelets, 7 U of leukocyte-reduced packed red blood cells, and 4 U of fresh frozen plasma. Estimated blood loss during the procedure was 6 L. He also received 1000 mL of 5% albumin, 50 mL of 25% albumin, and 4800 mL normal saline. The wound was closed by plastic surgery, and a pressure dressing was placed. The wound continued to bleed overnight. He was weaned from the ventilator postoperatively and met criteria for extubation. Because of the metastatic nature of the patient's disease, the palliative care service was consulted, and the patient and family elected to proceed with palliative care only.

DISCUSSION

Hepatocellular carcinoma is a common cancer. Worldwide, it is the fastest-growing cause of cancer death in men.[1] In North America, the incidence is lower than in other parts of the world at less than 5.0/100,000; however, it has been rising because of an increasing prevalence of hepatitis C virus. Hepatocellular carcinoma occurs more frequently in men than in women, with a 2:1 to 4:1 ratio.[1]

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Extrahepatic metastases from HCC are associated with a poor prognosis, with a mean survival of 7 months and a 1-year survival rate of 24.9%.[3] In 0.8% of patients with a form of internal carcinoma, cutaneous metastasis is the presenting sign. Four percent of cutaneous metastatic lesions occur on the scalp, and 6% on the face.[5] Cutaneous metastases of HCC may appear as rapidly growing nodules on the scalp, chest, or shoulder. They may be single or multiple, firm, painless, nonulcerative, and reddish blue nodules, typically 1 to 2.5 cm.[6] They may present similarly to basal cell carcinoma.

The hemorrhagic nature of metastatic HCC has been widely reported. Metastatic lesions to the skull have been associated with spontaneous epidural hemorrhages.[7] Spontaneous rupture of pleural metastases of HCC has been shown to cause massive hemorrhage, leading to hemothorax.[8] Kamiyoshihara and colleagues[9] reported a case of massive bleeding of biopsied rib tumor that was shown to be metastatic HCC; hemostasis of the biopsy site was achieved only by complete excision of the diseased rib. Cases of postoperative hemorrhage have also been described, including a case of massive hemorrhage after biopsy of a metastatic lesion to the mandible.[10]

The hemorrhagic quality of a metastatic HCC tumor is thought to be due to 2 distinct processes. First, patients with HCC and cirrhosis have declining liver function that affects the regulation of hemostasis on many levels.[11] The fibrotic changes associated with cirrhosis cause a decline in the synthetic function of the liver including a decline in production of coagulation factors such as factors V, VII, IX, X, and XI, as well as prothrombin and anticoagulation proteins such as protein C, protein S, and antithrombin.[4,12] The decreased production of coagulation factors and anticoagulation proteins disturbs the delicate balance of hemostasis and can lead to both hypercoagulable and coagulopathic states.[12] Declining liver function also affects hemostasis by causing a rise in nitric oxide, leading to decreased vascular tone.[13] There is thrombocytopenia due to increased splenic sequestration from splenomegaly. Declining renal function usually accompanies advancing liver failure, causing acquired platelet dysfunction.[12] These pathologic changes can tip the balance to cause a bleeding diathesis.[12]

In addition to the changes in liver function affecting the coagulation cascade, characteristics of the metastatic HCC tumor itself contribute to abnormal hemostasis. Hepatocellular carcinoma and its metastases are typically vascular in nature, with 1 study finding 89.2% of the tumors to be hypervascular. When a metastatic lesion becomes hemorrhagic, it becomes increasingly difficult to attain hemostasis. When standard options fail to control the hemorrhage, radiotherapy has been used to successfully stop the bleeding in a number of cases.[10,14,15]

Although cutaneous metastatic HCC is rare, it should be considered when evaluating a skin lesion in a patient with known cirrhosis or hepatitis. The hemorrhagic nature of metastatic HCC should be appreciated, particularly before any attempted manipulation or resection of the metastatic lesion.

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Unusual Odontogenic Keratocyst of the Maxillary Sinus

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Abstract: An odontogenic keratocyst that eroded into the sinus through the maxillary bone and occupied it, showed replacement of the sinus respiratory epithelium by lesional epithelium, and was associated with fungal rhinosinusitis is presented. A review of the literature disclosed that epithelial replacement has been described in 2 previous case reports, although there is no report on the coexistence of odontogenic keratocyst with fungal rhinosinusitis.

Key Words: Odontogenic keratocyst, maxilla, fungal rhinosinusitis

Odontogenic keratocyst (OKC) is established as a distinct entity because of its specific microscopic features and aggressive behavior that manifests with infiltration of adjacent anatomic

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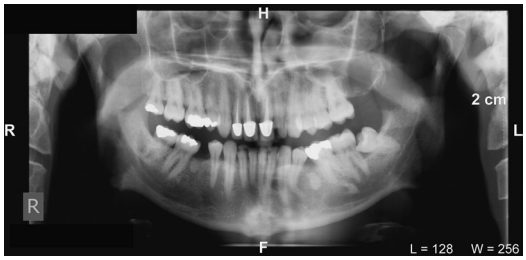

FIGURE 1. Panoramic radiograph shows well-circumscribed radiolucent lesion apical to the first molar tooth.

structures and high recurrence rate. This behavior is emphasized by the introduction of the term *keratocystic odontogenic tumor* in the recent classification of the World Health Organization,[1] although its neoplastic nature is debated. We describe an OKC that eroded into the sinus through the maxillary bone and replaced the sinus respiratory epithelium, while it was associated with fungal rhinosinusitis.

CLINICAL REPORT

A 38-year-old man presented with intermittent pain, swelling, and a feeling of "fullness and pressure" on the left side of his face that he had first noticed about 1 year ago and got worse during scuba diving. His medical history was significant for chronic rhinosinusitis that had repeatedly been treated with administration of antibiotics and local corticosteroids. He was otherwise healthy and not in any other kind of medication.

Clinical examination revealed a hard, nonfluctuant, and slightly tender swelling covered by normal mucosa in the maxillary vestibule, distant to the first molar tooth. Regional lymph nodes were not palpable. According to the referring dentist, the associated teeth were vital. Panoramic radiograph showed a well-circumscribed radiolucent lesion apical to the first molar tooth (Fig. 1).

A provisional diagnosis of OKC was made, and an incisional biopsy was performed under local anesthesia. Intraoperatively, the cystic cavity was found to contain a whitish material. Microscopic examination of 5-μm-thick, formalin-fixed, and paraffin-embedded tissue sections showed connective tissue lined by a uniformly thin parakeratinized epithelium without rete ridges that had a corrugated parakeratinized surface and a basal layer composed of columnar cells with reverse nuclear polarity (Fig. 2). Focal separation of the epithelium from the connective tissue was seen. The connective tissue was mildly infiltrated by inflammatory cells, predominantly lymphoplasmacytes. The diagnosis was OKC.

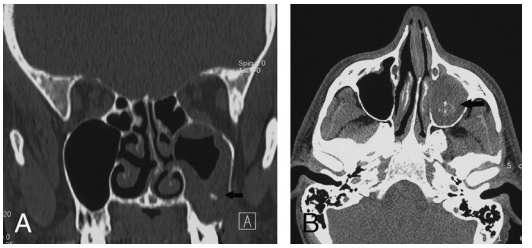

FIGURE 3. A, Computed tomography scan, coronal view, reveals diffuse radiopacity mainly toward the floor of the left maxillary sinus, bone resorption at the anterolateral wall of the maxilla close to the alveolar process, and microcalcifications (arrows). B, Computed tomography scan, sagittal view, reveals diffuse radiopacity mainly toward the floor of the left maxillary sinus and microcalcifications (arrows).

A computed tomography scan revealed a diffuse radiopacity mainly toward the floor of the left maxillary sinus. Bone resorption was evident at the anterolateral wall of the maxilla close to the alveolar process (Fig. 3). Radiopacity of the sinus with microcalcifications or "metallic dense" spots were interpreted as "consistent with aspergillomas."

The cyst was enucleated, the sinus cleaned through a Caldwell-Luc approach, and a drain was placed through a rhinoantrostomy. The patient was administered amoxicillin 500 mg plus clavulanic acid 125 mg, 3 times daily, for 5 days. The drain was removed after 72 hours; his postoperative recovery was uneventful, and 19 months after operation, he is free of disease.

Macroscopically, the cyst measured about 3.5 × 2 × 1 cm. Microscopic examination confirmed the diagnosis of OKC. Focally, the epithelium showed increased cellularity and drop-shaped rete ridges, whereas the cells in the basal third had large and hyperchromatic nuclei (Fig. 4). Mitoses were not identified. In addition, there was a focus of abrupt transition of the cystic epithelium to respiratory pseudostratified columnar epithelium that gave the impression of active replacement or "pushing" (Fig. 5). The connective tissue of the cystic wall was vascular, edematous, with foci of calcifications, cholesterol crystals, and mild lymphoplasmacytic infiltration, whereas no granulomatous

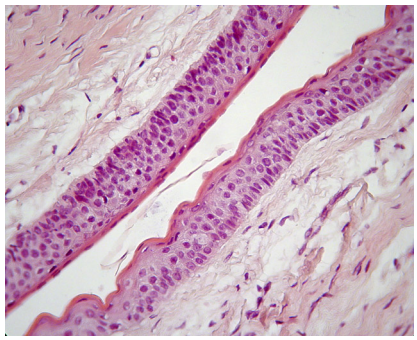

FIGURE 2. Connective tissue lined by a uniformly thin parakeratinized epithelium without rete ridges. Notice the corrugated parakeratinized surface and a basal layer composed of columnar cells with reverse nuclear polarity (hematoxylin-eosin stain, original magnification ×400).

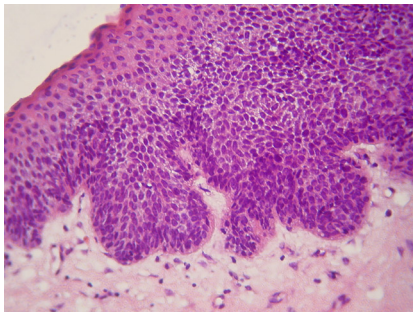

FIGURE 4. Odontogenic keratocyst epithelium shows increased cellularity, drop-shaped rete ridges, and cells with hyperchromatic nuclei in the basal compartment (hematoxylin-eosin stain, original magnification ×400).

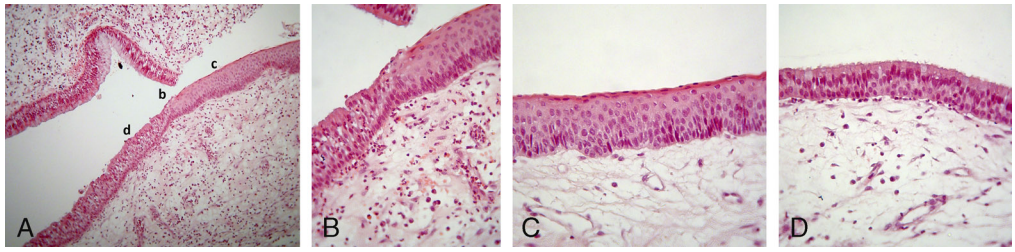

FIGURE 5. Abrupt transition of OKC epithelium to respiratory epithelium: (A) low-power view, (B) area of penetration, (C) OKC epithelium, (D) respiratory epithelium (hematoxylin-eosin stain, original magnification ×100 [A], ×400 [B–D]).

reaction, fibrosis, or necrotic foci were evident. Periodic acid-Schiff stain revealed fungal hyphae and spores in the mucosal connective tissue, close to but not infiltrating vessels (Fig. 6). Final diagnosis was OKC with yeast.

A complete blood count was not suggestive of hematologic disease or diabetes mellitus, and an HIV test was negative.

DISCUSSION

Sinus involvement by OKCs is estimated to occur in less than 1% of the cases,[2] but replacement of respiratory epithelium with OKC epithelium is unusual. Yamazaki et al[3] described a case of an OKC in a 39-year-old woman that involved an impacted maxillary premolar, expanded to the maxillary sinus, and finally reached the inferior nasal meatus. Replacement of the OKC lining by respiratory epithelium was attributed to the proximity of the cyst to the nasal cavity. Abrupt change of OKC lining to respiratory epithelium, as seen in our patient, was reported by Vencio et al[4] in a 27-year-old woman. The cyst was associated with an impacted right second maxillary molar and also invaded the sinus floor.

This microscopic finding is reminiscent of the extension of intraepithelial neoplasia of the oral cavity[5,6] and other carcinomas[7] along adjacent ductal epithelium basement membrane. "Ductal or glandular involvement" is considered an important pathway of spread of carcinomas. In our case, we assume that the OKC epithelium "crept" on the sinus wall replacing the normal respiratory epithelium and extended to involve most of the antral cavity. Thus, we agree with Vencio et al[4] in that replacement is consistent with the infiltrative behavior of the OKC reflected in its classification as a neoplasm. Dysplastic features, as seen in the present case, are occasionally described in OKCs, but have not been associated with an aggressive growth.

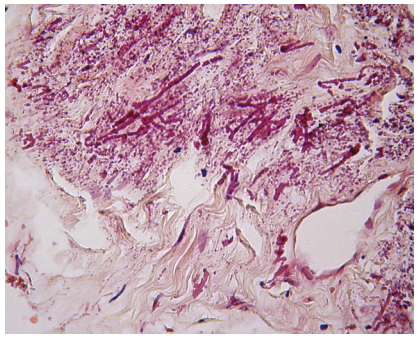

FIGURE 6. Fungal hyphae and spores in the mucosal connective tissue wall (periodic acid-Schiff stain, original magnification ×400).

Our patient had a history of chronic rhinosinusitis that is the most common form of rhinosinusitis, estimated to affect about 20% of the population.[8] Radiological evidence of sinus opacification with calcifications was consistent with fungus balls or aspergillomas, defined as noninvasive accumulations of dense conglomeration of fungal hyphae in 1 sinus cavity. Histological evidence, however, of spores and hyphae in the mucosal connective tissue is diagnostic of an invasive fungal rhinosinusitis, in particular chronic invasive fungal rhinosinusitis. This form is usually associated with *Aspergillus fumigatus* and may appear in patients under corticosteroid treatment who are subtly immunocompromised.

No other case of OKC associated with fungal rhinosinusitis and fungus balls was found in the literature. In our patient, the root canal therapies in the central incisors and right lateral incisor were not close to the sinus; thus, a role for the heavy metals from root filling materials, in particular, zinc from zinc oxide–containing materials, do not seem probable. The blockage of the ostium by the OKC, however, may have resulted in the obstruction of sinus ventilation and the creation of anaerobic conditions that allowed germination of fungi, which probably became invasive because of the local immunosuppression caused by the prolonged corticosteroid treatment.[9,10]

As our patient was proved to be immunocompetent, the complete surgical removal of the cyst and the sinus drainage and aeration achieved with the rhinoantrostomy were considered curative, both for the OKC and the fungal rhinosinusitis.[11]

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Complex Midfacial Reconstruction With an Implant-Supported Framework

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Abstract: This clinical report describes the treatment of a patient with osseointegrated extraoral implants supporting a framework retainer and acrylic resin mesostructures and a large silicone midfacial prosthesis. A metal framework was used to splint the implants together and provided satisfactory retention for the facial prosthesis. A 2-piece prosthesis that composed of an obturator and facial prosthesis was fabricated. Cosmetic improvements as well as the ability to speak, swallow, and, to a lesser degree, chew, were achieved for this patient.

Key Words: Extraoral implant, facial prosthesis, obturator, facial defect, framework

Patients with extensive maxillofacial defects are confronted with functional limitations of vision, speech, mastication, deglutition, and, perhaps most devastating, the psychologic impact that such defects have on the quality of life.[1] The goals of prosthetic rehabilitation for these patients include separation of oral and nasal cavities to allow adequate deglutition and articulation, possible support of the orbital contents to prevent enophthalmos and diplopia, and support the soft tissue to restore the midfacial contour and an acceptable aesthetic result.[2] Adequate retention and acceptable cosmetic results are required for successful prosthetic rehabilitation of a patient with a large facial defect. Osseointegrated implants can provide support and retention using the remaining bones.[3–8] In midfacial and oculofacial defects, fabrication of the retention component is often complicated by the orientation of the abutments, the interabutment distance, and the remaining anatomic structures.[9] Considerable attention has been given to framework design for the extraoral implant–supported prostheses[8,9]; however, reports on the attachment design of the larger combination maxillofacial prostheses are few.

This clinical report describes the treatment of a patient with osseointegrated implants supporting a framework retainer acrylic resin mesostructures and a large silicone facial prosthesis.

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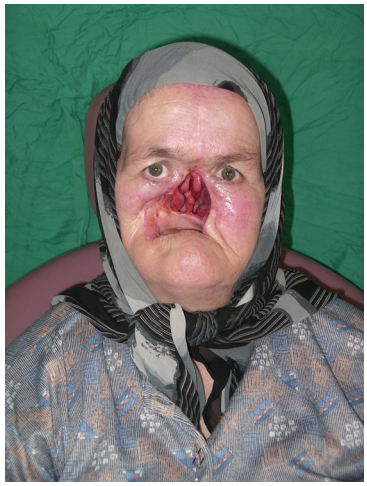

FIGURE 1. Initial facial appearance of the patient.

CLINICAL REPORT

A 55-year-old woman was referred to our clinic for the rehabilitation of a large midfacial defect. History revealed that the patient had been operated on for basal cell carcinoma that occurred in her nasal skin in 1994. During the next 11 years, she had underwent 2 operations for the penetrating basal cell carcinoma. The residual defect included total loss of the midface bilaterally (Fig. 1). The upper lip, alveolar bone, and hard palate were totally gone except for a small fragment of the tuberosity on the left side. Also, right oral commissure, nose to frontal bone, and parts of the cheeks bilaterally were absent. There was a significant impairment of deglutition and speech, which exhibited on articulation resonance disorder with moderate hypernasality. The patient was able to swallow liquids and pureed solids. Radiotherapy was performed, and the patient received 6000 cGy of external beam irradiation in 30 sessions for 6 weeks. Hyperbaric oxygen treatment was not applied.

After 2 years from radiotherapy, patient came to our clinic for treatment. She was examined to place extraoral implants with

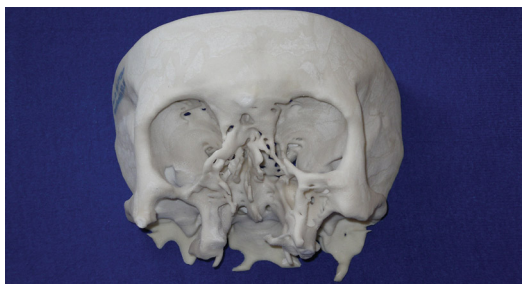

FIGURE 2. Bone model of patient.

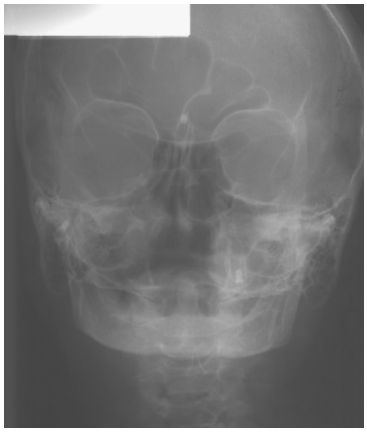

FIGURE 3. Radiographic image of the patient after placement of the implants.

computed tomographic (CT) image and Mimics 10.0 software (Materialise, Leuven, Belgium). Z-Printer 510 (Z Corporation) was used to form bone modeling (Fig. 2). According to the results of the analysis, three 5-mm self-tapping Straumann extraoral implants were placed, 1 in the glabellar region, 1 in the right zygomatic arch, and 1 in the left tuberosity intraorally under local anesthesia (Fig. 3). After 8 weeks of healing period, stage 2 surgery was performed to expose the implants. Then conical abutments were connected to the implants, and implant sites were allowed to heal approximately 2 weeks before impressions were made. Impression cylinders were placed on the abutments, and impression was made with a polyvinyl siloxane putty (Elite HD Zhermack, Bodia Polesine, Italy) and regular body (Elite HD Zhermack). Then impression cylinders were connected to conical implant analogs and placed into impression. A master cast was obtained. A rigid framework was fabricated to splint the implants together on the master cast. Dolder bar attachments were placed on to rigid framework (Fig. 4). Acrylic resin templates were attached with dolder bar matrix over the dolder bar attachment (patrix) on the framework. One template separated the nasal cavity from the oral cavity, and this part also provided the upper lip, and the other was used to support the silicon prosthesis (Fig. 5). The wax contours of the facial prosthesis were produced with the aid of a presurgical photograph of the patient. The wax pattern was evaluated on the patient for aesthetics and marginal adaptation. The wax facial prosthesis was invested and cast with silicone material (Platinum Slicone Elastomer; Factor II, Inc, Lakeside, AZ) that had a base pigment shade for intrinsic coloration. Platinum Primer A-306 (Factor II, Inc) was used for bonding silicone elastomer to acrylic resins. Extrensec Coloring Kit (Factor II, Inc) was used for external

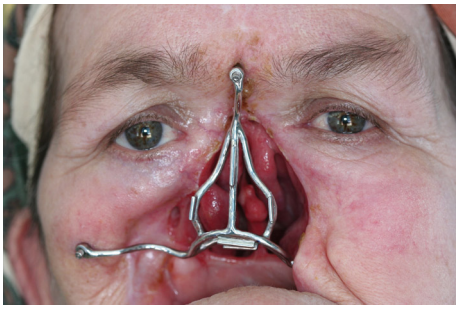

FIGURE 4. The metal framework.

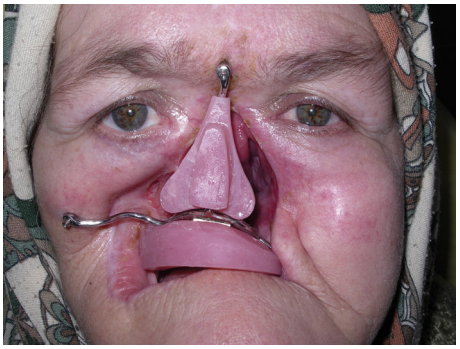

FIGURE 5. Acrylic resin templates.

coloration of facial prosthesis to match the patient's skin (Fig. 6). Conventional removable complete denture was made for edentulous mandibular arc.

DISCUSSION

Endosseous implant-supported prostheses are reported for midfacial defects.[1,3,6,8,9] Use of endosseous implants may be limited by the lack of viable bony structures. Sample size, treatment technique, dose, type, and fractionation scheme of irradiation, duration of follow-up, patient factors (eg, site and size of the tumor and general health status), and time interval between radiotherapy and implant placement, all of which may affect implant survival.[10]

Identifying an adequate residual bone bed for endosseous implant placement may be difficult for patients with bilateral maxillary resection defects.[6] A computed tomographic and computer-aided bone model is also used for customized implant preparation to reduce surgery time and risks.

In this case, for the stability of a facial prosthesis, 3 implants were placed in the glabellar region, the right zygomatic region, and the left tuberosity, which were the only suitable bony structures for placing these 3 extraoral implants. The preparation of the metal framework was difficult because of the implant angulations and anatomic structures. Although 1 of the 2 acrylic resin templates was supporting the silicone prosthesis, the other was used only to separate the nasal cavity from oral cavity as an obturator, and this part provided additional durability to prosthesis during nutrition.

A dentate prosthesis was not attached to this obturator. The stability of the metal framework supporting only 3 extraoral implants may not be enough to bear occlusal forces.

The prosthesis processed in the patient with facial defect; support the patient psychologically owing to the aesthetic and function it provides. It also affects the social activity.

The resulting restoration was retentive, provided acceptable appearance, and was well tolerated by the patient. The patient is under periodical follow-up medically for 3 years for any recurrence and the success of the prosthesis. Silicone prosthesis was remade during that time, and any failure of implants was observed.

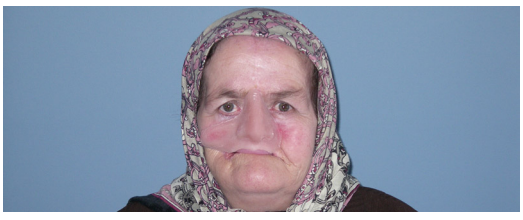

FIGURE 6. Silicone facial prosthesis.

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Salival Fistulae: A Rare Complication After Microtic Ear Reconstruction

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Abstract: Parotid fistulae are a very infrequent complication after ear reconstruction in microtia when the local conditions are favorable. We report 2 cases of salival fistulae after microtic ear reconstruction. Timing of the diagnosis is important to decide the treatment. We recommend conservative management by restricted citric diet in early postoperative salival fistulae and Botox injections in the case that it persists.

Key Words: Salival fistulae, microtia, ear reconstruction

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Microtic ear reconstruction is a complex procedure, a challenge even for experienced plastic surgeons, and it is not free of complications. The most frequent complications are related with flap viability and cartilaginous framework exposure. Salival fistulae are an infrequent complication with few reports in the literature. We report 2 cases and their management.

CLINICAL REPORTS

Patient 1

A 7-year-old girl, with hemifacial microsomia and lobular-type microtia, was presented for ear reconstruction. After surgery, she presented wound infection that was treated with surgical debridement and antibiotics. Four years later, a second stage for conchae reconstruction was performed. At the second postoperative month, salival fistulae, with saliva fluid draining from the conchae were observed (Fig. 1). Fistulography confirms the diagnosis (Fig. 2). Conservative treatment by restricted diet failed. The patient persisted with the salival fistulae at 1 year postoperative, so we decided to use botulinum toxin (Botox; Allergan Pharmaceuticals, Westport, Ireland) injection for gland defunctionalization. Under ultrasonic vision, 100 g of botulinum toxin was injected in the parotid gland. After 3 sessions of Botox injection, symptoms stopped, and an atrophic parotid gland was observed by ultrasound.

Patient 2

An 11-year-old patient with isolated lobular microtia underwent ear reconstruction. At postoperative period, wound infection with partial cartilage exposure was treated by debridement and antibiotics. Two years later, conchae reconstruction was performed. At the first postoperative month, fluid drained through the conchae, appearing in relation to feeding. As soon as the salival fistula was confirmed, restricted citric diet was started as an early conservative treatment. We observed progressive decrease of the fluid with complete resolution of symptoms at 2 months' follow-up.

DISCUSSION

Parotid fistulae are widely reported as a complication after trauma, rhytidectomy, and parotidectomy. The only report[1] related to microtic ear reconstruction was a patient with failed primary reconstruction, who developed a salival fistula after the second stage of new reconstructive attempt, confirmed with sialography. The authors describe spontaneous resolution of the fistulae, without reference to the specific treatment, only the fact that the patient did not require surgery. In our experience, both cases were related with

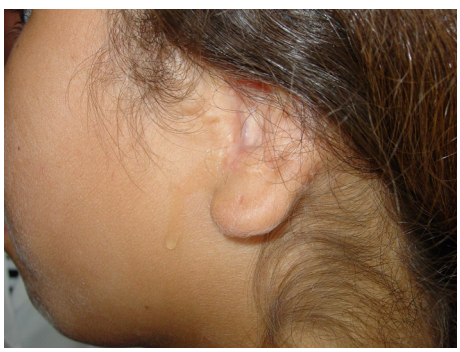

FIGURE 1. Reconstructed ear and salival fistulae.

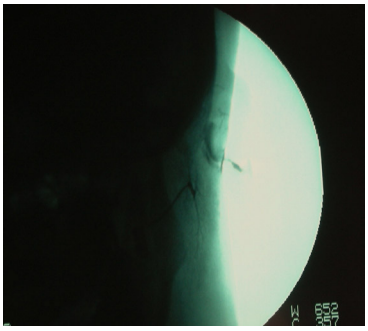

FIGURE 2. Fistulography shows salivary fluid through the reconstructed ear.

a history of inflammatory events in the postoperative ear reconstruction period. We consider that anomalous wound healing produces parotid gland traction next to the reconstructed ear, increasing the risk of parotid gland injury. In both patients, the second stage consisted conchae reconstruction with manipulation of the soft tissues adjacent to the gland.

The management of parotid fistulae uses the same approach independent of the lesion etiology. This is based on salival secretion decrease to prevent tissue lysis by salival enzymes, which perpetuates the fistulae. Traditionally, treatments are fasting with parenteral nutrition, drainage, dressing, surgical procedures, and/or local radiotherapy. However, it is well accepted that most salival fistulae have spontaneous resolution, and the main treatment is an early restricted citric diet. When associated to a sialocele, frequently in posttraumatic fistulae, suction drainage and compressive dressing appear to be the best solution.[2,3] When fistulae persist, fistulectomy is the alternative, unfortunately with high rate of recurrence and facial scars.

Alternative treatment suggested chemical defunctionalization of the parotid gland using somatostatin analogs or anticholinergic drugs (atropine). The disadvantage of this therapy is the daily injection, as well as their systemic effects.[4] Recent articles report the use of local injections of Botox as an alternative for chemical denervation, blocking acetylcholine release to reduce salivary flow.[5] Effective response has been reported (4 resolved fistulae with 1 injection, 2 with 2 injections, and 1 after 3 injections),[6–8] with a simple, safe, and reproducible technique, avoiding surgical treatment that could worsen the aesthetic result of the microtic ear reconstruction.

In the first case described, the patient did not respond to conservative diet restricting treatment, presenting persistent fistulae. The surgical option would have changed the aesthetic results of the ear reconstruction, and so treatments with local botulinum toxin injections were indicated. In the second case, the salival fistulae complication appeared early in the postoperative period (first month). The experience of the previous case permitted an early diagnosis and treatment initiation. The patient had an excellent response to the conservative restricted citric diet therapy.

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Isolated Hemifacial Hyperplasia

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Abstract: Hemifacial hyperplasia, formerly termed hemifacial hypertrophy, is a rare condition, which involves both bone development and soft-tissue enlargement. We present 2 pediatric cases of hemifacial hyperplasia. Soft-tissue reconstruction was applied for the severe case.

Key Words: Hemifacial hyperplasia, child, soft-tissue reconstruction

Hemifacial hyperplasia, first described by Beck in 1836, is a rare congenital developmental disorder characterized by unilateral enlargement of facial soft and hard tissues (bone, teeth, adipose

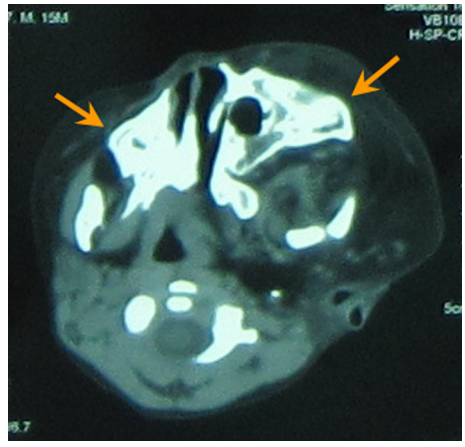

FIGURE 1. Axial computed tomography scan at 15 months of age (preoperatively), showing overgrowth of the left maxilla (arrows) and soft tissue.

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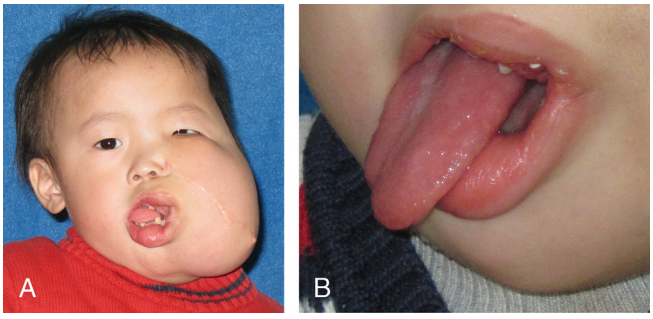

FIGURE 2. Frontal view shows remarkable facial asymmetry before the second operation (A) and hemihyperplasia of the tongue, which is deviated to the unaffected side (B).

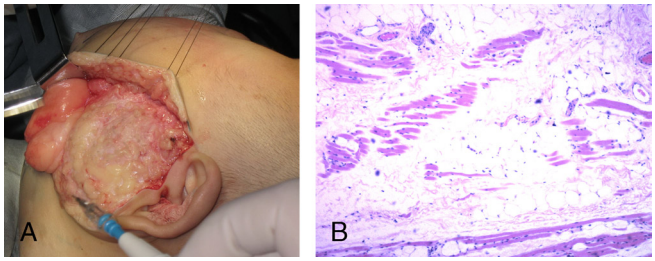

FIGURE 4. Intraoperative view shows diffuse lipomatosis and an enlarged buccal pad with intact capsule (A). Histopathologic examination shows fatty infiltration in muscle without hypertrophy of the adipocyte or muscle cell (B).

tissue, muscle, vessel, nerve, lip, gingival, mucous membrane, auricle). Sometimes, not all of the structures are involved. It is assumed to be a minor form of hemihypertrophy. The asymmetry is usually noticed at birth and grows gradually until puberty. The enlarged side grows at a rate proportional to the unaffected side.

Surgical treatment has a limited role, mainly because of postoperative recurrence, extensive deformities, and difficulty with airway management. This report presents 2 cases of hemifacial hyperplasia and discusses its features and surgical treatments.

CLINICAL REPORTS

Patient 1

A 2-year-old otherwise healthy boy presented to our department with marked left-sided facial asymmetry. He was the second child of young, healthy parents. The family history was unremarkable. Pregnancy and birth were uneventful. Facial asymmetry was first noticed at birth by his parents. The left side of the face grew slightly faster than the right side of the face. His physical and mental development were normal.

Fifteen months after birth, computed tomography scan revealed enlargement of the left zygomatic bone, maxilla, mandible, masseter muscle, and parotid gland. Diffuse subcutaneous adipose tissue was also demonstrated on the left side of the face (Fig. 1). Partial correction of the lips and perioral region was accomplished by dermolipectomy and mucosectomy in a dental hospital through intraoral and nasolabial fold incision. Postoperative pathology showed lipoma.

On physical examination, gross asymmetry of the left face and apparent nasolabial fold scar was noted (Fig. 2A). The left side of the face was severely enlarged, including the mandible, maxilla, zygoma, and upper and lower lips. There was a narrow palpebral fissure on the left side. The nose and chin were deviated to the right. Both auricles were within normal limits and seemed to be symmetrical. Facial expression was unchanged, possibly covered by adipose tissue.

On intraoral examination, severe enlargement of the left side of the tongue was noted, which was deviated to the right side (Fig. 2B). Alveolus in the premaxillary region was enlarged significantly with the midline deviated to the unaffected side. The teeth of the left side were larger in size and shape. The left gingival and buccal mucosa was thickened and enlarged.

Magnetic resonance imaging (MRI) revealed enlargement of subcutaneous fat, which penetrated into the left masseter, parotid, parapharyngeal space, and temporalis. Apparent augmentation of other structures was detected, including left zygomatic bone, maxilla, mandible, masseter muscle, parotid gland, and submandibular gland (Figs. 3A–C). The volume of the maxillary sinus was reduced, partially because of the thickened mucous membrane. Suspected mastoiditis was found without symptoms.

Given the fact that the patient's deformity was extensive and there was considerable time for physical and mental development, a decision was made to carry out liposuction and lipectomy. The premarked subcutaneous facial fat was removed with tumescent liposuction. Using a previous facial scar, a 2.5-mm cannula attached to a vacuum device was inserted and traveled beneath the skin, encountering relative greater resistance. A total of 150-mL subcutaneous fat was removed. Through a preauricular and submandibular approach, a large cheek flap was elevated in the subcutaneous plane. No distinct boundary of subcutaneous fat mass was detected, apart from an enlarged buccal pad with intact capsule (Fig. 4A). Under direct vision, the parotid duct and the main branches of the facial nerve were preserved. To our surprise, the distal facial nerve was underneath the fat tissue and not ending at extremely atrophic facial

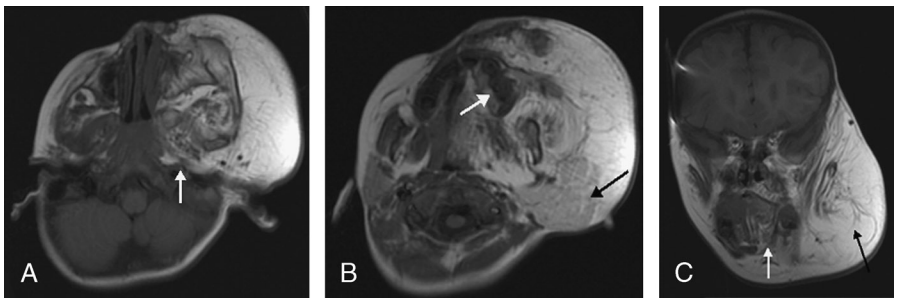

FIGURE 3. Axial MRI shows diffuse lipomatosis (A) and enlargement of left alveolus, palate, and parotid (B). Coronal MRI shows overgrowth of cutaneous adipose tissue and the left side of the tongue (white and black arrows) (C).

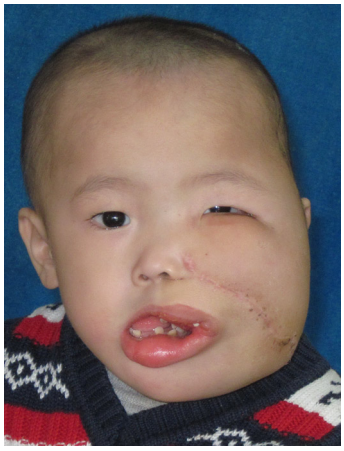

FIGURE 5. Postoperative frontal view after the second surgical treatment.

expression muscles. Thus, there was no need to preserve the distal facial nerve and facial expression muscles. An initial nasolabial fold incision was made to expose the premaxilla region. Facial fibrofatty tissue was excised, and histopathologic examination revealed fatty infiltration (Fig. 4B). A pedicled fibrofatty flap of the infraorbital region was applied to suspend the angle of mouth. Excessive skin was excised, the wound sutured in layers, and a drainage tube was placed and removed 5 days postoperatively to avoid a parotid duct fistula. The postoperative period was uneventful, and a relatively acceptable facial contour was achieved (Fig. 5).

Patient 2

A 4-year-old otherwise healthy girl presented to our department with moderate facial asymmetry. Her mother had a history of cold medicine ingestion and exposure to hair dyes during pregnancy. The child was born at full term by spontaneous vaginal delivery. The family history was negative. Facial asymmetry was first noticed 1 month after birth by her parents. For the past years, the right side of the face had been proportional to the unaffected side. Her mental development had been normal, except for a suspected hyperactivity disorder.

On physical examination, moderate enlargement of the right face was noted, including the mandible, maxilla, zygoma, and lower lip (Fig. 6A). The right auricle, with a lighter pigmentation (Fig. 6B), was slightly larger than the unaffected side. The nose and chin were slightly deviated toward to the left. A lightly pigmented lesion was noticed under the earlobe. The facial expression was normal.

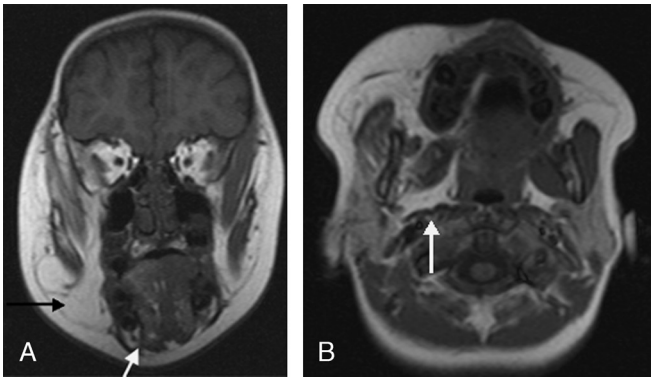

FIGURE 7. Coronal MRI shows overgrowth of cutaneous adipose tissue and right side of the tongue (black and white arrows) (A). Axial MRI shows fatty infiltration with affection of right parapharyngeal space (arrow) (B).

On intraoral examination, slight enlargement of the right side of the tongue to the midline was noted, which was deviated to the left side (Fig. 6C). The alveolus in the premaxillary region was almost symmetrical. The teeth were normal in size and shape. The left buccal mucosa was thickened and enlarged.

Magnetic resonance imaging revealed enlargement of subcutaneous fat, which penetrated into the left masseter, parotid, and parapharyngeal space. Moderate augmentation of other structures was detected, including the left zygomatic bone, maxilla, mandible, masseter muscle, and parotid gland (Figs. 7A, B). The volume of the maxillary sinus was also reduced slightly. The child is followed up regularly. An operation will be considered when growth ceases.

DISCUSSION

Pollock et al[1] suggested the word *hyperplasia* rather than hypertrophy, because the enlargement resulted from an increased number of cells rather than an increase in cell size.[2,3] Rowe[2] classified congenital hemihypertrophy as follows:

1. Complex hemihypertrophy, involving an entire half of the body, or at least an extremity;
2. Simple hemihypertrophy, involving a single limb; and
3. True hemifacial hypertrophy, unilateral enlargement of all tissues, including the ear (not including the eye); partial hemifacial hypertrophy, not all tissues are enlarged.

The etiology of hemifacial hyperplasia remains unclear, although various hypotheses have been suggested. The disorder has been attributed to endocrine diseases, lymphatic disorders, asymmetric

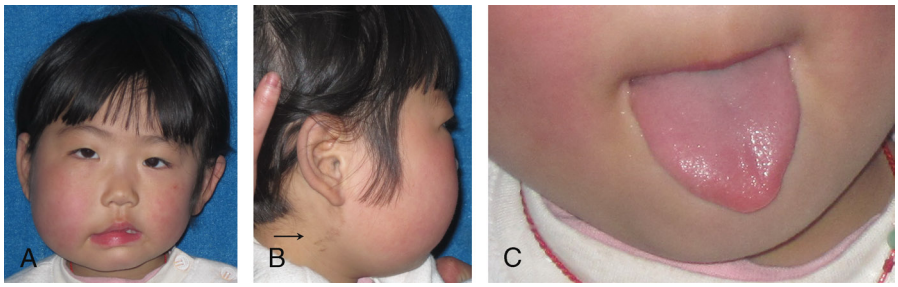

FIGURE 6. Frontal view shows moderate facial asymmetry with normal facial expression (A). The enlarged auricle and skin pigmentation (arrow) on the right side (arrow) (B). Hemihyperplasia of the tongue, which is deviated to the unaffected side (C).

development of the neural fold and hyperplasia of the neural crest cells,[1] and affection of first branchial arch during embryonic development.[3] Based on Staffenberg work, Lee et al[4] reported 3 cases of partial hemifacial hyperplasia and suggested the term of *hemifacial myohyperplasia*. Because all of the involved muscles belong to the first and second branchial arches, this disease has been attributed to disturbance of branchial arch formation.

The diagnosis should be made by a clinical geneticist experienced in the differentiation of other partial hypertrophies, including Beckwith-Wiedemann syndrome,[5] Silver-Russell syndrome, Proteus syndrome, CLOVE syndrome (newly delineated syndrome of congenital lipomatous overgrowth, vascular malformations, and epidermal nevi),[6] neurofibromatosis type 1, Klippel-Trenaunay syndrome, and lymphaticovenous malformation. Bou-Haidar et al[7] reported a case similar to our patient 2, which was manifested almost exclusively as lipomatosis.

Hemihyperplasia is assumed to be a minor form of hyperplasia. In a 10-year prospective multicenter clinical study of 168 isolated hemihyperplasia children, 10 tumors developed in 9 patients, with an incidence of 5.9%, including 6 Wilms tumors, 2 andrenal cell carcimoma, 1 hepatoblastoma, and 1 small bowel leiomyosarcom.[8] One hundred thirty-four malignancies in association with isolated hemihyperplasia were reviewed by Lapunzina,[9] drawing a similar conclusion. Thus, regular tumor surveillance was suggested, such as abdominal examination, abdominal ultrasound, and serum alpha-fetoprotein measurement.[10] However, tumors associated with isolated hemifacial hyperplasia have not been reported to our knowledge.

Reconstructive procedures, such as orthognathic surgery, osteotomy of the mandible and maxilla, soft-tissue excision, cheiloplasty, are mainly used when growth ceases.[11–15] However, a patient's needs should also be considered.[1] In our case, soft-tissue debulking was performed for the 2-year-old child to obtain a relatively acceptable facial contour and prevent social stigma.[16] Facial liposuction, with less trauma, is an effective liposuction procedure for facial contouring, especially the temporal region. Subcutaneous fibrofatty tissue was largely resected subsequently, but the enlarged masseter and parotid were preserved, which influenced the operative result. Because the facial expression muscles did not function well, a compromised static suspension was accomplished by pedicled fibrofatty flap of the infraorbital region. To our knowledge, treatment of such an infantile case with severe hemifacial hyperplasia has not been reported. In addition, these combined procedures, especially the combination of liposuction, were also unique for treatment of this rare disorder. When growth ceases, secondary or tertiary procedures may be required,[1,13] including soft-tissue excision, intramuscular injection of botulinum toxin, cheiloplasty, and remodeling of the mandible and maxilla. Function of the temporomandibular joint should be followed by a dentist. Orthognathic surgeries may be required to adjust the occluding relation.

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Patients' Preoperative Expectations and Postoperative Satisfaction of Dysgnathic Patients Operated On With Resorbable Osteosyntheses

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Background: This study evaluated whether personal expectations and satisfaction throughout orthognathic surgery were fulfilled. In addition, patients were interrogated about their experience of resorbable osteosynthesis.

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Methods: A total of 50 patients were interviewed 3 times each throughout the study by a mixed questionnaire of standard psychologic tests and a tailored itemized questionnaire regarding their expectations regarding resorbable osteofixation and their postoperative satisfaction.

Results: A postoperative increase in self-esteem and approach to life were evident. An examination of Oral Health–Related Quality of Life showed constant quality of life; an examination of Oral Health Impact Profile–Germany) showed no postoperative difficulties in dental hygiene and nutrition. No statistically significant change in any of the tests could be expressly determined. Avoidance of secondary surgery motivated 94% to choose resorbable osteofixations, although a mere 66% had heard of them before; 90% of patients were satisfied with the operation result.

Conclusions: Orthognathic surgery cannot change preexistent depression or a problematic social background. Mastication and oral health improved, and postoperative happiness and confidence increased. When given the choice between resorbable fixation and titanium osteofixation, patients generally preferred resorbable fixations.

Key Words: Orthognathic surgery, questionnaire, resorbable osteosynthesis

It is often suspected that orthognathic surgery positively changes self-esteem and self-disclosure for patients. Even oral function, like dental hygiene, should improve and become simplified. This study therefore evaluated by anonymous questionnaire whether personal preoperative expectations are fulfilled, and posttreatment satisfaction is adequate. In addition, patients were interrogated about their knowledge and expectations of resorbable osteosyntheses and their preference for either titanium or resorbable osteosynthesis.

Resorbable osteosyntheses have been in regular surgical use for many years, especially in craniofacial surgery.[1–3] There has been widespread investigation concerning clinical application, stability, and compatibility.[4–7]

Health insurance covers the costs for bimaxillary surgery in most countries when osteofixation uses titanium osteosyntheses, but not yet when using resorbable fixations, and contradictorily, costs for secondary surgery of metal removal are paid.

Although some previous studies investigated the psychologic aspects of orthognathic patients before and after surgery, this study tried to evaluate the psychologic outcome after orthognathic surgery treated with resorbable osteosynthesis, which excluded secondary surgery of metal removal.[8,9] None of the polled patients underwent the operation for mere aesthetic improvement without a dysgnathic occlusal malformation; that is, all patients were primarily transferred because of orthodontic problems.

Therefore, an anonymous questionnaire asking about personal feelings regarding oral health and general psychologic well-being was handed out to each of 50 orthognathic surgery patients preoperatively, postoperatively, and at the follow-up. It was hypothesized that the patients have more self-esteem and self-disclosure after treatment. Furthermore, patients were asked which osteosynthesis systems they would prefer if given the choice, the reasons for their decision, and adverse effects they could imagine.

PATIENTS AND METHODS

This study has a retrospective design. All patients were operated on and polled in a single center. All were patients of the Frankfurt University Medical Center; the catchment area was mainly local with few patients from the rest of Germany. The sample group comprises of patients who were nearly all whites, and some with African origin. The outlines in the declaration of Helsinki were adhered to, and the formal ethical approval obtained from our local board. All included patients were at least 16 years of age; all patients with dysgnathic and occlusal malocclusion were included. Those with dysgnathic malformation after trauma or patients with congenital malformation were not excluded from the study. Both patients of Angle class II and those of class III were included. The study included altogether 64 patients who received questionnaires, but 14 patients did not return the questionnaires, so 50 patients could at least be included. Fifty patients completed the questionnaire before operation, 47 completed the questionnaire postoperatively, and 48 patients 1 year after surgery. Forty-five completed all 3 questionnaires: 2 at preoperative and postoperative and 3 at preoperative and follow-up. The primary intention was that 50 identical patients received all 3 questionnaires, but because of geographical mobility, not all patients received all 3 questionnaires. Exclusion criteria were disability to fill out the questionnaire autonomously and refusal to participate. The preoperative questionnaire was handed out between 2 and 6 weeks before the operation; the postoperative questionnaire was given 4 to 8 weeks afterward. The follow-up questionnaire was handed out 11 to 14 months after the operation.

The questionnaires contained 112 questions preoperatively, 107 postoperatively, and 107 in follow-up, subdivided into 5 different test groups. The 5 preoperative questions not included in the postoperative and follow-up survey were part of a group of questions specifically constructed for this study questionnaire. The tests were Rosenberg[10] Self-esteem Scale, Zung Depression Scale,[11] OHIP (Oral Health Impact Profile), and Oral Health–Related Quality of Life, which correlates with the Orthognathic Quality of Life Questionnaire (OQOL).

The authors considered 7 tailored preoperative questions and 2 postoperative questions on resorbable osteosynthesis relevant for the study. These asked the patients about their knowledge on resorbable osteosynthesis, their expectations of adverse effects, and their opinion in favor of or against such materials. The questionnaire also contained questions on problems in healing, confidence with the result, attitude regarding the removal of metal osteosynthesis, and the legitimacy of higher costs in specific. The questions in detail were as follows:

1. Have you ever heard of resorbable osteofixation? (Plates and screws that disintegrate by themselves in the body and are applied to fixate fractures and continuity defects.) *This should show the level of preoperative information.*
2. What would you think are their benefits and potential adverse effects? *This should show eventually undefined preoperative anxiety or superstition.*

The patients were further given background information: Using titanium osteofixation, in 12% of cases adverse effects such as heat or cold sensitivity, inflammation, and plate loosening do occur. In resorbable osteofixation 6% of patients encounter adverse effects that mainly consist of foreign body granuloma (ie, a kind of chronic subacute infection with occasional fistulation and drainage) that requires curettage.

The next questions were related to this information.

3. With this information in mind, would you prefer titanium or resorbable osteofixation? *This should show the informed preference.*
4. Why have you decided to receive titanium or resorbable fixation? *This shows the reason for their preference.*
5. What was the decisive point? *This shows the patient's interest within the issue.*

6. Does the idea of a later metal removal bother you? *The most important benefit according to the literature.*
7. Do you consider it justified to demand 50% higher implant costs for resorbable fixation, keeping in mind the successful avoidance of subsequent metal removal? *The most used argument of public health providers' issue.*

In the postoperative and follow-up questionnaires, the following 2 questions replaced the earlier listed question (7):

1. Have you suffered any adverse effects?
2. How content are you with the outcome of the operation?

The Rosenberg[10] Self-esteem Scale is a standard test in clinical outcome studies. It can be used for adolescence as well as for elderly persons. It consists of a 10-item self-report measure of feelings of self-worth or self-acceptance. The items are answered on a 4-point scale ranging from strongly agree to strongly disagree, scoring 1 to 4 points. Overall scores range from 10 to 40, with higher scores indicating greater self-esteem.[10]

The Zung Self-rating Depression Scale[11] is a 20-item self-report questionnaire that is widely used as a screening tool, covering affective, psychologic, and somatic symptoms associated with depression. The items are framed in terms of positive and negative statements. Each item is scored on a Likert scale ranging from 1 to 4 points. A total score is derived by summing the individual item scores and ranges from 20 to 80. Most people with depression score between 50 and 69, whereas a score of 70 or greater indicates severe depression. The Zung scale provides a simple tool for monitoring changes in depression severity over time in research studies.[12]

The OHIP Questionnaire measures the quality of life relating to oral health.[13–15] In this context, it was needed to measure the improvement of oral health through orthognathic surgery. The OHIP-G scale used in this study contained 53 questions (49 from the original English study plus 4 additional questions for the German population), subdivided into 5 possible answers ranging from never to often. *Never* counted for 0 points; *often* counted for 4 points. Higher scores indicate greater quality of life.

The OQOL consists of a catalogue of 22 questions subdivided into 5 possible answers ranging from never to often concerning the social consequence of facial deformity, facial aesthetics, function of mastication, and aesthetic facial self-perception (5 questions on oral function, 5 questions on oral aesthetic, 4 questions on awareness, and 8 questions on social aspects). The OQOL primarily addresses mental rather than physical health. *Never* counted for 0 points, increasing to 4 points for *often*. Lower scores here indicate better quality of life, and higher scores indicate poorer quality of life.[16,17] This inverse value system was chosen to avoid inattentive completion of the questionnaires. All questionnaires were anonymized by numbers and evaluated by an independent research fellow who was not performing the surgeries to avoid bias. All statistic calculation has been done using SPSS (SPSS Inc, Chicago, IL).

RESULTS

The Rosenberg Self-Esteem Questionnaire

By merely comparing the median before and after (postoperatively and in follow-up a year later) surgical intervention, preoperatively 24 of 40 points were reached. This result increased postoperatively up to 25 points and finally ended up at 26 points in the year's follow-up.

However, these findings did not prove statistically significant in the Wilcoxon signed rank test (preoperative to postoperative: $P = 0.9$; postoperative to follow-up: $P = 0.08$; follow-up to preoperative: $P = 0.1$).

The Zung Depression Scale

A maximum of 80 points in the Zung Depression Scale corresponds to severe depressions; 50 to 69 points are analogous with moderate depressions. By regarding the preoperative results, 32 of 80 points were reached in the median. Throughout the observation, the level of depression increases from 32 preoperatively up to 35 points postoperatively; after a year's follow-up, the values decrease to 34 points. No patient reached a severe or moderate level of depression in the observation.

These changes in the results cannot be proved statistically significant in the Wilcoxon signed rank test (preoperative to postoperative results: $P = 0.4$; follow-up to postoperative results = 0.6; follow-up to preoperative results $P = 0.4$).

The OHIP Scale

Preoperative results present a median of 33 points of a possible maximum of 212 points. An increase to 35 points can be perceived in the postoperative results. However, values decreased to 31 points after a year's follow-up.

No statistically relevant data could be assessed in the Wilcoxon signed rank test (preoperative to postoperative results: $P = 0.2$; postoperative to follow-up results $P = 0.4$; follow-up to preoperative results: $P = 0.7$).

The OQOL Score

Concerning the social aspects domain, the median preoperative results reached 4 of 32 points, the postoperative as well as the follow-up results reached 5 points. No statistically relevant data could be attained in the Wilcoxon signed rank test (preoperative to postoperative results: $P = 0.9$; postoperative to follow-up results: $P = 0.6$; follow-up to preoperative results: $P = 0.6$).

Regarding the dentofacial aesthetic value, the median preoperative results reached 9 of 20 points; the postoperative and the follow-up results reached 10 points each. Statistically significant changes could not be shown in the Wilcoxon signed rank test (preoperative to postoperative results: $P = 0.7$; postoperative to follow-up results: $P = 0.1$; follow-up to preoperative results: $P = 0.1$).

The median functional mastication value preoperatively reached 5 of 20 points. The median postoperative and follow-up values were each 6 points. Statistically significant changes in the data in the Wilcoxon signed rank test could not be shown (preoperative to postoperative results: $P = 0.8$; postoperative to follow-up results: $P = 0.4$; follow-up to preoperative results: $P = 0.6$).

Regarding the awareness scale, preoperative 7 of 16 points were achieved in median. In postoperative and follow-up results, 7 points were reached. Wilcoxon signed rank test could not prove statistically significant changes in the data (preoperative to postoperative results: $P = 0.4$; follow-up to postoperative results: $P = 0.5$; follow-up to preoperative results: $P = 0.5$).

The null hypothesis assumed that there is no difference in the named values. The study was performed on a significance level of 0.05. This implies that there are statistically important changes at $P < 0.05$. No significant changes could be found in the tested questionnaires. In none of these tests, the null hypothesis could be rejected.

The additional questions about the expectations of the operation and the patients' knowledge about resorbable osteosynthesis preoperatively were answered as follows: 66% of the patients had

heard of resorbable osteosynthesis, but 98% preferred resorbable osteosynthesis rather than a titanium plating system after having gathered information about resorbable osteosynthesis. The reason for the decision was the chance to avoid a second operation for metal removal in 94% of cases; in 6%, the recommendation of others led to the decision. Asked about their expectations for adverse effects, 31% of patients answered inflammation, 11% answered early resorption, and 11% feared that the plates do not resorb at all or at least incomplete over time. Nevertheless, 41% did not expect adverse effects. When asked for the eligibility to charge higher costs for the resorbable materials from health insurance, 55% thought it justified.

Postoperative questionnaires showed the following results when asked about adverse effects that occurred: 73% did not have any adverse effects at all. When there were adverse effects, most were hypoesthesia (12%), followed by mastication problems, swelling, fistulation, inflammation, pain, and so on, all in 2% or 3% of cases. In total, 55% of the patients operated on were very happy with the outcome of the operation, 37% were satisfied, 2% were unhappy with the outcome, and 6% were not sure.

Asked about their confidence with the result 1 year after the operation, 64% were very happy with the result, 23% were confident, and still 6% were not sure. The discontent patients' percentage, however, increased from 2% to 7%.

DISCUSSION

A study by Motegi et al[18] evaluated the psychologic function and the health-related quality-of-life outcome in a 5-year follow-up of patients who received orthognathic surgery. In conclusion of their evaluation, there has been a significant change in many measured parameters 2 years after surgery, but there has been no improvement from 2 to 5 years. In our study, there were no significant changes in the psychosocial parameters 1 year after operation. One explanation could be that patients need more time than a year to feel changes or change their psychosocial behavior. Rispoli et al[19] basically came to the same conclusion that a negative presurgical body image persists in the patient's mind, and a long process is necessary to achieve acceptance of the new body image.

According to Rispoli et al,[19] the level of depression in this study measured by the Zung scale was also not influenced by the surgical treatment. Apparently, the outcome of orthognathic surgery does not affect depressive tendencies, although it has to be kept in mind that, in the presented study, no patient had severe depression, which may affect the perception of the operation.

The OQOL score is an adequate method for measuring quality of life in orthognathic surgery.[16] Unlike Cunningham et al,[17] where the follow-up questionnaire was given after removal of orthodontics, our patients received the questionnaire 12 months after surgery. This, on the one hand, ensures that all patients are in the same stage of healing and having had the same time to cope with the new situation. On the other hand, not every posttreatment with braces might be finished, and the results may differ because of that. Rispoli et al[19] just took 3 months for the postsurgical evaluation, which from our point of view may be too short after intervention. The healing process may need more time, residual swelling may still mask the surgical result, or the new facial symmetry may not be incorporated into the patient's body concept.

The OQOL in our study showed a slight but no significant improvement in mastication and functional parameters but not in the expected range. According to this, the OHIP scale change was, over time, also not in the expected dimension. One possible explanation could be that patients do not sense their malfunction as severe as many surgeons or orthodontics expect. Another explanation may be that soon after surgery the problems with the preoperative malfunction are displaced by general problems with dental hygiene or braces. The slightly but not significantly increased facial aesthetic in our study has also been shown in studies by Motegi et al[18] and Rispoli et al.[19] Even if they have shown higher increase, an improved facial aesthetic profile can be assumed. The change here was also nonsignificant as referred to the questionnaires. The clinical and objective impression from the surgeon's point of view was a large increase in facial aesthetic. This impression again does not align with the questionnaires. Possibly most patients do not have a personal problem with their appearance, and mainly, a beauty ideal in our society is the real problem.

A convincing majority of the patients do prefer resorbable implants (98%) because almost every patient dislikes the possibility of metal removal (94%). These data are in line with the findings of Mittal et al,[20] who have asked the same questions in their orthopedic patient sample and obtained nearly the same percentage in answers (95% decision for resorbable fixations, 91% were concerned about metal removal). In comparison to the same study by Mittal et al,[20] none of our patients expected the resorbable plates to be less strong than titanium according to 29% in the Mittal study, but 22% in our study sample expected problem with the resorption of the implants; only 7% in the study of Mittal et al feared that.

In a study by Cunningham et al,[21] they state that a detailed discussion with their preselected patient sample would be necessary for a successful outcome of surgery and patients' confidence. The presented study did not preselect patients with personal motives, but patients in this study had occlusal and associated dentofacial dysgnathia. The results show that the same satisfaction percentage as in the previously mentioned study was reached throughout the study (Cunningham et al: very pleased 67%, reasonably pleased 28%; present study: 64% very pleased, 23% reasonably happy).

Overall, most patients were indeed satisfied with the outcome, but this interestingly did not express in the psychologic tests. The increase (2%–7%) between postoperative questionnaire and follow-up of discontent patients may result from discrepancies between high expectations and the outcome of the operation. It is therefore important that patients receive widespread education before surgery so that unrealistic expectations can be corrected.

Cunningham et al[21] found no significant differences in self-esteem preoperatively and postoperatively. This is also reflected in the presented study, where no significant increase in self-esteem can be seen. Cunningham et al[21] also found that the overall depression was low; this did not change drastically through operation. The evaluated Zung index in this study mirrors these results. Depression reported immediately after the operation could be considered as normal, regarding the special situation for the patients, coming primarily healthy to the hospital and feeling ill after the intervention.

Many patients complained about the extensive questionnaires. That, in some cases, led to disinterested and noncritical answering of the questionnaires. Because of that, some questionnaires showed discrepancies in the way of answering, for example, a self-confident and happy patient with major depression index. Perhaps fewer questions but those with special focus on craniofacial surgery might be asked.

CONCLUSIONS

Orthognathic surgery certainly changes the facial aesthetic but does not influence depression or social interaction in general. Most patients' self-assurance improves after the operation; the slightly increased values of self-consciousness and positive attitude toward

life may underline these facts. As expected, nutrition and dental hygiene are difficult directly after operation, which in the course of healing leveled off to the preoperative standard.

The personal decision for the osteosynthesis material led, in most cases, to the resorbable materials. The acceptance of resorbable osteosynthesis is very high because of the avoidance of a second operation. Even higher costs for these materials are felt to be justified by most patients.

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Pilomatrixoma Imitating Infantile Hemangioma

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Abstract: Many lesions can masquerade as infantile hemangioma, the most common tumor of infancy. We describe an infant with pilomatrixoma mimicking hemangioma. The patient presented at 8 months of age with an 1.2-cm, red, asymptomatic lesion of the right ear. The mass was noted at 4 months of age and subsequently enlarged. Ultrasonography demonstrated hypervascularity "consistent with infantile hemangioma," and the infant was observed. At 12 months of age, the lesion continued to expand and became ulcerated; it was excised, and histopathology showed pilomatrixoma. Diagnostic confusion was caused by atypical features of this pilomatrixoma that overlapped with infantile hemangioma: onset in infancy, ulceration, red color, and fast flow on imaging. Deviation from the predictable clinical features of an infantile hemangioma should prompt consideration for other types of pediatric lesions, including pilomatrixoma.

Key Words: Hemangioma, pilomatrixoma, pilomatrixoma, pediatric, ear, infant

Infantile hemangioma is the most common tumor of infancy. This lesion has distinctive characteristics including a pathognomonic clinical course with rapid expansion in early infancy and subsequent regression, a classic red appearance, and fast flow on ultrasonography. We present a patient who had a pilomatrixoma of the ear with atypical features that paralleled the history, physical examination, and imaging findings of infantile hemangioma.

CLINICAL REPORT

A full-term, 8-month-old healthy male infant presented with an asymptomatic lesion of the right ear that had developed at 4 months of age. The mass slowly enlarged over the next few months. There was no history of trauma or bleeding.

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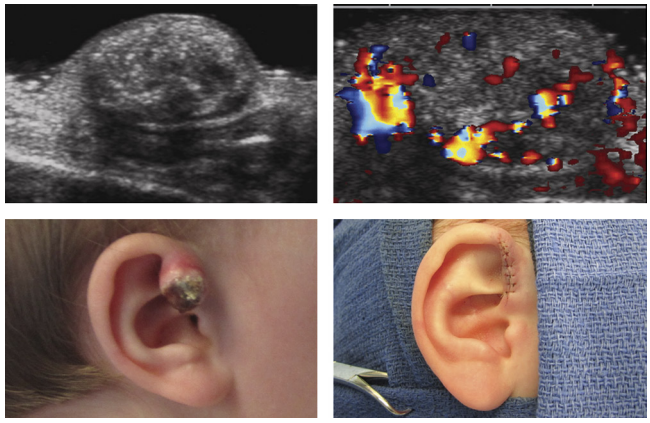

FIGURE 1. Infant with a history of an enlarging, red lesion of the ear. At 8 months of age, gray-scale ultrasonogram showed a well-demarcated, heterogeneous, oval mass in the skin and superficial subcutis (above, left). Multiple discrete hyperechoic foci suggest microcalcifications. Color-flow Doppler ultrasonography demonstrates hypervascularity with enlarged feeding arteries around and within the lesion consistent with ''infantile hemangioma'' (above, right). At 12 months of age, the mass ulcerated and continued to expand (below, left). After resection (below, right).

On physical examination, a 1.2-cm, red, firm, nontender mass was present at the base of the right helical root. Handheld Doppler examination revealed fast flow. Physical examination was most consistent with infantile hemangioma. However, because the lesion was firmer and noted later than a typical infantile hemangioma, an ultrasonogram was obtained. Imaging showed a well-circumscribed, echogenic lesion with hypervascularity that was "consistent with infantile hemangioma" (Fig. 1). The patient was observed.

Four months later, on follow-up examination, the lesion was larger, ulcerated, painful, and bleeding. Because the mass was now symptomatic and continued to expand at 1 year of age, which was inconsistent with infantile hemangioma, it was resected. Histopathology showed pilomatrixoma (Fig. 2).

DISCUSSION

Infantile hemangioma is common, occurring in 4% to 5% of infants.[1] The head and neck are most frequently affected (62%).[2] Ninety percent are diagnosed by history and physical examination.[3,4] The tumor appears at a median age of 2 weeks, proliferates to 80% of its maximal size by 5 months of age, and usually completes growth by age 9 months.[3–5] Involution begins at approximately 1 year; the tumor flattens, and its color fades. In most children, regression stops by 6 years of age.

Infantile hemangioma may be confused with other skin lesions that are common in the pediatric population. Pilomatrixoma, a benign calcifying tumor of the hair follicle matrix cells, also predominantly affects the head and neck (68%).[6,7] Eighty-eight percent occur before 10 years; the average age at excision is 5.8 years.[6] In contrast to infantile hemangioma, pilomatrixoma presents before 2 years of age in less than 10% of patients.[6,7] In 1 large series, the youngest patient was 18 months old.[7] Most masses are slowly growing, firm, and flesh-colored or bluish and may have small, white calcifications.[7] Although the average nodule size is 1.2 cm, rapid enlargement can occur from intralesional bleeding.[6]

If pilomatrixoma cannot be differentiated from infantile hemangioma based on history and physical examination, imaging or histopathology may be necessary. Ultrasonogram of infantile hemangioma shows a well-circumscribed, hypervascular mass with enlarged arteries and draining veins.[8] Pilomatrixoma appears as an ovoid, complex mass with a hypoechoic rim and echogenic center at the junction of the dermis and subcutaneous fat; calcification can cause shadowing.[9]

Histopathologically, proliferating infantile hemangioma has mitotically active plump endothelial cells that stain for erythrocyte-type glucose transporter, GLUT 1.[10] Pilomatrixoma contains stroma with irregularly shaped islands of epithelial cells, basophilic and eosinophilic keratinized (shadow) cells, and calcification and foreign-body granuloma formation.[6,7]

Differentiating pilomatrixoma from infantile hemangioma is important because the lesions are managed differently. More than 90% of hemangiomas do not require intervention; problematic lesions are treated with pharmacotherapy or resection. In contrast, most pilomatrixomas are excised to obtain a definitive diagnosis or to alleviate symptoms, for example, visible deformity or pain.[6,7]

In conclusion, we present an infant with pilomatrixoma imitating an infantile hemangioma. Diagnostic confusion was the result of atypical findings exhibited by the pilomatrixoma, which overlapped with infantile hemangioma: (1) unusually early onset in infancy, (2) a red appearance with subsequent ulceration, and (3) hypercvasularity on ultrasonography. Imaging and/or biopsy should be considered if there is deviation from the expected appearance and natural history of infantile hemangioma.

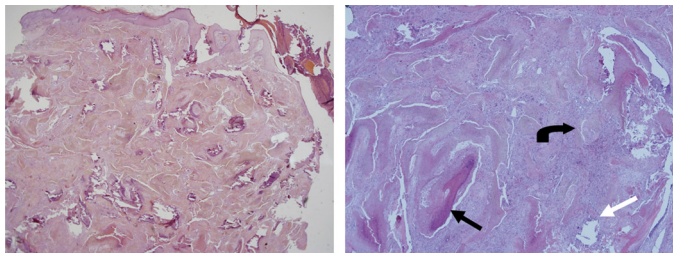

FIGURE 2. Histopathology of the ear lesion shows pilomatrixoma. Hematoxylin-eosin staining on low-power (original magnification ×2) magnification exhibits intradermal proliferation (left). Higher magnification (original magnification ×20) demonstrates shadow cells (curved arrow), basophilic areas (straight arrow), and reactive histiocytes (white arrow) (right).

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Giant Temporalis Muscle Metastasis of Esophageal Carcinoma

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Abstract: Temporalis muscle metastasis of a tumor is a rare condition. Basaloid squamous cell carcinoma is an uncommon variant of squamous cell carcinoma, which often occurs in the aerodigestive tract. To the best of our knowledge, there have been no previous reports dealing with temporalis muscle metastasis from esophageal carcinoma in the literature.

Key Words: Temporalis muscle metastasis, esophageal carcinoma, basaloid squamous carcinoma

Head metastasis of a tumor is easily misdiagnosed at the early stages. The differential diagnosis is important for primary disease diagnosis, treatment, and in preventing large destructive masses.

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Esophageal carcinoma has a high mortality rate, with a 5-year survival rate of 10%.[1] Patients with esophageal cancer usually present with disease that is locally advanced and that is already at a metastasized stage at the time of initial diagnosis. Cancer of the esophagus exists in 2 main forms with different etiologic and pathologic characteristics: squamous cell carcinoma and adenocarcinoma. Basaloid squamous cell carcinoma (BSCC) is an uncommon variant of squamous cell carcinoma (SCC), which often occurs in the aerodigestive tract. Primary BSCC of the esophagus is uncommon and is usually associated with a poor prognosis.

Temporalis muscle metastasis of a tumor is a rare condition.[2] In the current article, we present an interesting case of a large metastasis of esophageal BSCC in the temporalis muscle, which, to our knowledge, represents the first such location described in the literature.

CLINICAL REPORT

A 62-year-old woman presented with an 8 × 5-cm^2 painless, slow-growing subcutaneous mass on the temporal region of a 2-year history (Fig. 1). It started as a 2 × 2-cm^2 nodular mass and regarded to be a benign lesion with no biopsies. The lesion was painless, solid, and fixed. The covering skin appeared normal and unattached. There was no facial nerve weakness or trismus, and no neck nodes were palpable. Her medical and surgical histories were significant for esophageal squamous carcinoma treated 2 years earlier with surgery (negative surgical margins and lymph node status at that time) but no radiotherapy or chemotherapy.

In magnetic resonance images, the mass had a malignant appearance and infiltrating temporalis muscle. There was no destruction of the underlying temporal bone (Fig. 2). An incisional biopsy was performed, and a pathologic diagnosis revealed basaloid squamous carcinoma. The tumor was completely resected with 1-cm safety margins along with the bulk of the temporalis muscle, with an intact appearance of the temporal bone (Fig. 3). A pathologic diagnosis revealed metastasis of basaloid squamous carcinoma with negative surgical margins. On microscopic examination, nests

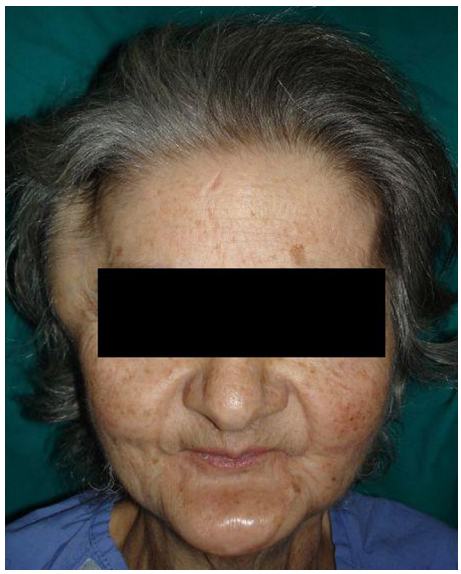

FIGURE 1. Patient presented with an 8 × 5-cm^2 painless subcutaneous mass on the temporal region.

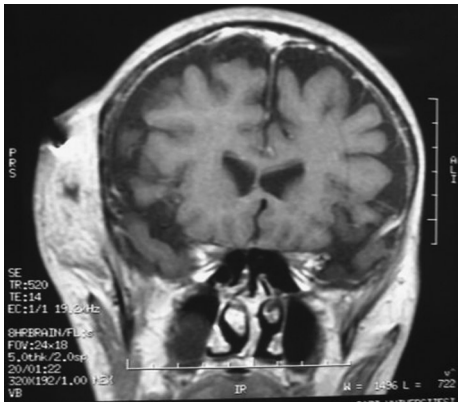

FIGURE 2. Appearance of the tumor using MRI.

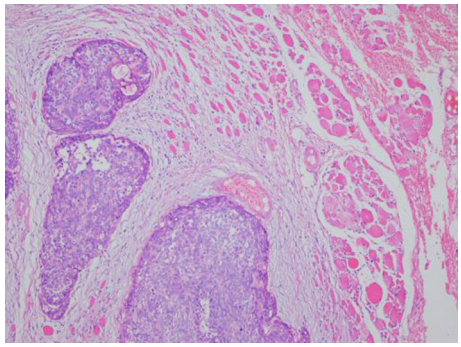

FIGURE 4. Biopsy specimen showing atypical basaloid cells adjacent to the muscular layer (hematoxylin and eosin stain, original magnification ×100).

of basosquamous carcinoma were seen. The basosquamous carcinoma cells spread in and adjacent the temporalis muscle. The peripheral layer of the tumor nests showed a palisade arrangement of the nuclei (Fig. 4).

DISCUSSION

Diagnosis of esophageal carcinoma is often delayed until late stages because of its inaccessible anatomic location. It is well known that esophageal cancer may cause distant metastases. Initially, it tends to spread locally followed by metastasis to the lymph nodes and then to the distant organs. It has a variable tendency to metastasize to other parts of the body.[3,4] Although esophageal carcinoma frequently metastasizes to the lung, pleura, liver, stomach, peritoneum, kidney, adrenal gland, and bone, muscle metastases have been infrequently reported in the literature as few small series and clinical reports.[5–8] There are no reported cases of temporalis muscle metastasis from esophageal carcinoma. Our patient had no other malignancy except for ESCC. In addition, the potential for double primary tumors with one being a primary muscle squamous cell carcinoma is extremely unlikely. It is therefore reasonable to assume that the tumor in the temporalis muscle was a metastasis from primary esophageal cancer.

In the present case, temporalis muscle metastasis of esophageal carcinoma, which represents the first such location described in the literature, along with its vague symptoms and clinical appearance, resulted in the patient's delayed referral, thus the tumor was quite large when diagnosed. Although excision of this metastasis was palliative surgery, it should be kept in mind atypical localization metastasis may occur at the head region and early diagnosis may have some beneficial effect on the patient's survival and palliative surgeries.

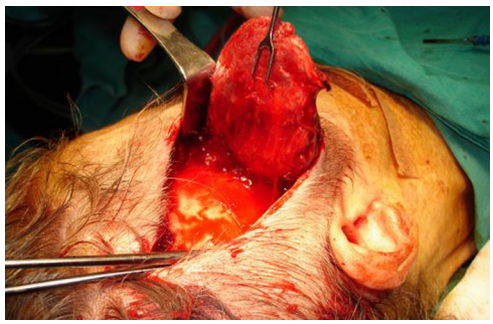

FIGURE 3. Intraoperative appearance of the tumor.

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Median Palatine Cyst

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Abstract: The median palatine cyst is a rare benign nonodontogenic lesion that attacks the median palatine suture. There is controversy about its pathogenesis; however, its origin is generally attributed to

the enclavement of epithelial remnants within the palatine suture between the 2 lateral maxillary processes during their fusion in the origin of the hard palate. The purpose of this report was to relate a case of a median palatine cyst, discussing the rarity of the lesion, its pathogenesis, and the different modalities that could be used for the correct treatment of this pathologic entity.

Key Words: Nonodontogenic cyst, hard palate, pathology, jaw cyst

The median palatine cyst is a rare fissural cyst of nonodontogenic origin, located in the midline of the hard palate, posterior to the palatine papilla.[1–4] Only 20 cases have been reported in the literature up to now, and the last one described in 2001 measured 5 cm in diameter.[5]

There is controversy about its pathogenesis. Its origin is generally attributed to the enclavement of epithelial remnants within the palatine suture between the 2 lateral maxillary processes that fuse to originate the hard palate.[6–9]

Often discovered during routine clinical or radiographic examinations,[2] it has been shown to develop as a circular or ovoid lesion, producing a defined, asymptomatic, and symmetrical swelling that may extend as far as the molar region.[10] In radiographs, it is shown to be radiolucent and could involve the nasal floor.

The teeth associated with the median palatine cyst are often vital, and their migration has been observed in several cases.[4] Most of those cysts are asymptomatic, although some patients have complained of pain on palpation. Enucleation is the most widely used treatment for this lesion, with a low rate of recurrence.

The differential diagnosis of the median palatine cyst includes the cysts of the incisive canal and palatine anterior maxillary cysts, such as the nasopalatine duct cyst.[4] Zachariades and Papanikolaou[11] affirmed that the subdivision of palatal suture cysts into median alveolar (anterior) and median palatine (posterior) cysts was purely academic. However, the median palatine cyst differs from other defects of the palate by presenting specific characteristics, including epithelial lining, absence of salivary glands, vascular or neural elements on the cystic wall, and its location in the posterior palate.[12]

Histologically, it presents a cystic cavity covered by epithelium that contains stratified squamous cells, often with respiratory epithelial components. The wall is composed of moderately dense collagenous fibrous connective tissue.[13,14]

As a result of the rarity of the median palatine cyst, and the difficulty in establishing a differential diagnose with other maxillary cystic lesions, the purpose of this study was to report a clinical case of a patient who presented a lesion in the palate consistent with the median palatine cyst and to discuss details of its pathophysiology, diagnosis, and treatment.

CLINICAL REPORT

The patient, a 28-year-old man, came to the Oral and Maxillofacial Surgery Division of Araraquara Dental School, referred by his doctor for evaluation and treatment of a swelling in the hard palate, suggestive of a tumor.

There was no history of allergic symptoms, and the patient presented good general health and was in no pain. No pathologic finding was related that enabled the nature of the lesion to be identified. In the clinical intraoral examination, the presence of a swelling was observed in the palatine area (Fig. 1), related to the maxillary right central incisor and maxillary right lateral incisor, which were vital. There were no alterations in the color and texture of the hard palate.

An occlusal radiograph revealed an ovoid and well-circumscribed radiolucency, extending from the right central incisor to the right first molar, crossing the palatine suture (Fig. 2). There was no divergence between the roots of the maxillary central incisors.

The diagnostic hypotheses were nasopalatine duct cyst, median palatine cyst, and keratocyst. The treatment plan was surgical enucleation of the lesion. The root apices of the central and lateral incisors were projected inside the lesion; therefore, rupture of the vasculonervous bundle of the teeth during surgery would be inevitable. This finding justified the choice of performing previous endodontic treatment of the teeth involved.

Aspiration of the palatal swelling was performed, and a seropurulent secretion was withdrawn. Therefore, the patient was submitted to a cyst-enucleation procedure.

The lesion measured 4 × 2.5 cm and presented a flat surface. The tissue was brown and fibrous, and the specimen was sent for histopathologic examination. Microscopic examination revealed the presence of a stratified squamous epithelium, partially ulcerated, with edema between the squamous cell layers, without atypia in the remaining areas (Fig. 3), covering the cystic structure. In the nucleus,

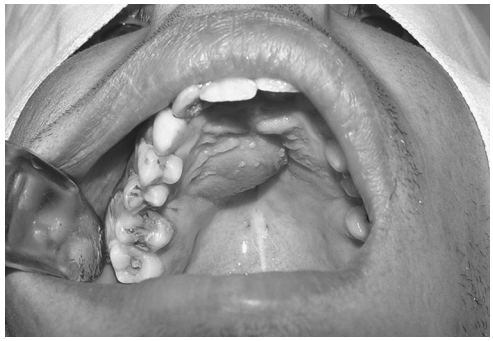

FIGURE 1. Clinical aspect of the lesion—presence of a swelling in the palatine area.

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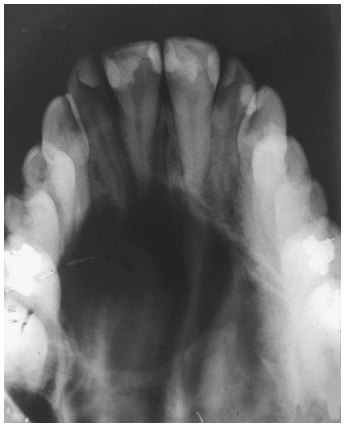

FIGURE 2. Initial occlusal radiograph—an ovoid and well-circumscribed radiolucency was observed, extending from the right central incisor to the right first molar, crossing the palatine suture.

there were areas of edema, infiltrate of lymphocytes and plasma cells, and dense fibrous connective tissue. Trabeculae without atypia were found in the base. There were no salivary glands or any vascular or neural elements in the cystic wall; therefore, the histologic diagnosis was median palatine cyst with an unspecific inflammatory process of the adjacent stroma, without evidence of malignancy.

The patient's postoperative course was uncomplicated. The postoperative radiographic evaluation after 1 year revealed bone regeneration of the palatine area (Fig. 4) and absence of clinical signs and symptoms.

DISCUSSION

The infrequent discoveries of median palatine cyst can be confirmed by the presence of only 2 cases among approximately 25,000 surgical specimens that were reviewed in the Oral Pathology Division files of the University of California in San Francisco.[15] Killy and Kay[16] did not find any case among the 471 benign cystic lesions in the maxillae, and Bradley[17] reported 1 case among 104 cysts in the maxillary bones in a period of 45 years, which corroborate the rarity of the lesion.

The median palatine cyst is commonly confused with the nasopalatine duct cyst, because they present a similar anatomic

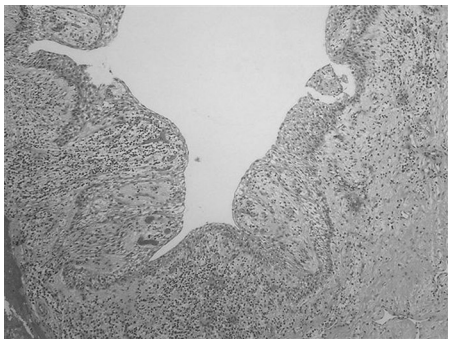

FIGURE 3. Histological findings—presence of a stratified squamous epithelium, partially ulcerated, with edema between the squamous cell layers, without atypia in the remaining areas.

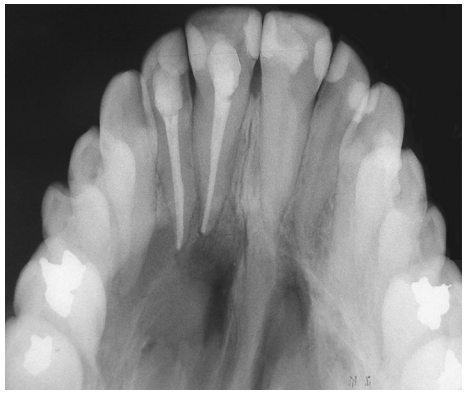

FIGURE 4. Final occlusal radiograph—postoperative radiograph after 1 year reveals bone regeneration of the palatine area.

localization. Lack of mucous glands and lack of involvement of vessels and nerves tend to exclude the nasopalatine duct cyst from the diagnosis of median palatine cyst.[17] Radiographically, the median palatine cyst is circular, and nasopalatine cysts are quite often heart-shaped, because of their anterior location and superimposition of the nasal spine.[18]

The clinical presentation is characterized by a well-defined swelling along the midline of the palate. Most of these cysts are asymptomatic, as the patient of this study related; the cysts may become painful when the nasopalatine nerve is involved by local extension, or when the cyst is secondarily infected and causes painful swelling and a sinus opening into the oral cavity.[11]

Although there are a limited number of examples, the median palatine cyst appears to display a definite predilection for men (the ratio of men to women in reported cases is 4:1). Patient ages ranged from 13 to 52 years, with a mean age of 37 years. Known duration of the cysts ranged from 1 week to 2 years, but most were found to have existed for about 6 months.[2,5,19] All these characteristics described in the literature were also observed in this clinical case.

The pathogenesis of the median palatine cyst is very controversial. It may be a result of abnormal palatal development during embryogenesis.[20] Lack of complete disintegration of epithelium at the fusion line may result in the growth of such cysts along the midline of the hard palate.[8] However, for unknown reasons, these epithelial remnants proliferate later in adult life to form cysts.[21] The range of masticatory movements causing slight movement of bones[22] is thought to forestall the synostosis; this movement may be enough to excite proliferation of potentially cystic epithelial remnants. Other researchers consider these cysts to be derived from odontogenic cell rests.[11,16]

The median palatine cyst was mainly treated by enucleation, although there was 1 case of marsupialization in the literature.[23] Killy and Kay[16] state that marsupialization of fissural cysts is unsatisfactory, because the cavity is not obliterated; the entire specimen is not available for histopathologic diagnosis, and repair is delayed.

In this clinical case, the lesion was treated by enucleation. The cyst was approached intraorally by elevating a posteriorly based mucoperiosteal flap and enucleated, and the surgical area was curetted, debrided, and irrigated to ensure complete removal of epithelial cells.

The median palatine cyst is considered a rare fissural non-odontogenic lesion that must be distinguished from the nasopalatine duct cyst.[24] The treatment of choice is enucleation, and the surgical access depends on the size and extension of the cyst. There should be complete excision to avoid recurrence.

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Giant Postauricular Fibrolipoma

Vipula Perera, MD, Amila Sasanka, MD, Dammika Dissanayake, MS

Abstract: Fibrolipoma belongs to the benign tumor variant of lipoma characterized by the presence of adipose tissue and large volume of fibrous tissue. Our patient who was a 50-year-old mother of 2 children presented with a recurrent pedunculated large lump from right postauricular region of 13 years' duration. She underwent en mass excision of the lump under general anesthesia with primary closure of surgical wound. The tumor weighed 2.55 kg, and histopathologic examination revealed fibrolipoma. She was followed up for 6 months and found no recurrence.

Key Words: Fibrolipoma, postauricular lump, benign tumors

CLINICAL REPORT

Our patient was a 50-year-old mother of 2 children who presented with a lump hanging from the right side of the head (Fig. 1). She had no pain, but lump was disfiguring. She had undergone surgery thrice at ages of 10, 16, and 33 years. She possessed 2 diagnosis cards with past surgical interventions, but neither containing histopathologic diagnosis.[1]

The current lump was there for 13 years and gradually increased in size. There was an ulcer for 4 months' duration, which was resistant to treatment. The growth of the lump was painless. She was reluctant to expose the lump in public clinic room, although she continued her job as a garment factory worker. At the time of initial clinic visit, she was covering her lump and scalp with a towel. Her medical history was irrelevant.

On exposure, she had a large pedunculated, well-mobile lump originating from the right postauricular area. The lump was hanging over the clavicle, almost covering the upper half of the right breast. Her right ear was hardly visible. The lump contained multiple punctuate lesions, sebaceous cysts, and single decubitus ulcer over the most dependent area. The skin surface was shiny. The base of the lump was about 6 cm in diameter and well mobile without pain.

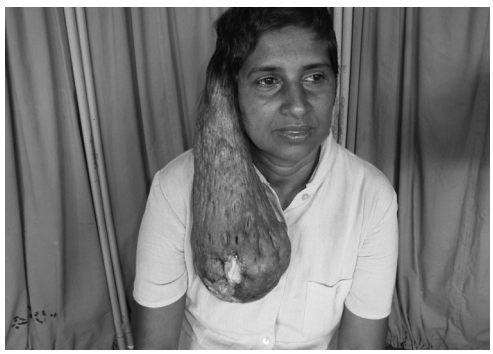

FIGURE 1. Patient at the time of presentation.

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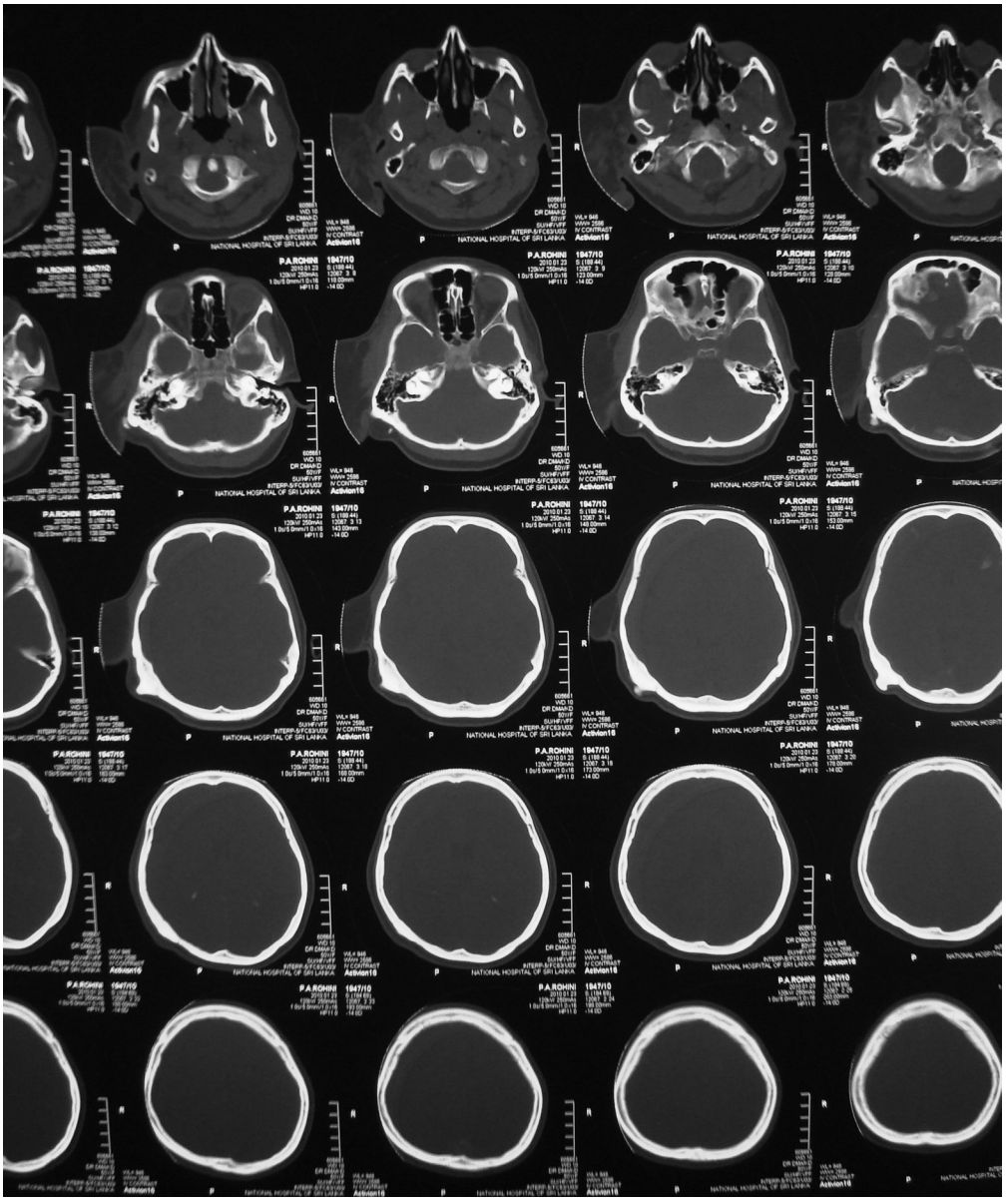

FIGURE 2. Computed tomography scan of tumor.

Although the lump covered the right ear with pressure on it, definitions of the ear were well preserved with intact normal hearing. Posterior to the lump, there was a hard hemispherical lump about 4 × 3 cm in dimension. We suspected an osteoma or intracranial extension of the lump.

A computed tomography scan of the head was obtained to exclude intracranial extension and incisional biopsy under local anesthesia to confirm histopathologic diagnosis (Fig. 2). Incisional biopsy revealed tissue-containing lipoma with fibrous tissue.[2–4] There were no features of malignancy. Unfortunately, biopsy area got ulcerated requiring every-other-day dressing with povidone-iodine. A computed tomography scan revealed no intracranial extension, with bony lump posterior to the tumor. The patient consented to undergo excision of the postauricular soft-tissue lump and not the bony prominence in the right parietal region.

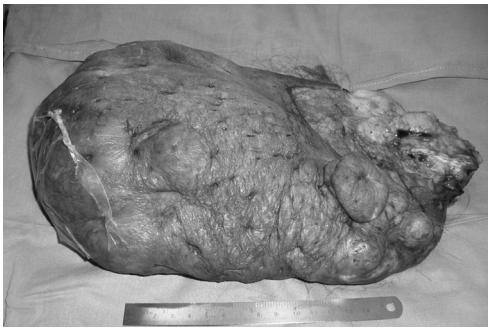

FIGURE 3. Tumor after removal.

FIGURE 4. Patient, postoperative view.

She underwent en mass excision of the lump under general anesthesia. The base of the lump was infiltrated with adrenalin saline (1:200,000). Three flaps were raised circumferentially around the base. There was a well-defined plane separating the tumor from the scalp tissue. Primary closure of the surgical wound was done in layers after insertion of single suction tube drain. Blood loss was about 200 mL. The tumor weighed 2.55 kg (Fig. 3).

She had an uneventful postoperative period. Her drain was removed on the second postoperative day, and all sutures were removed on day 12.

Histology revealed a fibrolipoma. She was followed up monthly in the clinic, and no recurrence was found at 6 months. She has developed relative prominence of the right ear, which was not considered by the patient for further correction. She was delighted with surgery and started her normal day-to-day activities (Fig. 4).

DISCUSSION

Fibrolipoma belongs to the group of benign tumors. It is common in males, according to few case reports found in references. Although fibrolipoma is rare in children, our patient had undergone excision thrice starting at the age of 10 years. Unfortunately, none of the diagnosis cards revealed histopathologic diagnosis. The asymptomatic nature of these tumors allows them to grow for many years, and cosmesis makes the patient seek medical consultation. Although there are clear tissue planes, it is mandatory to make complete excision to avoid recurrence. Literature review does not emphasize such large tumors.[5–8]

ACKNOWLEDGMENTS

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Sequential Maxillary Bifocal Transport Distraction Osteogenesis After Radiotherapy

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Abstract: Maxillary defects secondary to ablative surgery are common, and functional maxillary reconstruction is challenging. Adjuvant radiotherapy further complicates reconstructive surgical planning. In this article, successful functional reconstruction of a partial maxillary defect was achieved using segmental osteotomy, sequential bifocal distraction osteogenesis, and prosthodontic treatment. The current evidence is reviewed, and the surgical planning and technique are described in detail.

Key Words: Maxillary defect, distraction osteogenesis, bifocal transport, radiotherapy, reconstruction

Maxillary defects can lead to long-term functional disabilities and aesthetic complications for the patients. However, the clinical role of surgical maxillary reconstruction is still controversial.[1] Maxillary reconstruction often involves multiple procedures including vascularized free-tissue transfer. The result is satisfactory, but donor-site morbidity is unavoidable.[2–4] Distraction osteogenesis (DO) is a useful treatment modality in maxillofacial reconstruction. McCarthy et al[5] reported the first successful craniofacial reconstruction with distraction in 1992. It has become an important treatment option for maxillofacial defects.[6–8] The use of DO in reconstruction of head and neck oncology is limited by the effect of radiotherapy on the remaining hard and soft tissue.[9] Radiation can

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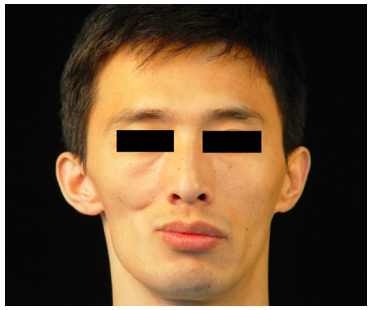

FIGURE 1. Frontal photograph showing depression on the right cheek.

result in hypoxic, hypocellular, and hypovascular changes in the irradiated field.[10] Gantous et al[11] reported successful DO in the irradiated canine jaw, which received radiation dose that is equivalent to human dosage. There were reports of successful DO of mandibular reconstruction in both human and animal models with preoperative radiotherapy.[6,12,13] However, Holmes et al[14] reported 2 cases of failed distraction in an irradiated mandible and found no evidence of bone formation in the distraction gap. Patients who received more than 60 Gy of radiation are considered to be contraindicated for DO.[15] The effect of radiotherapy on DO remains controversial.[6,11–14,16]

Taub et al[17] reported a case of successful closure of oronasal fistula in an irradiated palate using DO. A successful orbitozygomatic complex distraction after radiotherapy was reported by Grover et al[18] recently. However, reconstruction of partial maxillectomy defect using bifocal transport DO after radiotherapy has not been reported.

This article presents the first documented case of maxillary bifocal transport DO after radiotherapy. The surgical technique and biomechanics of a custom-made distractor design are discussed and illustrated.

CLINICAL REPORT

A 33-year-old male patient presented for maxillary reconstruction secondary to malignant melanoma management. The patient had right partial maxillectomy, cryotherapy, and adjuvant postoperative radiotherapy 11 years ago. The radiation dose was 60 Gy, and the radiation field was from the upper dental midline to the right maxillary tuberosity. He was otherwise fit and well. Clinical examination showed obvious facial deformity, and there was a class 2A maxillary defect distal to the right lateral incisor. Three-dimensional reconstructed model showed the maxillary and hard and soft palatal defects (Figs. 1 and 2).

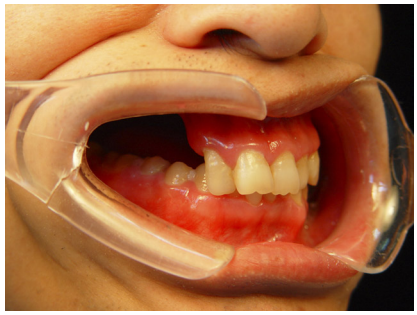

FIGURE 2. The right partial maxillary and alveolar bone defect.

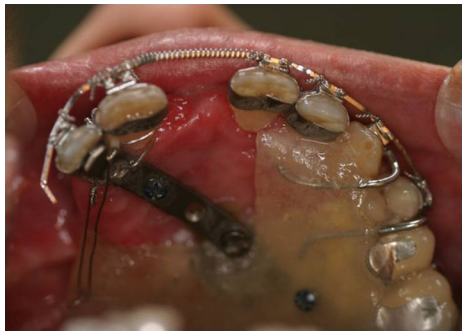

FIGURE 3. The first appliance: dentoalveolar transport disk was distracted by flexible rubber chain and open coil spring under the guidance of edgewise and supporting palatal bar.

Preoperative radiographic and clinical examination was performed to rule out any local and distant metastasis. Treatment plan for reconstruction of the maxillary and palatal defect was discussed in detail with the patient. The patient elected and consented to undergo DO. Stereolithographic model was constructed, and a distractor was designed on the model preoperatively.

SURGICAL TECHNIQUE

Surgery was performed under general anesthesia. A small buccal mucoperiosteal flap was raised with preservation of the alveolar crest and palatal mucoperiosteum to maintain vascularity. The transport disk was created by a horizontal subapical osteotomy 5 mm above the tooth apex and vertical interdental osteotomy between the central incisors. The distractor components were fitted and tested. Mini-implant anchorage was implanted in the region of maxillary tuberosity and a figure-of-eight ligature was placed between central and lateral incisors. The wound was irrigated and closed.

Distraction was started after 5 days of latency period. The palatal supporting bar was connected to the palatal side of the lateral incisor tooth band. Flexible rubber chain and open coil spring on the archwire were used to provide traction. Traction force was adjusted according the transport disk movement to ensure a rate of 0.8 to 1 mm daily for 14 consecutive days. Once the transport disk reached the canine area, 6 months of consolidation was followed (Fig. 3).

In the second procedure, another transport disk was created with the same technique as the first procedure, and mini-implants were

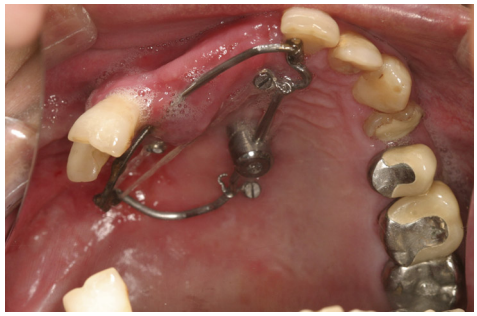

FIGURE 4. The second appliance of sequential DO: transport disk was moved along the curve of the dental arch.

anchored on its palatal side for distraction. A 1.2-mm stainless steel palatal archwire that was secured by 3 palatal mini-implants midpalatally, mesially, and distally guaranteed the transport disk moved along the curve of the dental arch. Flexible rubber chain was used to provide traction on the palatal side (Fig. 4). After a latency period of 5 days, distraction was started at 0.8 to 1 mm/d for 17 consecutive days.

Three months later, the distractor was removed (Fig. 5). Cast cobalt-chromium alloy crowns were made for the central and lateral incisors, which act as retainers for a removable cobalt-chromium partial denture (Fig. 6).

DISCUSSION

Maxillary reconstruction with DO after malignancy management is a less invasive treatment option compared with a free vascularized bone graft. Although treatment time of DO seems longer when compared with other treatment modalities, the use of free-tissue transfer and implant-retained prosthesis would take a similar time frame to complete. Distraction osteogenesis provides functional reconstruction of the hard and soft tissue without donor-site morbidity. The complications associated with free-tissue transfer are also avoided.

The anterior maxillary alveolus is a reliable site for the transport disk selection for DO because of blood supply from the incisive foramen. The transport disk in this case was designed to include the lateral and central incisors. It not only acted as the base for the osteogenesis, but the teeth were also used as abutments for the removable partial prosthesis.

To reconstruct the posterior maxillary dental alveolus across the canine region, DO has to be performed along the dental arch.[19–21] Therefore, sequential DO was necessary, and customized distractors were designed. The first distractor was designed in 3 parts. The first part is a partial acrylic palatal plate that covers the remaining palate with clasps on the contralateral premolar and molar. The second part is a palatal bar that attaches to the transport disk and connects to a palatal screw that acts as a rotational center. The third part is the fixed orthodontic appliance, which acts as a guidance tract for the transport disk. The orthodontic archwire can be easily changed and modified to facilitate three-dimensional movement of the transport disk.

The second distractor consists of a palatal wire that is secured by 3 palatal screws and flexible rubber chains, which provide adjustable palatal traction. Additional rubber chain can be connected to the mini-implant on the transport disk to improve vector control. The palatal wire provides the path of DO.

Maxillary distraction after radiotherapy can be achieved with careful case selection and correct distractor design. Development of a new maxillary distractor to enable single-stage maxillary alveolus reconstruction is necessary. A well-designed prospective study will be needed for further evaluation of this technique.

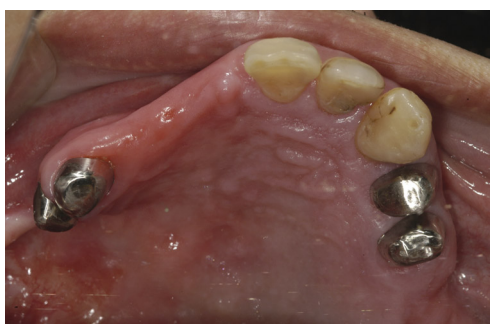

FIGURE 5. Postdistraction occlusal photograph showing complete soft- and hard-tissue maxillary reconstruction.

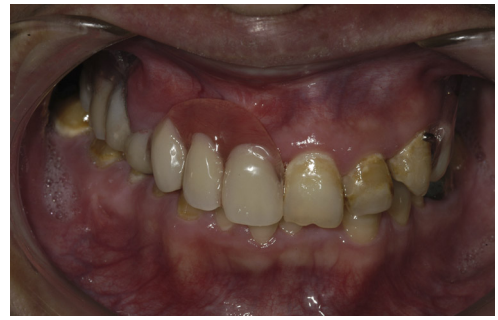

FIGURE 6. Occlusion was rehabilitated by removable partial denture.

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Giant Aneurysmal Bone Cyst of the Mandible

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Abstract: The aneurysmal bone cyst is a type of pseudocysts of the jaw. It is a nonneoplastic lesion of the bone, characterized by replacement with fibro-osseous tissue containing blood-filled sinusoidal or cavernous spaces. The lesion remains a relatively uncommon finding in the facial bones, and the cause and pathogenesis are yet to be elucidated.

Aneurysmal bone cyst was first described as a distinct clinical and pathologic entity by Jaffe and Lichtenstein in 1942. Aneurysmal bone cyst comprises 1.5% of all nonodontogenic cysts of the jaws and 1.9% of all aneurysmal bone cysts of skeleton.

A rare case of giant aneurysmal bone cyst of mandible in a 10-year-old child is presented, which was managed by surgical curettage with a long-term follow-up.

Key Words: Aneurysmal bone cyst, jaw cyst, nonodontogenic cyst, aneurysms of jaw, benign lesion of jaw

Aneurysmal bone cyst (ABC), first described by Jaffe and Lichtenstein in 1942, is a nonneoplastic bone lesion, characterized by replacement with fibro-osseous tissue containing blood-filled sinusoidal or cavernous spaces, which has been reported to affect mainly the long bones of the skeleton.[1] Only 2% of the lesions appear in the jaws.[2]

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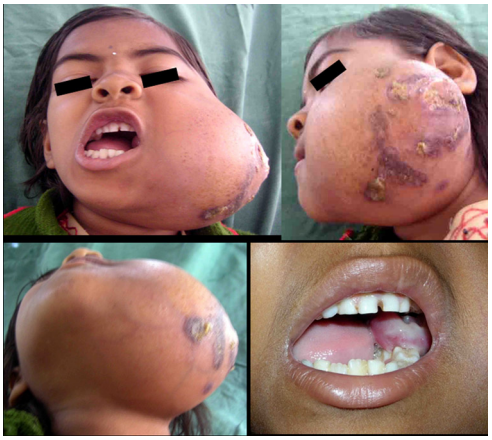

FIGURE 1. Preoperative photographs.

The World Health Organization defines ABC as a benign intraosseous lesion, characterized by blood-filled spaces of varying sizes associated with a fibroblastic stroma containing multinucleated giant cells, osteiod, and woven bone.[3] The pathogenesis is, however, controversial. It has been suggested that the lesion may have a developmental or traumatic origin that it may represent a vascular anomaly or that it may show features of a benign or malignant neoplasm.[4]

The theory that the lesion is the result of secondary change in a primary bone lesion was first suggested by Ewing who described what appears to be an ABC and advanced the concept that it represented a benign giant cell tumor modified "apparently as a result of free communication with large blood vessels." Jaffe advanced the theory that the ABC may result from modification of some other lesion of bone, most of which had been destroyed by hemorrhage. Other authors disputed this theory and subscribed to that proposed by Lichtenstein that the ABC is the result of some vascular disturbance.[5]

We report a rare case of giant ABC of the mandible in a 10-year-old child managed by surgical curettage and review of literature.

CLINICAL REPORT

A 10-year-old female child reported to us with a chief complaint of swelling over the left half of her face and facial asymmetry for 2 months. Other relevant medical and personal histories were not contributory.

Extraoral examination revealed a well-defined swelling of 8 × 7 cm^2 on the left side of the face extending superiorly from the

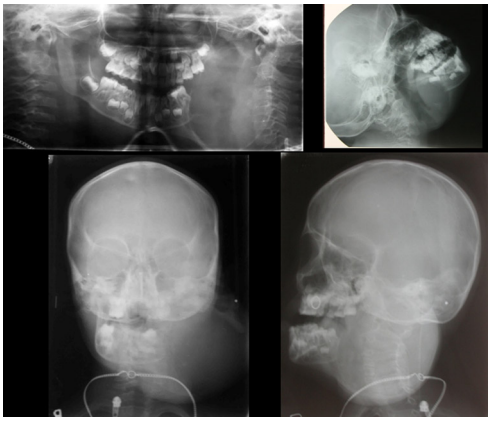

FIGURE 2. Preoperative radiographs.

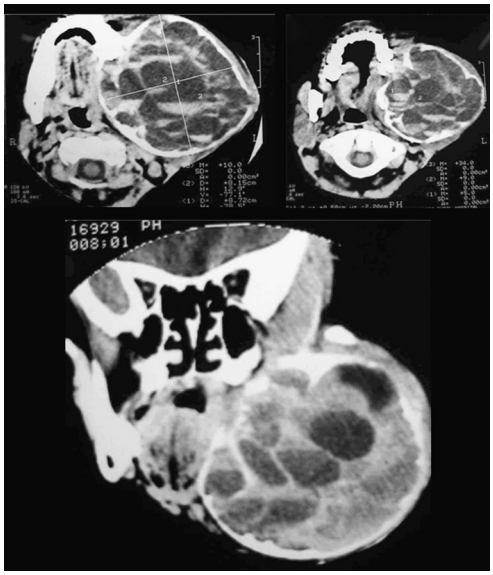

FIGURE 3. Computed tomographic scan.

left lateral canthus of the eye to inferiorly approximately 3 cm below the lower border of the mandible (Fig. 1). The swelling extended from the submental region to approximately 7 cm laterally and anteroposteriorly from the lateral side of the nose to the preauricular region, pushing the auricle distally and superiorly. Overlying skin showed some ulcerations with necrosis, and the skin was discolored. The left nare was elevated, obliterating the nasolabial fold, and there was difficulty in mouth opening with deviation of mouth toward the right side. The swelling was tender, firm, and not fixed to the overlying skin. There was no lymphadenopathy.

Intraorally, swelling was present in the lower left alveolus and vestibular region, extending from the left lower deciduous second molar distally to the anterior border of the ramus. Overlying mucosa was erythematous with sloughing. The swelling was soft to firm, tender with no spontaneous bleeding on palpation. There was displacement of the lower left first permanent molar onto the lingual aspect.

Radiographic examination showed a solitary well-defined thinly corticated radiolucency, extending from the distal aspect of the permanent first molar posteriorly to involve the entire ramus, superiorly up to the maxillary tuberosity region and zygomatic bone and inferiorly approximately 3 cm below the inferior border of the mandible. The radiolucency was extending approximately 2 cm laterally on the left side. Developing tooth buds were present except the tooth bud of the second permanent molar (Fig. 2).

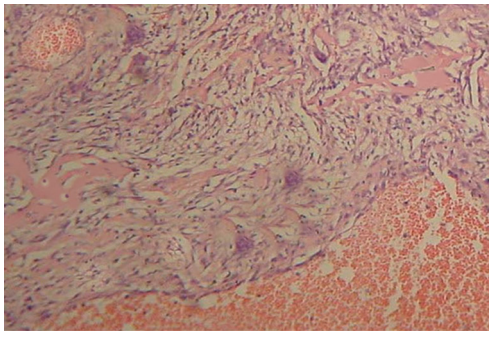

FIGURE 4. Histomicrograph; hematoxylin and eosin stain, original magnification ×40.

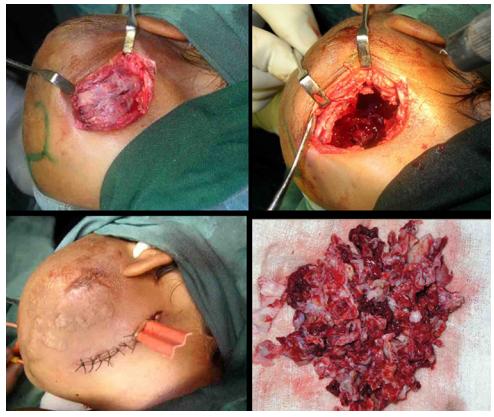

FIGURE 5. Intraoperative photographs.

Findings in the CT scan revealed a large (8.72 × 8.15 cm^2) round heterogenous mass seen involving the mandible on the left side with an expansion of the lesion laterally with multiple hypodense areas within the main lesion suggestive of cystic areas in it (Fig. 3).

Incisional biopsy report revealed ABC (Fig. 4). Surgical curettage was performed using an extraoral approach under general anesthesia (Fig. 5). The postoperative recovery and healing were uneventful. The patient is kept on long-term follow-up with no signs of recurrence (Figs. 6 and 7).

DISCUSSION

The literature contains conflicting reports on the clinical and radiologic features of ABC.[6–8] Most explanations about their cause and pathogenesis have been based on fragmentary evidence gained from case reports and small series of cases. Consequently, ABCs have been variously thought to be neoplasms, developmental errors, and local reactions to injury.

Three stages may be distinguished in the course of the development of ABC[9]:

1. initial phase—with prevalent osteolysis and noncharacteristic appearance.

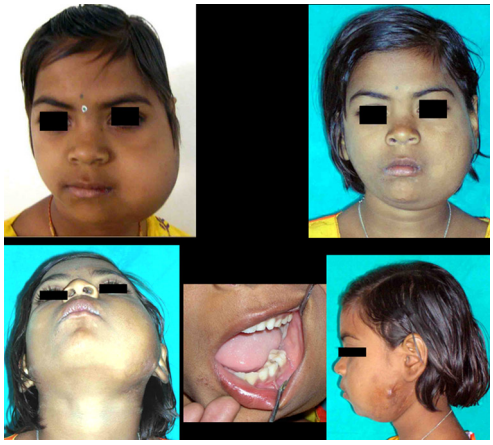

FIGURE 6. Photographs 18 months after surgery.

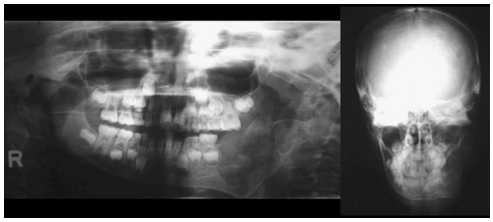

FIGURE 7. Postoperative radiographs.

2. growth phase—with rapid increase of the tumor, expansion of bone, and marked bone destruction. The tumor is not demarcated and septa are indistinct.
3. stabilization phase—with fully developed radiologic syndrome.

Aneurysmal bone cyst may be present at any age; it is most frequently diagnosed in the second decade[2,5] and is said to have a slight female predominance. Aneurysmal bone cyst is reported to occur more frequently in the mandible than in the maxilla, with a ratio of 2.4:1. However, Giddings et al[10] found that the mandible is involved less often than the maxilla.

Clinical presentations of the ABC range from a mild, slowly expanding, semisolid growth, causing slight facial asymmetry, to a rapidly expanding vascular swelling, causing extensive bone destruction and mimicking malignant lesions.[11–13] This lesion does not have a clear clinical specificity.

Despite some reports that ABC may cross the midline, it is rare and only 2 cases have been reported.[2] Goaz and White[7] describe ABC as an expansive, osteolytic process that is manifested as a unilocular radiolucent lesion. They suggest 3 stages in its development, which determine its radiologic features.

1. initial stage characterized by a defined but not corticated lytic area,
2. growth stage, showing an enlarged area of bone destruction, and
3. mature stage, exhibiting bony expansion, cortication, and faint septa coursing through the lesion in a random pattern.

Hernandes et al[14] claim that the radiologic appearance of ABC may "change rapidly" and recommend that, before biopsy, CT and magnetic resonance imaging (MRI) be used in addition to plain film radiography to establish the differential diagnosis.

Kaffe et al[2] found that MRI and CT were helpful in the differential diagnosis because MRI shows the soft tissues and fluid-containing cavities more definitively, whereas the expansive character of the lesion and its thin bony walls are better depicted by CT as was seen in this case.

The characteristics like the sudden growth, cortical destruction, osteoid formation, and tumor-like appearance can easily cause confusion with malignancy. It is important to differentiate the ABC from other pathologic lesions that occur in the maxillofacial region. These include peripheral and central giant cell reparative granuloma, traumatic bone cyst, brown tumor of hyperparathyroidism, myxoma, fibrous dysplasia, desmoplastic fibroma, fibrous histiocytoma, hemangioma, osteogenic sarcoma, globulomaxillary cyst, hemangioendothelioma, and hemangiopericytoma.[12,15] However, definitive diagnosis requires histopathologic examination of the surgical specimen.

Aneurysmal bone cyst should be considered as a distinct clinical entity, despite its association with other primary bone lesions, because additional diagnostic evaluation and surgical precautions are then needed. Various treatment options have been described for ABCs, of which curettage and en bloc excisions are the methods of choice.[16] Other modalities include radiation, cryotherapy, percutaneous intralesional injection, calcitonin therapy, and embolization. There are reports of embolotherapy (as an adjunct to surgical treatment or as a definitive treatment modality) being used to treat ABCs of the spine, long bones, and pelvis. Intralesional embolotherapy has been used as a definitive treatment wherein the lesions show complete involution after embolization[17] or as an adjunct treatment wherein embolotherapy limits blood loss during surgery.[18] There is no literature regarding the use of this treatment modality for mandibular lesions.

Although cryotherapy[19] and radiotherapy[20] have supplemented curettage in decreasing the recurrence rate, the use of the latter is strongly discouraged because it is likely to induce sarcomatous change in the irradiated bone.[21] The treatment modality most likely to have a complete cure is en bloc resection,[5] but this is restricted to large and recurrent lesions owing to the morbidity of the procedure.

Beisecker et al[19] reduced the recurrence rate by supplementing curettage with cryotherapy. Supplementation of curettage by radiotherapy has also been used; however, Tillman et al[21] reported sarcomatous change after radiotherapy. Radiation should be reserved for those cases in which surgical removal is not feasible, and the recommended dosage seems to be between 600 and 2000 rad. The risk of postradiation sarcoma increases with larger or excessive dosages of radiation.[20]

In conclusion, it is believed that ABC of the jaws is best treated by surgical curettage and resection in selected cases.[16] Recurrence seems to occur within 1 year after initial treatment, and incomplete removal is quoted as the most common reason for recurrence.[12] It is therefore recommended for scrupulous surgical curettage and conscientious follow-up for all cases of ABC of the jaws.

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Preventing Iatrogenic Injury From Inadvertent Intracranial Migration of a Urinary Foley Catheter While Controlling Profuse Epistaxis After Severe Craniofacial Trauma

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Abstract: The massive nasopharyngeal bleeding that may accompany complicated comminuted fractures of the craniofacial bones can be controlled by pressure tamponade using an inflatable urinary Foley catheter. However, inadvertent intracranial catheter penetration poses a serious risk in such situations. Management of a relevant case is described, and a simple preventive measure is suggested.

Key Words: Iatrogenic intracranial introduction, craniofacial trauma, urinary Foley catheter, nasal packing

Nasal insertion is traditionally contraindicated in trauma victims with extensive craniofacial bone fracture but continues to be performed in selected situations such as severe craniofacial trauma with massive oral-nasal bleeding. There are few approaches to manage profuse bleeding in these situations. Posterior nasal packing with a urinary Foley catheter in a patient with complicated craniofacial trauma is an effective, rapid, and nonsurgical alternative for the management of posterior epistaxis. In particular, insertion of a urinary Foley catheter with an inflatable balloon is the most popular and time-saving approach.[1–3] Like other invasive procedures, however, use of an inflatable urinary Foley catheter in this situation is not free of risks or complications. Although the potential for intracranial migration of a Foley catheter during posterior nasal packing exists, clinical reports describing such migration are rare.[4,5] This report describes a case of such migration and a procedure to manage the resulting complications.

CLINICAL REPORT

A 45-year-old man was sent to our hospital after a severe motorcycle crash. Active bleeding from his nasal and oral cavities accompanied by massive nasopharyngeal blood clots were noted. In addition, multiple facial contusions and deformity were observed. Initially, craniofacial trauma with cranial base injury was suspected. The patient was comatose with a Glasgow Coma Scale score of 6 and was also in shock. After endotracheal intubation, posterior nasal packing with a Foley catheter and an inflatable balloon was performed to stop the bleeding.

A computed tomographic (CT) scan of the brain revealed that the catheter was lodged in the left basal ganglia through the fractured anterior ethmoid sinus (Fig. 1). After removal of the Foley tube, a second set of CT scans showed minimal intracranial hematoma. The nasal bleeding was stopped after reinsertion of the catheter. The scans also revealed the presence of a Le Fort type III craniofacial fracture and left frontal-temporal subdural hemorrhage.

After emergent brain surgery and administration of empirical antibiotics, the patient regained consciousness and recovered progressively without significant neurologic deficits. A Glasgow Outcome Scale score of 4 was obtained at 3 months after injury.

DISCUSSION

Complicated craniofacial injuries are often followed by cranial base fractures. The Le Fort type III fracture is characterized by fractures of the central and lateral midface. This fracture, which usually results in separation of the midface from the cranial base, involves the nasomaxillary suture and lamina papyracea medially, the pterygoid plates, and the superior orbital fissure.[6] In a study at the Johns Hopkins Medical Institutions, 268 Le Fort fractures were treated and followed; 107 (39%) of patients with these fractures had a

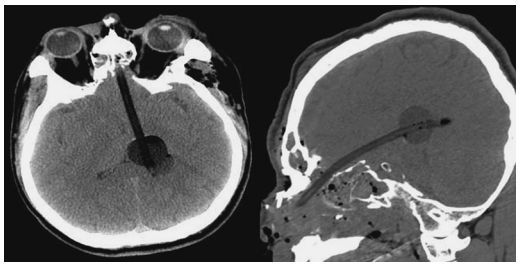

FIGURE 1. Axial (right) and sagittal (left) brain CT scans revealing a severe cranial base fracture, passage of the inflated Foley catheter through the anterior ethmoid sinus, and lodging of the catheter into the brain parenchyma.

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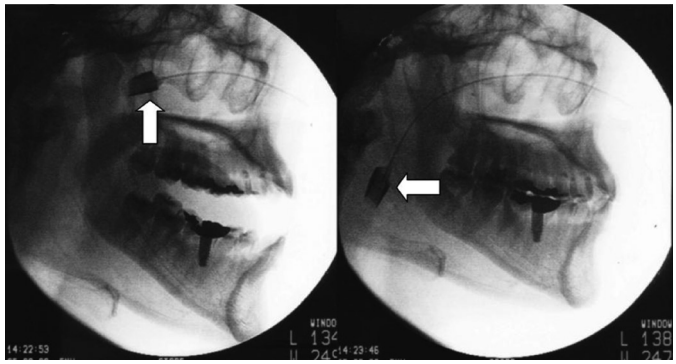

FIGURE 2. Use of a Foley catheter injected with 1 mL of contrast medium (Omnipaque) to confirm the position of the catheter and balloon tip. The catheter is clearly identified (arrows) by the C-arm (lateral view). The trajectory of the Foley tip within the nasal cavity is visible approximately 10 cm from the nostril (left). The balloon is located in the oropharynx at a distance of 13 cm from the nostril (right). These photographs were obtained from a patient of the second author (S.-T. Wei).

nasoethmoidal fracture, which is the major of cause of misdirection of nasal catheter insertion.[7]

The use of a Foley catheter in the treatment of severe posterior epistaxis is an old and well-established technique.[2,3,8] A Foley catheter with an inflated balloon is the most commonly used device and is recommended because most clinicians are familiar with its use and can obtain it easily. Despite the fact that nasal packing with this device represents a fast and effective approach, stimulation of the nasopulmonary reflex by balloon pressure can lead to hypoxemia, apnea, and cardiac arrhythmia. Palatal necrosis and subsequent perforation can also occur because of overinflation of the balloon. Furthermore, balloon deflation failure, rebleeding, and nasal alar necrosis have been observed during securing of the catheter.[1,5] If conventional techniques fail to control posttraumatic oronasal hemorrhage, which can result in life-threatening exsanguination, transarterial embolization of the hemorrhaging artery is reported to be an effective choice for hemostasis.

Intracranial placement of a Foley catheter is much less commonly reported in the literature compared with placement of a nasogastric tube. However, the latter is a routine maneuver in daily practice that would not be commonly considered for a patient with severe craniofacial trauma. Although uncommonly observed, this bony barrier may be fractured owing to the rigidity of the nasogastric tube. The consequences of inadvertent nasogastric tube positioning within the cranial cavity are serious, with a reported mortality of 64%, and severe complications can occur including hemiparesis, blindness, anosmia, and development of persistent cerebrospinal fluid fistulas.[9,10]

The best procedure for removing a displaced intracranial tube is debated. Some authors recommend craniotomy with removal of the previously segmented tube under direct visualization. Others advocate retrieval through the nose. No scientific evidence is presently available to suggest that either technique offers any prognostic advantage.[10] Regardless, the best approach is to use all appropriate measures to prevent this complication from occurring.

Several procedural modifications for the prevention of intracranial Foley catheter migration have been described. For example, Woo et al[5] suggested use of a large-sized catheter in a straight direction parallel to the floor of the nasal cavity and identification of the tip before inflation of the balloon. An insertion length of less than 13 cm was considered the most important modification for prevention of migration because the average distance from the nostrils to the oropharynx is approximately 11 to 13 cm.

Because of its radiolucency, identification of a Foley catheter on a radiograph is often difficult. Once misplacement of the catheter has been established, an emergency CT study is essential. To confirm the appropriate position of the balloon tip, we suggest the use of a Foley tube filled with 1 mL of contrast medium (Omnipaque, 300 mg/mL, GE Healthcare, Carrigtohill, Ireland) before nasal packing. Initially, the first 10 cm length of Foley catheter should be inserted from 1 nostril. Identification of the trajectory and position by the C-arm or portable x-ray is mandatory for the prevention of inadequate upward migration. After confirmation that the trajectory of the Foley tube is correct, further advancement to a length of 13 cm from the nostril should allow the tube to reach the oropharynx (Fig. 2). Routine implementation of these simple measures should prevent potentially serious iatrogenic complications from occurring during the management of profuse epistaxis in the setting of severe craniofacial trauma.

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Benign Nodular Hidradenoma of the Face

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Abstract: A very rare case of nodular hidradenoma with an atypical clinical presentation of the face of a 70-year-old woman is presented. Although this rare benign tumor has the potential of local recurrence and malignant transformation, it is commonly reported in the dermatology, pathology, and oncology literature, with limited clinical information. The purpose of this report was to emphasize the clinical features of this lesion for plastic surgeons.

Key Words: Nodular hidradenoma, clear cell hidradenoma, acrospiroma

Nodular hidradenoma is a rare benign sweat gland tumor arising from the apocrine glands. This tumor has a low incidence of neoplastic transformation and metastasis.[1] The differential diagnosis of the malignant form of the tumor is extremely difficult not only because of their benign appearance but also because of their similarity to other cutaneous lesions.[2] Some of the excised lesions initially diagnosed as a benign tumor may be proved to be a malignant transformation once reexamined after the recurrences and the metastasis have been occurred.[3] Here, a case of a nodular hidradenoma with atypical demonstration on the face of an old woman is presented.

CLINICAL REPORT

A 70-year-old woman with an enlarged mass on the left malar area of the face, diagnosed as basal cell carcinoma or squamous cell carcinoma without histologic proof, was referred to our clinic by the dermatology department of our hospital. The patient stated that she had palpated a pea-sized nodule on her left malar area 1 year previously, becoming larger especially in the last 2 months. Physical examination revealed a 2.5-cm mass with crusty irregular surface (Fig. 1). The lesion was hard, and there was lack of peripheral induration.

Total excision of the lesion with 2-cm clear margins and reconstruction with a local flap under general anesthesia were performed owing to the malignant appearance of the tumor and advanced age of the patient. The patient has been discharged on the first postoperative day, and the postoperative period was uneventful (Fig. 2).

Histopathologic evaluation of the lesion demonstrated polyhedral cells with clear cytoplasm surrounded by cystic spaces and ductal structures (Fig. 3). Squamoid differentiation, keratinization, and trabecular structures, which were anastomosing together, were also seen in some areas (Fig. 4). The histopathologic diagnosis was benign nodular hidradenoma with clear surgical margins. The pathologic reexamination of the lesion confirmed the initial diagnosis.

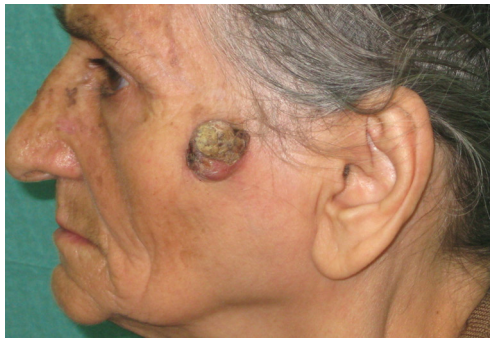

FIGURE 1. Preoperative view of the lesion.

DISCUSSION

Nodular hidradenoma is an uncommon benign tumor of the distal portion of sweat glands and is also known as acrospiroma, clear cell hidradenoma, dermal duct tumor, hidroacanthoma simplex, or poroma.[4] This tumor is most commonly seen on the head, neck, trunk, and extremities and is more commonly reported in females.[1]

The lesion appears usually as a small, firm, solitary dermal nodule with intact overlying skin. Some tumors may have ulceration on the surface together with serous fluid leakage.[5] Macroscopically benign nodular hidradenomas are well demarcated compared with the larger and asymmetric nodular hidradenocarcinomas.[3] The mean age at presentation of the benign nodular hidradenoma is 37.2 years.[6] However, malignant nodular hidradenoma occurs in the sixth and seventh decades.[2]

The case presented here had a serous leakage and ulceration on the surface. The lesion was multilobulated, irregular, and crusty. Both the appearance of the tumor and the age of patient were more likely to be a part of a malignant skin tumor.[6]

Although nodular hidradenomas regarded as a benign lesion, they can reoccur and might have a malignant transformation with the rate of 6.7%.[6] In fact, most of the malignant hidradenomas are reported to be malignant from their inception[5] but are incorrectly

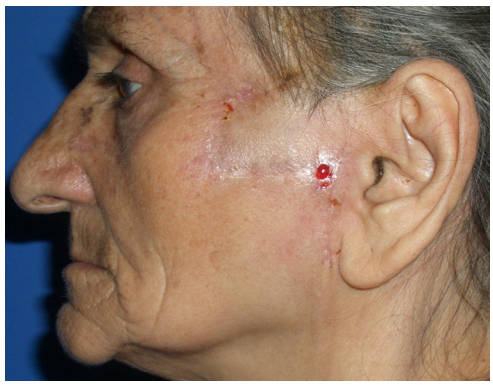

FIGURE 2. Postoperative second week view of the lesion.

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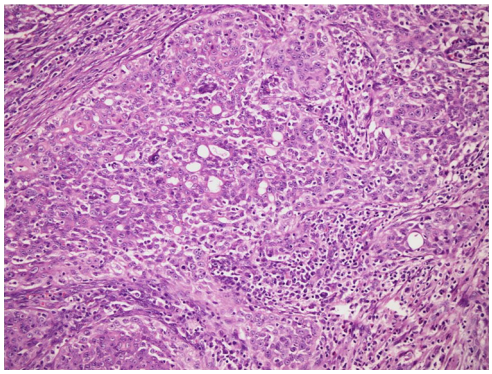

FIGURE 3. Ductal differentiation areas in the tumor tissue (hematoxylin and eosin, original magnification ×200).

evaluated as benign in their initial pathologic examination.[2] Malignant nodular hidradenoma has an aggressive behavior with local recurrence (50%) and metastases (60%).[2] The survival of this tumor is generally poor, with a 5-year disease-free survival of less than 30%.[3] These information clearly demonstrate that even the pathologic evaluation reveals a benign nodular hidradenoma and that the tumor may reoccur and become an aggressive malignant form.

Primary excision[5] or Mohs micrographic surgery[2] may be used for the treatment of the tumor. In cases of malignant nodular hidradenoma, resection with at least 2 cm of clear margins was suggested and prophylactic lymph node dissection is advocated by many authors owing to the high propensity of regional lymph node spread.[2] By treating skin tumors with malignant appearance, an incisional biopsy can be considered before the definitive treatment because the result of the biopsy can change the direction of the planned therapy.

CONCLUSIONS

Nodular hidradenoma is a rare tumor with a risk of local recurrence and malignant transformation. A 2.5-cm-sized lesion, which was diagnosed as a nodular hidradenoma of the face with an atypical presentation and possible suspicion of malignant features, is presented. We believe that these lesions, especially if they are observed in elderly patients and if they have a malignant appearance like in our case, have to be resected with possible wide clear margins, and the patient has to be followed up with short time intervals owing to the risk of local recurrence and malignant transformation.

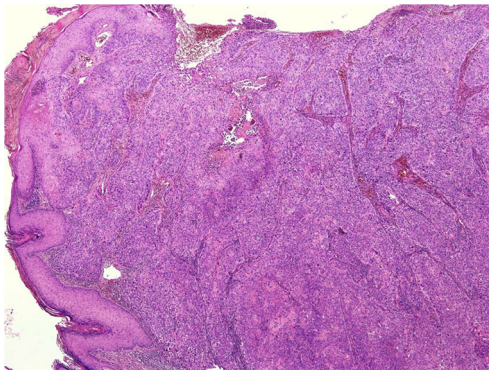

FIGURE 4. The tumor has a protruding growth pattern with ulceration on the surface. The tumor tissue has anastomosing trabecular structures near the dermis.

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Unexpected Tumor Incidence in Surgically Removed Unilateral and Bilateral Nasal Polyps

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Objective: The objective of the study was to investigate the postoperative tumor incidence on routine histopathologic examination of surgical specimens in patients who underwent endoscopic sinus surgery with the preoperative diagnosis of unilateral or bilateral nasal polyposis.

Methods: Patients who underwent endoscopic sinus surgery with the preoperative diagnosis of unilateral or bilateral nasal polyposis between 2000 and 2009 were included in the study as the 2 separate groups. In both groups, tumor incidence that had been noticed on routine postoperative histopathologic examination was recorded retrospectively. Patients who had a preoperative diagnosis other than nasal polyposis, determined on biopsy, were excluded.

Results: Of 251 patients included, 197 had the preoperative diagnosis of bilateral nasal polyposis, and 54 had unilateral nasal polyposis. No tumor was diagnosed on postoperative histopathologic examinations in patients with preoperative bilateral nasal polyposis.

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Seven patients (12.96%) with the preoperative diagnosis of unilateral nasal polyposis had tumors on postoperative histopathologic examinations. Two of these 7 patients had malignant tumors that were reported preoperatively by intranasal biopsy as inflammatory polyps.

Conclusions: Diagnosis of a neoplasia is an extremely rare situation, unless there are special findings of tumor in patients with preoperative diagnosis of bilateral nasal polyposis. On the other hand, histopathologic examinations should be carried out in every case operated with preoperative diagnosis of unilateral nasal polyposis, as the tumor incidence is higher.

Key Words: Nasal polyp, tumor, endoscopy, surgery, histopathology

Nasal polyposis is a chronic inflammatory mucosal disease of the nasal cavities and paranasal sinuses. Prevalence of nasal polyposis is approximately 1% to 4% in the overall population.[1] It is a common cause of nasal obstruction and chronic sinusitis and requires surgical treatment frequently. An accurate history and physical examination including nasal endoscopy, significantly contribute to the diagnosis of nasal polyposis. Computed tomography (CT) of the paranasal sinuses also supports the preoperative diagnosis. However, there may be unexpected diagnosis in postoperative histopathologic examinations of these patients. Benign and malignant tumors of the nasal cavities and the paranasal sinuses always should be kept in mind in the differential diagnosis.[2,3]

Essential treatment of nasal polyposis is composed of endoscopic sinus surgery and postoperative steroid administration.[4] During surgery, the concept of being suspicious in the diagnosis of unilateral nasal polyps has wide acceptance.[5] Nevertheless, routine postoperative histopathologic examination in every patient who undergoes surgery for the treatment of nasal polyposis is still a controversial issue.[2,6]

The aim of this study was to investigate the frequency of tumor on routine postoperative histopathologic examination in patients who were operated on with the preoperative diagnosis of bilateral and unilateral nasal polyposis and to find out whether histopathologic examination of all surgically removed nasal polyps is a necessity or not.

CLINICAL REPORT

Patients who underwent endoscopic sinus surgery with the preoperative diagnosis of nasal polyposis in our department between 2000 and 2009 were included in the study. Medical records of patients, including preoperative and postoperative examination notes, surgical reports, pathologic examination request forms, and histopathologic examination reports, were evaluated retrospectively. Patients with the diagnosis other than nasal polyposis (eg, granulomatous nasal diseases or tumors) diagnosed by preoperative biopsy because of appearance suggestive for intranasal soft tissue were excluded from study.

Preoperative diagnosis of nasal polyposis was based on findings of nasal endoscopy and paranasal CT. Patients were divided into 2 groups as cases with bilateral and unilateral nasal polyposis in regard to their preoperative diagnosis. Preoperative nasal biopsy was performed in those patients with suspected tumor appearance of intranasal soft tissue and with the findings of bone erosion on paranasal CT. All the patients underwent endoscopic sinus surgery. If a suspicion of tumor arose intraoperatively, a biopsy was taken for frozen-section examination, and surgery was extended accordingly in case of detection of malignity. All samples, taken during surgery, were fixed with 10% neutral-buffered formalin and sent to the department of pathology for histopathologic examination. After routine processing and embedding, sections from paraffin blocks were obtained and stained with hematoxylin-eosin for microscopic examination. Incidental tumors detected during postoperative histopathologic examination in the 2 different groups with preoperative diagnosis of unilateral and bilateral nasal polyposis were recorded.

RESULTS

A total of 251 patients, 174 male and 77 female patients, were included in the study. Mean age of patients was 39.3 (SD, 1.6) years (range, 5–72 years).

Of 251 patients, 197 (78.4%) with a preoperative diagnosis of bilateral nasal polyposis and 54 (21.6%) with unilateral nasal polyposis underwent endoscopic sinus surgery. Inflammatory polyp was detected on the postoperative histopathologic examination in all patients who underwent surgery with the preoperative diagnosis of bilateral nasal polyposis. On the other hand, postoperative histopathologic examination revealed tumors in 7 (12.96%) of the patients who were operated on with the preoperative diagnosis of unilateral nasal polyposis (Table 1).

The most frequent tumor reported postoperatively was inverted papilloma (n = 5). These patients were reoperated on under endoscopic vision, and the surgery was extended to obtain clear surgical margins. No local recurrence of inverted papilloma was detected in 4 years' follow-up of these patients.

Malignant tumors were detected on postoperative histopathologic examination in 2 patients. These were squamous cell carcinoma (Fig. 1) and chondrosarcoma of which preoperative intranasal biopsy was reported as inflammatory polyps. Intraoperative biopsies for frozen-section examination were carried out by taking into account of radiologic findings and intraoperative tumor image of soft tissues. The operation was extended in these patients as endoscopic anterior cranial base surgery and endoscopic medial maxillectomy after obtaining a positive report of malignancy by frozen-section examination. The patient with squamous cell carcinoma underwent postoperative radiation therapy because of microscopic intimacy of tumor to the periorbita, and no local recurrence was seen in the 5-year postoperative follow-up. As the surgical margins of chondrosarcoma were clear histopathologically, no postoperative

TABLE 1. Postoperative Histopathologic Findings in Patients With the Diagnosis of Unilateral and Bilateral Nasal Polyposis

Histopathologic Findings	Preoperative Diagnosis of Bilateral Nasal Polyps (n = 197), n (%)	Preoperative Diagnosis of Unilateral Nasal Polyps (n = 54), n (%)
Inflammatory polyp	**197 (100)**	**47 (87)**
Inverted papilloma	0	**5 (9.2)**
Squamous cell carcinoma	0	**1 (1.9)**
Chondrosarcoma	0	**1 (1.9)**

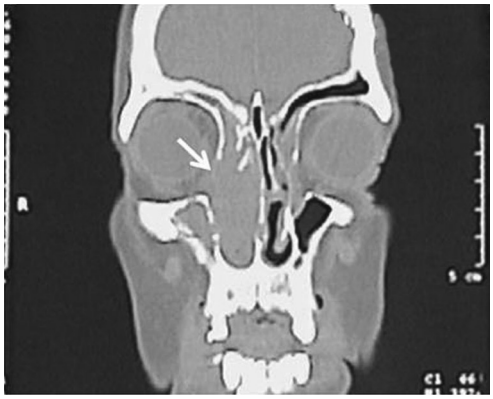

FIGURE 1. Computed tomography images of the patient with squamous cell carcinoma reported as inflammatory polyp by preoperative biopsy. Arrow indicates bone erosion of the lamina papyracea.

radiotherapy was given. This patient was followed up for 9 years without postoperative local recurrence or distant metastasis.

DISCUSSION

Endoscopic sinus surgery is one of the most frequently used methods in the treatment of nasal polyposis. Differential diagnosis between nasal polyps and tumors is always needed for the planning of an appropriate treatment. Preoperative diagnosis of nasal polyps is commonly based on its endoscopic appearance and CT findings. However, the final diagnosis of tumor and nasal polyp can be done by histopathologic examination. Yet, it is still controversial if the tumor incidence in patients who undergo surgical treatment with the preoperative diagnosis of nasal polyposis is as high that requires routine histopathologic examination of tissue samples in every case.

Romashko and Stankiewicz[6] reported different results of histopathologic examination from preoperative diagnosis in only 0.2% of cases that underwent endoscopic sinus surgery because of nasal polyposis and sinusitis. In their study, sinonasal lymphoma was detected in 1 patient and inverted papilloma in another one.[6] As a result, the authors reported that histopathologic examination is not required for every surgical samples of nasal polyps, taken during endoscopic sinus surgery, but it is recommended for cases with intraoperative findings suggestive for a tumor and those ones with unilateral sinus opacity on CT of the paranasal sinuses.[6] However, in a study of Irfan and Shamim,[2] including 95 patients who underwent endoscopic sinus surgery with the preoperative diagnosis of nasal polyposis, the rates of benign nasal mass and malignant tumor were as high as 5.3% and 1.1%, respectively, on postoperative histopathologic examination of all surgical tissue, and they advocated postoperative routine histopathologic examination of surgically removed polyps. Garavello and Gaini[7] found 0.37% of different diagnosis other than nasal polyps on postoperative histopathologic examination in a large patient group who were operated on for nasal polyposis, and they reported that further studies were required to determine whether histopathologic investigation in these patients was cost-effective.

Postoperative histopathologic findings, obtained in relevant studies on this subject, have been interpreted differently in several studies. These varieties may be due to evaluation of histopathologic findings of bilateral and unilateral nasal polyposis in a single group. Therefore, we divided patients into 2 groups as with bilateral and unilateral nasal polyposis, based on preoperative diagnosis, and compared postoperative histopathologic results in these groups. In our study, postoperative histopathologic examination of surgical samples in patients with the preoperative diagnosis of nasal polyposis revealed no tumor. Nasal tumors generally emerge as unilateral and extend to the other side of the nasal passage only in well-advanced stages. Although advanced-stages tumors involve the nasal passage bilaterally and may be confused with nasal polyp initially by its appearance, accompanied findings besides intranasal mass generally alert the surgeon for taking preoperative biopsy. Therefore, if there is no specific finding of tumor on intranasal endoscopic examination or on radiologic studies, routine postoperative histopathologic examination may not be necessary in cases with the diagnosis of bilateral nasal polyposis.

On the other hand, the rate of postoperative incidental tumors that are recognized by histopathologic examination of surgical materials in cases that were operated with the preoperative diagnosis of unilateral nasal polyposis is higher.[8,9] This ratio was 12.96% in our study. In other words, intranasal tumors generally appear unilaterally unless they reach to advanced stages. Therefore, surgeons always should be suspicious in patients with unilateral nasal polyposis even if there is no tumor appearance on preoperative examinations. Treatment modality should be reviewed and rearranged in case of a tumor diagnosis reported by the postoperative histopathologic examinations. In our study, 5 cases had incidental inverted papilloma on postoperative histopathologic examination and were reoperated to obtain clear surgical margins.

Preoperative biopsy should definitely be performed to plan the appropriate treatment in patients with preoperative tumor such as findings of nasal polyposis.[10] However, results of preoperative biopsies of intranasal masses are not always the same as the postoperative histopathologic findings.[11] In these patients, chronic inflammation caused by the tumor may result in the development of additional nasal polyps, which mask the real pathology. Biopsy samples taken from the nasal passage in such cases may give rise to a false-negative diagnosis. That was the situation experienced in our 2 cases. Postoperative malignancy was encountered, even though inflammatory polyps were reported in preoperative biopsy. Bone erosion on CT should be a warning for the tumor. If there is suspicion of a tumor, intraoperative biopsies should be taken for frozen-section analysis. In our study, the malignant tumor was reported by frozen-section examination of intraoperative tissue samples, and surgical plan was changed accordingly.

In conclusion, diagnosis of a neoplasia is an extremely unlikely situation, unless there are special findings of tumor in patients with preoperative diagnosis of bilateral nasal polyposis. On the other hand, preoperative and postoperative histopathologic examinations should be carried out even if there is no clear preoperative tumor finding in cases operated with a preoperative diagnosis of unilateral nasal polyposis, as the tumor incidence is higher. However, preoperative intranasal biopsies sometimes may give false-negative results because of nasal polyps that develop anterior to the tumor.

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Intraorbital Isolated Mucocele

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Abstract: A 55-year-old man was presented with an orbital mass. The magnetic resonance imaging revealed that the dimensions of the mass were 29 × 19 × 17 mm. The most probable diagnosis with the magnetic resonance imaging findings was dermoid cyst. The mass was a cyst and marsupialized by functional endonasal endoscopic sinus surgery without any complication. Intraoperative findings of the mass were thought to be a hydatid cyst, but the diagnostic tests were negative, and the pathologic result was consistent with mucocele.

Key Words: Intraorbital mucocele, intraorbital mass, endoscopic removal

Mucoceles are inflammatory, cystic lesions of the paranasal sinuses containing a mucous membrane.[1] The most common paranasal sinus mucoceles are found in the frontal and ethmoidal sinuses; they are infrequent in the sphenoid sinus, and they occur rarely in the maxillary sinuses.[2]

The characteristic feature of mucocele on imaging is the thinned remodeling of the bony wall of the affected paranasal sinus. The locations of the mucoceles are usually paranasal sinuses, but because of the anatomic close relations, periorbital mucoceles with the involvement of the nearest paranasal sinus are not very rare. However, isolated intraorbital mucoceles are not very common.[3]

CLINICAL REPORT

A 55-year-old man was referred with a swelling in his left medial canthus for 3 months. He had a minimal exophthalmos. He had no history of trauma or positive history sinus pathology. Although he said that his vision was decreased, according to eye consultation there were no changes of his vision, and there was a slight proptosis. His orbital magnetic resonance imaging showed that there was a mass at the anteromedial part of the left orbit, which compresses and displaces the medial rectus superomedially. The dimensions of the mass were 29 × 19 × 17 mm, and in T1-weighted imaging series, the anterior part of the mass was hyperintense because of fat, and in T2-weighted series, it showed solid components that enhanced contrast, and the mass had very well-defined margins; with these findings, the most probable radiologic diagnosis was dermoid cyst (Fig. 1). There is a slight exophthalmos in the left orbit. He underwent excision of the left orbital mass endoscopically under general anesthesia. The cyst wall was very fragile, and cyst contents came down to the nasal cavity. It was observed that there was no communication between the cyst and the adjacent sinus. Because of the feature of the cystic components, the authors thought that this might be hydatid cyst and cleaned the area with hypertonic saline, because Turkey is endemic for hydatic cyst.[4] The remnants of the cyst were excised safely. There was no complication postoperatively.

Systemically administered albendazole was started, and the patient was scanned for hydatid cyst by abdominal ultrasonography and

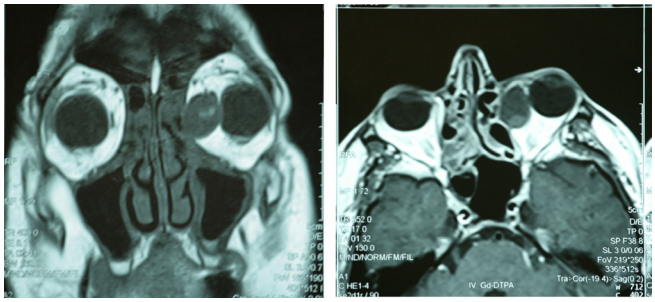

FIGURE 1. Preoperative orbital magnetic resonance imaging of the patient. There is a mass in the right intraorbital part of the patient. The dimensions of the mass were 29 × 19 × 17 mm, and in T1-weighted imaging series, the anterior part of the mass was hyperintense because of fat, and in T2-weighted series, it showed solid components that enhanced contrast, and the mass had well-defined margins.

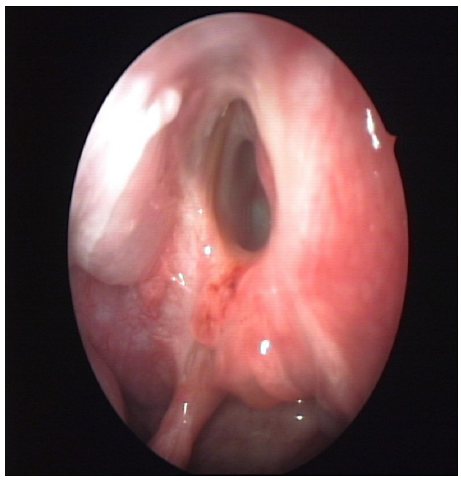

FIGURE 2. Postoperative endoscopic image of the right osteomeatal complex and orbital wall.

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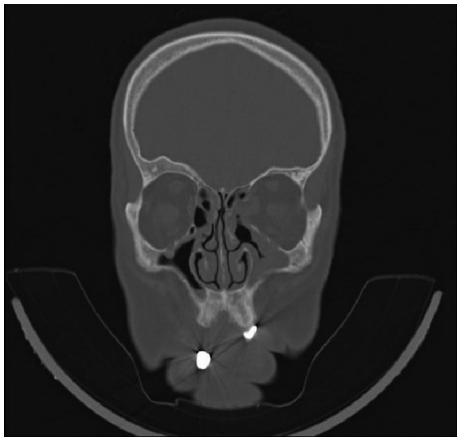

FIGURE 3. Paranasal sinus computed tomography of the patient 1 month after the surgery. The intraorbital structures are normal, and there is a lack of bone at the periorbital wall.

Echinococcus immunoglobulins. There was no evidence for hydatid cyst.[5] The pathologic examination revealed the mucocele, so albendazole treatment was ceased.

DISCUSSION

Orbital mucoceles may originate from trauma, inflammation mass lesions, or the anatomic anomalies that obstruct the paranasal sinuses. Mucoceles are mostly derived from accidental or iatrogenic trauma. As the anatomy of orbital region has close proximity with the brain and eye, there is a great risk of morbidity and mortality.[6]

Mucoceles are epithelial-lined and mucus-filled sacs. They have benign properties, but they can gradually expand through bony resorption. This bony resorption may lead to local expansion. Expansion of the mucoceles may give result to headache, orbital swelling, diplopia, and other visual symptoms.[7]

The differential diagnosis includes abscess, intraorbital hematoma, dermoid cyst, hydatid cyst, lacrimal tumor or cyst, and lymphangioma. In our patient, as Turkey is endemic for echinococcosis, the diagnosis of a hydatid cyst was considered intraoperatively; the operation area was washed by hypertonic saline, and systemic albendazole treatment was started after the operation.[4] But after the diagnostic tests, it is thought to be unlikely because there was no hepatic cyst, and indirect hemagglutination assay test for cystic echinococcosis was negative as well.[5]

The pathologic result was confirmed as mucocele. As we questioned the patient, there was no history of trauma, operation, infection, or any other predisposing factor. It is important that there may be no major or minor predisposing factor for the mucoceles.[3,6,7]

In this case, the orbital structures were intact, and there were not any anatomic preformed pathways, so the orbital mucocele was isolated from other paranasal sinuses. These types of cases are rare, as Asamoto et al[3] reported before. Although there was no history of trauma in our case, there is usually a history of minor trauma with these types of cases mentioned. Rodríguez-Marco et al[8] hypothesized the ectopic mucinous glands, which appeared by the development of the optic canal; this is unlikely in our patient because of the localization of the mucocele.

Intranasal marsupialization of mucoceles was presented by Horwarth in 1921, and it has been used by rhinologists since then in appropriate cases.[9] Although the most suggested treatment is complete removal of the mucocele by endoscopic surgery or open technique according to the localization and dimensions of the mucocele in a series of 16 patients with paranasal sinus mucoceles, marsupialization was used, and no recurrence was observed in the follow-up period.[10]

In our case, as the dimensions were not very small, it was not possible to completely remove the mucocele endoscopically, but marsupialization, without any complication, was done successfully; his vision and eye movements were normal, and after 1 month, the patient is free of disease (Figs. 2 and 3).

CONCLUSIONS

The diagnosis of isolated orbital mucoceles might be a challenging issue in differential diagnosis. But we recommend the endoscopic approach at the first step (if the mass is not aggressive or massive) for removal and the pathologic diagnosis. The outcomes of the endoscopic approach are satisfactory, and morbidity is less than external approach.

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Pituitary Duplication With Nasopharyngeal Teratoma and Cleft Palate

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Abstract: Pituitary gland duplication is a rare malformation of unknown cause that is often associated with a nasopharyngeal teratoma, among other secondary malformations. This clinical report describes a case of pituitary gland duplication with a nasopharyngeal teratoma, cleft palate, and hypothalamic hamartoma, as well as the surgical management of this patient. This case also raises the question of whether the nasopharyngeal teratoma is the cause of the pituitary

duplication above and the cleft palate below or whether it is a result of the primary duplication of the notochordal process. Various theories are presented in an attempt to answer this question, but the exact cause of these malformations remains equivocal. Future research in this topic may elucidate the answer to this question.

Key Words: Pituitary gland duplication, hypophyseal duplication, nasopharyngeal teratoma

Pituitary gland duplication is a rare malformation, with approximately 40 reported cases documented in the medical literature.[1–19] Aside from a poor survival rate, pituitary gland duplication usually contains several secondary malformations such as cleft palate, agenesis of the corpus callosum, abnormalities of the circle of Willis, hypothalamic hamartoma, and an abnormality of the dentate nucleus.[16] On the other hand, duplication of the pituitary gland can also be an isolated finding.[13]

Among the cases of pituitary duplication containing secondary malformations, an association has also been made with the presence of nasopharyngeal teratoma. This article raises the question of whether the teratoma is the cause of the pituitary duplication above and the cleft palate below or whether it is a manifestation of the primary duplication of the notochordal process. We report a case of congenital nasopharyngeal teratoma in association with pituitary duplication, as well as the surgical management of the associated cleft palate.

CLINICAL REPORT

A 3710-g white female infant was referred to the craniofacial surgery service on day 3 of life because of feeding difficulties secondary to a large nasopharyngeal mass and cleft palate deformity. She was born to a 29-year-old woman, G1P1A0, at 38 weeks after a complication- free pregnancy and delivery. APGAR scores were 8/9 at delivery. The family history was significant for epilepsy in two paternal uncles and Down's syndrome in one paternal uncle. A physical examination of the patient revealed a normal external facial examination. The intraoral examination revealed a soft, nodular mass covered with fine hair protruding down through the middle third of the vomer (Fig. 1). This mass measured approximately $3 \times 2 \times 2$ cm^3 and was confluent with the vomer, which was hypoplastic posteriorly. There was a bilateral postincisive cleft palate, and the anterior vomer was not contiguous with the premaxillary segment.

Computed tomographic scans demonstrated the nasopharyngeal mass (which involved the vomer as well as the nasal septum) extending through a large defect in the hard palate. It was determined to contain both lipomatous and ossific components (Fig. 2). The nasopharynx was partially obliterated by the mass, which was closely associated with the cranial base. There was also a defect in the midline sphenoid bone consistent with a persistent craniopharyngeal canal. A magnetic resonance (MR) image was obtained to further evaluate the intraoral mass (Fig. 3) and any other associated cranial anomalies.

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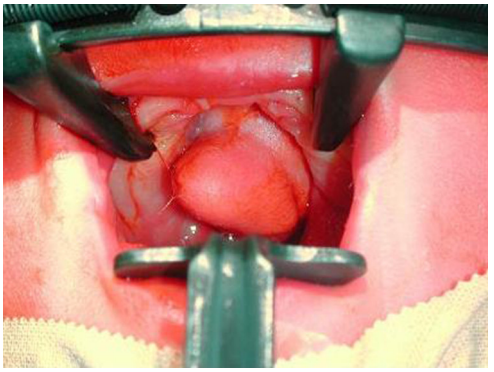

FIGURE 1. Intraoral examination of the patient showing a nasopharyngeal teratoma extending through a cleft in the middle third of the vomer, as well as a bilateral postincisive cleft palate.

Abnormal bifurcation of the clivus was demonstrated on imaging, and it was closely associated with the patent craniopharyngeal canal (Fig. 4). Intracranially, 3 main findings were observed: a duplicated pituitary gland and stalk (Fig. 5), tubomammary fusion, and a midline hypothalamic mass along the floor of the third ventricle. The corpus callosum and dentate nuclei were normally formed. Note that the prenatal ultrasound did not demonstrate the mass or the cleft palate. One could conjecture that the mass filled the palatal area obliterating the cleft on the ultrasound study.

The patient underwent surgical excision of the nasopharyngeal mass on day 4 of life. The tumor was dissected off the dura and was the superior extent of the excision. Coverage of the dura, as well as closure of the defect, was accomplished with mucosal flaps from the remaining septum. Gross pathology noted a $3.2 \times 1.5 \times 1.4$-cm^3 skin covered fingerlike mass with internal white hairs up to 3 cm in length. Microscopic examination revealed adipose tissue, hair follicles, salivary glands, and a tooth bud. The tissue diagnosis was a benign teratoma.

The patient was noted to be healing well from the excision at 1 month. However, because of poor feeding and gastroesophageal reflux, the patient underwent a Nissen fundoplication with placement of a feeding gastrostomy. The palatal defect was closed at the age of 9 months using a V-Y pushback technique (ie, Veau-Wardill-Kilner).

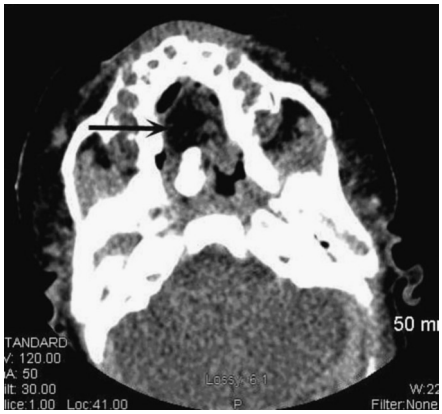

FIGURE 2. Transverse computed tomographic scan demonstrating the nasopharyngeal teratoma (arrow) extending through a large defect in the hard palate and containing both lipomatous and ossific components.

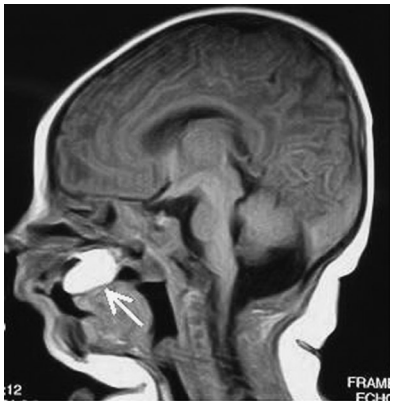

FIGURE 3. Sagittal MR image showing another view of the nasopharyngeal teratoma (arrow).

The patient' thyroid function tests, chromosome profile, electroencephalograph, echocardiogram, and ophthalmologic evaluation were normal. The clinical examination at 1 year revealed a normal growth curve with no evidence of any neuroendocrine deficiencies. The patient was seen at the age of 5 years. The feeding tube has been removed. Her swallowing function and endocrine function remains normal. There was no evidence of recurrence of the teratoma clinically and by MR imaging.

DISCUSSION

The embryological origin of the pituitary gland is often misquoted as arising from 2 separate structures, an upward extension of oral ectoderm, and a downward evagination of diencephalon neuroectoderm.[10] The true origin of the pituitary, as first described by Gilbert[20] in 1934, is from a single structure: the adhesion of the surface and neuroectoderm in a small area on the ventral surface of the developing head. The primordium of this gland is first distinguished at the gestational age of 22 days.[13]

Duplication of the pituitary gland was first described in 1880 by Ahlfeld[21] in a patient with partial duplication of the brain. At that time, the hypophyseal duplication was attributed to a partial twinning of the anterior part of the body.[21] However, now, many different theories exist to explain this phenomenon, which is not yet completely understood.

One of the more recent theories to describe the cause of pituitary duplication was initially postulated by W.M. Morton in 1956. He hypothesized that this malformation resulted from a duplication of

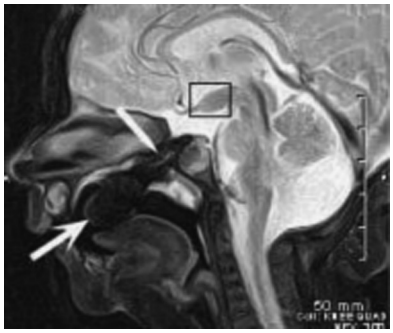

FIGURE 4. Sagittal MR image demonstrating a nasopharyngeal teratoma (large arrow) and a patent craniopharyngeal canal (small arrow). The box demonstrates a hamartoma at the base of the third ventricle or fusion of the tubomammary bodies.

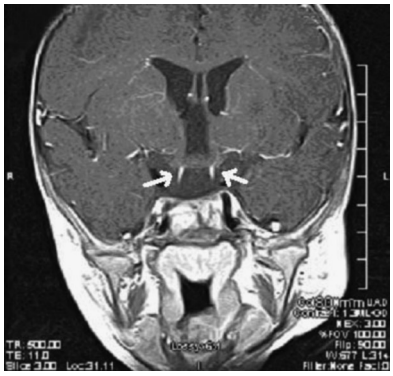

FIGURE 5. Coronal MR image showing duplicated pituitary gland and stalk (arrows).

the prechordal plate and the anterior end of the notochordal process at the 15th to 16th day of pregnancy.[1] This event presumably leads to the formation of 2 areas of persistent contact between neural and surface ectoderm resulting in 2 pituitary glands. The cleft created by the splitting of the prechordal plate and the notochordal process is filled by invaginating mesenchyme. In the developing face, this medial collection of mesenchyme (organized into a mass) hinders the fusion of normally formed lateral structures resulting in a midline cleft. The location of the midline mass governs the location of the clefts.[10] If, as seen in the case of our patient, the mass interferes with the development of the lateral palatine processes, the result is a hard and soft palate cleft. Our case seems to be very similar to other documented cases of pituitary duplication with a nasopharyngeal teratoma.[10]

The most commonly associated anomaly described with a duplication of the pituitary is the presence of a midline hypothalamic mass along the floor of the third ventricle. This finding was observed in our patient, who exhibited a hypothalamic hamartoma. The abovementioned theory first proposed by Morton also supports an explanation of this phenomenon. The notochord plays an important role in influencing ependymal differentiation, and experimental splitting of the notochord has demonstrated thinning of the ependymal layer. Therefore, duplication of the notochord leads to 2 areas of low mitotic activity in the diencephalon and, subsequently, the formation of 2 median eminences. The mass observed in these patients exists between these 2 eminences and consists of abnormal cells, which normally migrate laterally to form the hypothalamic nuclei.[10]

Another one of the more recent theories of pituitary duplication, described by Shah et al,[11] explains the duplication in the presence of the median cleft face syndrome. The formation of a median cleft in early embryology by a midline inclusion dermoid leads to the splitting of the pituitary anlage and the formation of 2 pituitary glands. Therefore, in contrast to the first theory, the mass is the cause of the split rather than an effect of it. The presence of pituitary duplication in the existence of a midline mass, cleft, palate and sphenoid, according to Shah et al, may represent a spectrum of the median cleft face syndrome. This includes cranium bifidum occultum frontale, V-shaped hairline, and clefting of the nose, upper lip, and premaxilla. There was no evidence of any of these findings in our patient.

Although no mechanism or exposure has been consistently described in association with pituitary duplication, reports have cited uterine bleeding early in pregnancy,[1] maternal surgery early in gestation,[4] vaginal herpes,[5] digoxin,[5] meclizine,[7] smoking,[11] cortisone,[11] and naproxen[11] as possible contributors. None of these events or exposures occurred in our patient.

Teratomas, defined as true neoplasms arising from all 3 germ layers, occur in roughly 1 in 4000 live births. Of these, most occur

in the sacrococcygeal area. Head and neck teratomas comprise approximately 1% to 10% of congenital cases. Conditions with a pure oral presentation, as in our case, occur very rarely.[22] Prenatal ultrasound has been shown useful in diagnosis, largely in cases where oral obstruction has occurred resulting in polyhydramnios.[23] However, a large portion of these tumors, 78% in one study,[22] are not detected until birth even with regular prenatal ultrasound examinations.

Because teratomas of the oral cavity tend to protrude out of the mouth as they enlarge and because infants are obligate nasal breathers, infants with pure oral teratomas tend to have better respiratory outcomes than children with other head and neck presentations.[24] At presentation, ultrasound examination of the mass is useful to document solid and cystic components. Computed tomographic scans are important for clinical suspicion of extensive cranial involvement or communication with intracranial structures, whereas MR imaging gives a clear picture of the neuroanatomy. True diagnosis of the mass is often only possible with histopathologic examination.[22]

Treatment of oral teratomas is focused on complete excision of the tumor, with early excision advised especially if the airway is compromised.[25,26] Although malignant degeneration in oral teratomas is not well documented, a 90% degeneration rate has been documented in other areas of the head and neck.[27] Adjuvant chemotherapy has not been shown to be beneficial even in tumors with immature features.[28] Surgical management of the clefts should proceed at the normal time course.

Whereas initial management of pituitary duplication includes ruling out associated anomalies, continued clinical surveillance for neuroendocrine disorders is also important. Reviewing the literature reveals about 30 cases of pituitary duplication with less than half of these cases involving a nasopharyngeal teratoma.[1–19] Whether this case represents a spectrum of notochord duplication or is the manifestation of a teratogen on the morphogenesis of embryologic structures is open for debate. Future research further defining the stage-by-stage processes at the microscopic and cell signaling level involved in the development of the embryologic cranial base may eventually define the true cause-and-effect relationship in this constellation of malformations.

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Corticosteroids or Cyclooxygenase 2–Selective Inhibitor Medication for the Management of Pain and Swelling After Third-Molar Surgery

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Abstract: The dental-impaction pain model is the most commonly used and widely accepted acute pain model for assessing the analgesic effect of drugs in humans. The aim of this randomized crossover

clinical trial was to observe and compare the anti-inflammatory effects of corticosteroid and nonsteroidal cyclooxygenase 2–selective inhibitor medication on pain, swelling, and trismus after third-molar surgery. For this, 50 adult subjects, 25 male and 25 female, with ages ranging between 18 and 29 years (mean, 22.5 years) and no local or systemic problems, presenting bilateral impacted lower third molars in similar position with surgical extraction indicated were selected. The subjects were submitted to 1 surgical procedure for each side with interval of 3 weeks between each procedure, in which they were given 120 mg etoricoxib, nonsteroidal anti-inflammatory (group 1), or 4 mg dexamethasone, corticosteroid anti-inflammatory (group 2), 1 hour before the procedures. Data were registered at preoperative baseline and 24 and 48 hours postoperatively. Postoperative pain was evaluated using a visual analog scale, and the degree of swelling was evaluated through facial reference points' variation. The presence of trismus was analyzed through measurement of the interincisal distance. These assessments were obtained before the operation and at 24 and 48 hours after the surgeries. There was no statistically significant difference between anti-inflammatory treatments. However, at 48 hours, the facial swelling increased in both groups despite trismus reduction. The effects of nonsteroidal and steroidal anti-inflammatory drugs were similar for pain, swelling, and trismus.

Key Words: Corticosteroids, COX-2 selective, third-molar surgery

The surgical extraction of lower third molars is the most frequent intervention in oral surgery.[1,2] This procedure often results in postoperative pain, swelling, and trismus due to the surgical trauma.[1,3–7] The pain reaches a maximum intensity 3 to 5 hours after surgery continuing for 2 to 3 days, then gradually diminishes until the seventh day.[8] The swelling peak intensity occurs at 12 to 48 hours after surgery and resolves only between the fifth and seventh days.[9] The control of inflammation and associated symptoms is necessary to decrease the social impact of this procedure.[1,3,10–14] These postoperative sequels are a consequence of the conversion of phospholipids into arachidonic acid by the phospholipase A2 enzyme (PLA2). This activates the formation of mediators of the inflammatory response such as prostaglandins, leukotrienes, or thromboxane-related substances.[5,15,16]

The anti-inflammatory therapy of corticosteroids is well known, and they are widely used to decrease the swelling related to third-molar surgery.[1,5–7,12,14,15] Corticosteroid mechanism of action includes the inhibition of PLA2, which reduces the release of arachidonic acid in the cells of the inflamed focus and consequently decreases prostaglandin and leukotriene synthesis. Previous studies[1,5,12,14,15] have shown that a preoperative single-dose administration of dexamethasone can effectively reduce the inflammatory response after oral surgical procedures.

Another approach to block the nociceptive response of endogenous mediators of inflammation is the use of nonsteroidal anti-inflammatory drugs (NSAIDs), which reduce the synthesis of prostaglandins from arachidonic acid by inhibiting 2 different isoforms of cyclooxygenase enzymes (COX-1 and COX-2).[15,16] The effect is greatest in tissues that have been subjected to injury and trauma, representing a pathway that can be used to prevent primary hyperalgesia.[4,17] However, the use of COX-nonselective NSAIDs in the postsurgical concerns about prolonged bleeding times due to inhibition of platelet aggregation factor (thromboxane A2) and gastrointestinal intolerance, including ulcers and bleeding.[4,9]

Because of the nonblockage of COX-1 enzymes, the COX-2–selective NSAIDs do not affect platelet function[4] and are associated with fewer gastrointestinal adverse effects due to preservation of the gastric protective functions of prostaglandins, which are responsible for protecting the gastrointestinal mucosa and platelet aggregation.[9] Etoricoxib is an NSAID more than 100-fold selective for COX-2 than COX-1 in whole-blood assays with similar efficacy to traditional NSAIDs.[18] Also, its half-life of approximately 25 hours supports once-daily dosing.[19] In a previous randomized, placebo-controlled, double-blind, single-center study in a third-molar extraction model, 120 mg etoricoxib was shown to be the minimum dose that had maximal efficacy in patients with moderate to severe acute pain associated with dental surgery,[3,20] showing to be a single-dose oral medication better than other commonly used analgesic.[4] However, no comparisons have been made between this COX-2–selective NSAID and corticosteroids.

Therefore, the aim of this randomized clinical trial was to observe and compare the anti-inflammatory effects of the 2 proposed protocols, corticosteroid and COX-2–selective inhibitors, on pain, swelling, and trismus after third-molar surgeries.

CLINICAL REPORT

This study was a prospective, randomized, double-blind, and controlled clinical trial in which subjects acted as their own control (crossover), and this study was approved by the local ethical committee. Fifty subjects from Dental College of Juiz de Fora Federal University, 25 men and 25 women, with ages ranging between 18 and 27 years and no local or systemic problems, requiring surgical removal of the 2 mandibular impacted third molars under local anesthesia were included. The inclusion criteria were asymptomatic mandibular third molars with class I or II position and A or B impaction level according to Pell and Gregory radiographic classification[1,4] (Fig. 1), no periodontal disease, and without signs of

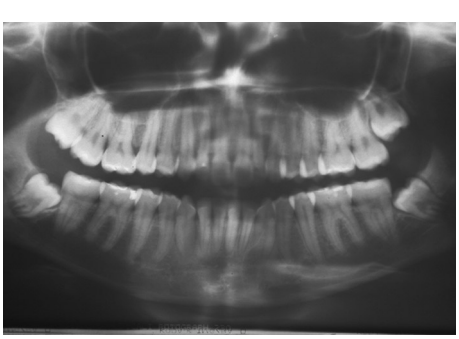

FIGURE 1. Radiographic classification according to Pell and Gregory, class I or II position and A or B impaction level.

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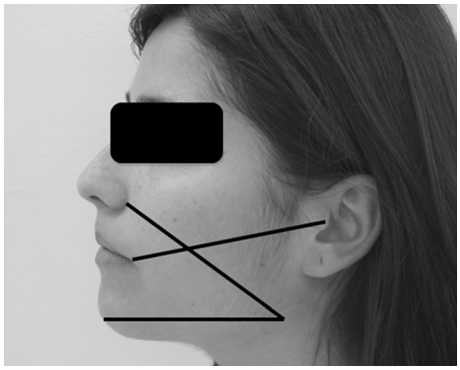

FIGURE 2. Tape-measuring method for evaluation of facial edema was performed preoperatively and at 24 and 48 hours postoperatively.

acute pericoronitis at the time of surgery. Exclusion criteria included systemic diseases, recent anti-inflammatory treatment, chronic use of medications that would obscure assessment of the inflammatory response and pregnant or lactating women. The nature of the surgical and experimental procedures was informed to all subjects, and informed consent was obtained before surgery.

A split-mouth design was used, in which each side constituted the experimental unit and each subject acted as his/her own control, increasing statistical efficiency. The sequence of treatments and surgical side were randomly assigned by numbered containers. One hour before the first surgical third-molar removal, the subjects received 120 mg etoricoxib (COX-2–selective NSAID) (group 1) or 4 mg dexamethasone (corticosteroid) (group 2). The second surgical visit was scheduled 3 weeks later when the other contralateral third molar was removed with administration of the other medication 1 hour before the surgery.

Surgical Procedure

To minimize differences due to operator variability, all of the surgical procedures were performed by the same oral surgeon, who does not know the preoperative medication regimen. Surgical removal of the impacted third molars was performed with the patient under local anesthesia using 2% lidocaine with 1:200,000 epinephrine. The inferior alveolar, lingual, and long buccal nerves were anesthetized. The access to the third molar was done by a mucoperiosteal incision and a full-thickness flap reflection. When necessary, osteotomy and tooth sectioning were done with burs under constant irrigation with sterile isotonic saline. Once all the tooth components were extracted, the socket was carefully inspected and the flap sutured with interrupted sutures using 4-0 silk. The duration of surgery and osteotomy was recorded.

TABLE 2. Comparison of the Total Number of Analgesic Doses and VAS Scores Between the 2 Groups

	Total No. Analgesic Doses	VAS Scores
Group 1	2 (1.8)	6 (1.8)
Group 2	2 (1.1)	5 (2.3)
P	0.972	0.793

Values are presented as mean (SD).

All patients received a prophylactic preoperative dose of oral antibiotic (1 g amoxicillin 1 hour before surgery), and postoperative regimen was not prescribed. If necessary, the subjects were instructed to take 750 mg paracetamol tablet at each 6 hours.

Data Collection

The pain was assessed using a visual analog scale (VAS), 10 mm in length, where 0 was marked as "no pain" and 10 as "most severe pain imaginable," and by the report of the total number of analgesic paracetamol tablets required until 48 hours after surgery.

Facial swelling was evaluated by a modified tape-measuring method described by Gabka and Matsumara.[6] Three measurements were made between 5 reference points: tragus, soft tissue pogonion, the corner of the mouth, angle of the mandible, and lateral corner of the nose (Fig. 2), and the sum of this 3 measurements was considered as the swelling data for that side, and the difference between the preoperative (baseline), 24 hours, and 48 hours postoperative measures indicated the facial swelling for that day.

Trismus was evaluated by measuring the distance between the mesial-incisal corners of the upper and lower right central incisors at maximum opening of the jaws preoperatively, on the 24 and 48 hours postoperatively. The difference between each measure indicated the trismus for that day. Data are reported as means and SD and analyzed by a blind researcher at significant level of 0.05. The pain scores of VAS were evaluated by nonparametric test (Wilcoxon test). The data on edema, trismus, and number of analgesic tablets violate normality assumptions (Shapiro-Wilk test) and were transformed [$\log(\chi)$]. The difference between anti-inflammatory treatments was analyzed by the paired *t*-test.

RESULTS

The time of surgery in group 1 was 29.59 (SD, 3.76) minutes and in group 2 was 27.74 (SD, 5.54) minutes without statistical difference among them ($P = 0.47$). At follow-up, no patients developed wound infection or serious postoperative complications.

TABLE 1. Comparison of the Facial Swelling and Trismus Between the 2 Groups

	Swelling		Trismus	
	24 h Postoperative	48 h Postoperative	24 h Postoperative	48 h Postoperative
Group 1	5.67 (1.2)	8.66 (2.4)	17.8 (8.2)	17 (6.01)
Group 2	5.38 (1.8)	8.21 (1.9)	16.5 (6.3)	15 (5.2)
P	0.856	0.759	0.359	0.344

Values are presented as mean (SD).

At 48 hours postoperative, the edema increased in both groups; however, the difference between the 2 groups was not statistically significant (Table 1).

Regarding trismus, there was no significant difference between the groups in opening-mouth-reduction scores (24 hours postoperative: 17.8 mm for group 1 and 16.5 mm for group 2; 48 postoperative hours: 17 mm for group 1 and 15 mm for group 2) (Table 1).

The evaluation of pain was similar in both groups. The number of analgesic tablets taken and the VAS pain scores in group 1 have no significant difference from those in group 2 (Table 2).

DISCUSSION

Surgical removal of impacted third molars is one of the most frequently performed procedures in oral and maxillofacial surgery and can lead to immediate and postoperative discomfort.[11,13] The extent of swelling, trismus, and the severity of pain are the chief indicators of a patient's morbidity during the postoperative period after third-molar removal.[1,7,9,11,13,14] The dental-impaction pain model is the most commonly used and widely accepted acute pain model for assessing the analgesic effect of drugs in humans.[3]

Therefore, many clinicians have attempted to reduce postoperative sequelae, using anti-inflammatory drugs.[1,4–7,9,12–16] The pattern of postsurgical pain experienced by the patients in this study was similar to that in previous reports; the greatest levels of pain were experienced on the day of surgery,[1,12] in accord to the results in the study of Chopra et al,[8] who state that the pain peak occurs within 3 to 5 hours after surgery. In the current study, there was no statistical difference for pain, swelling, and trismus between the 2 anti-inflammatory protocols evaluated, but both were effective. Ustün et al[21] and Laureano Filho et al[14] investigated 2 doses of corticoid after third-molar removal surgery and concluded that both have good analgesic and anti-inflammatory action.

Grossi et al[13] evaluated the effect of 4 mg dexamethasone on discomfort after mandibular third-molar surgery, and the authors concluded that corticoid is effective in the prevention of postoperative edema. Varner et al[20] evaluated the analgesic efficacy of doses of a COX-2 inhibitor compared with placebo in the extraction of third molars, with analgesic effect superior to placebo in this acute pain setting. However, Troullos et al,[22] in a comparative study with NSAIDs and steroids, found different results from this study; the NSAIDs produced greater initial analgesia than did steroids, whereas steroids result in greater suppression of swelling and less loss of function.

This study evaluated 2 different proposed protocols for anti-inflammatory therapy, the use of corticosteroids and COX-2–selective inhibitor. It was thought that corticosteroids were much more powerful anti-inflammatory medications than NSAIDs[15]; however, in this study, a significant difference between them was not found. The absence of significant difference may be explained by the dose of corticosteroid used in this study; according to Kim et al,[15] the dose must be equal or exceed the physiologic amount released by the body, suggesting a dose equivalent to 300 mg cortisol for maximum anti-inflammatory effect, which is equivalent to 9 mg dexamethasone. Therefore, the 4-mg dose of dexamethasone used in this study may not be sufficient to achieve the maximum anti-inflammatory effect. The use of corticosteroids in oral surgical procedures may present as an area of ambiguity for surgeons. Whereas some authors recognized the administration of corticosteroids, others consider its use only as a supplement in patients undergoing extensive oral surgery procedures.[15] Many studies have determined the effectiveness of corticosteroids after oral surgical procedures[1,7,15] by the inhibition of the enzyme PLA2, leading to a reduction in prostaglandins and leukotrienes.[5,15] Corticosteroids such as dexamethasone and methylprednisolone have been used extensively in dentoalveolar surgery for oral surgery,[1,7,12–15,21] despite having no standard on dosing regimen.[15]

The administration of NSAIDs decreases inflammation and fever while providing analgesia by inhibiting the COX enzymes COX-1 and COX-2, which are needed for the production of inflammatory mediators such as prostaglandins, prostacyclins, and thromboxanes.[15,16] When administered preoperatively, NSAIDs have been shown to be particularly effective in combating postoperative pain.[23] However, the association of corticosteroids and NSAIDs offers a better therapeutic outcome.[24,25]

CONCLUSIONS

Oral surgical procedures involving the removal of at least 1 partial bony or full bony impacted third molar provide a considerable amount of soft and hard tissue trauma. Therefore, preoperative use of anti-inflammatory medication appears to be a valid method to increase patient comfort postoperatively. In this study, the effects of nonsteroidal and steroidal anti-inflammatory drugs were similar for pain, swelling, and trismus.

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Malignant Peripheral Nerve Sheath Tumor of Mandible

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Abstract: Malignant peripheral nerve sheath tumor is a common tumor that rarely affects the head and neck region. The patient presented in this report is a teenage girl presented with a lesion in the right body of the mandible with severe disfigurement of the lower face. The lesion was first histopathologically diagnosed as embryonal rhabdomyosarcoma. After excision, however, the histopathology report proved the diagnosis of malignant peripheral nerve sheath tumor.

Key Words: MPNST, neurosarcoma, schwannoma, mandible, malignant peripheral nerve sheath tumor, malignancy, tumor

Malignant peripheral nerve sheath tumor (MPNST) is an aggressive and rapidly growing malignant tumor that infiltrates the surrounding tissues and metastasizes early.[1,2] Incidence of MPNST is about 0.1:100.000 of population per year. They arise either as a de novo solitary mass or in association of with neurofibromatosis type 1 (NF1) in 38%[3,4] to 60% of cases.[5] Malignant peripheral nerve sheath tumors account for 5% of all soft-tissue tumors[6–8] and less than 5% of all malignant neural tumors.[9] Of these, 15% are in the head and neck region; however, they are extremely rare in the oral cavity.[10,11]

They occur mainly extraosseously in association of major nerve trunks as sciatic nerve, sacral plexus, and brachial plexus.[12] They likely originate from peripheral nerve sheath cells, including Schwann cells and perineural cells. Tumors that arise from endothelial cells and the epineurium are not included under this category.[13,14]

These tumors infrequently show histologic evidence of focal divergent differentiation to rhabdomyosarcoma, osteosarcoma, chondrosarcoma, angiosarcoma, epithelial elements, glandular element, or a combination of some of the abovementioned elements. This display of variable phenotypic expression is thought to be due to the tumor's origin from Schwann cells, which are derived from the pluripotent neural crest cells.[15–17]

Reported cases of MPNST started mostly in the soft tissues,[18] but can arise in bone,[19] with the age at onset somewhere between the third and fifth decades. However, cases have been reported from 3 weeks to 80 years old.[6–10,13,20–22] There is no sex predilection in sporadic cases,[3,4,23] whereas there is male predomination in cases associated with NF1.[24,25] Previous cases have been reported in the maxilla,[20] maxillary sinus,[1] tongue,[4] skull base,[26] parapharyngeal space,[27] and parotid gland.[28] In this report, a case of MNST of the mandible is presented.

CLINICAL REPORT

A 16-year-old female patient presented to the Oral and Maxillofacial Department of Garyonis University Dental School; Benghazi, Libya, with moderate-size intraoral mass. The mass extruded from the extraction socket of the lower right second premolar that had been

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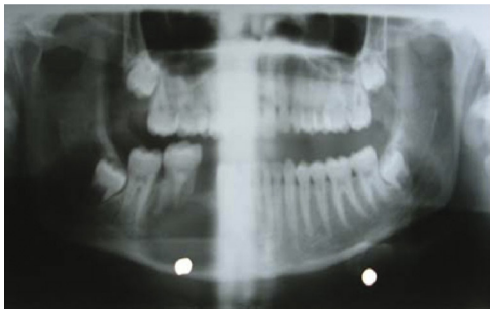

FIGURE 1. Orthopantomogram showing radiolucent area extending from mandibular right canine to mandibular second molar and reaching the inferior border of the mandible.

extracted 10 days earlier. The mass measured about 10 × 10 cm and was soft and friable, easily bled on touch, and had an offensive odor. Regional lymph nodes were impalpable, and the medical history was unremarkable.

Orthopantomogram revealed a radiolucent area extending from the mandibular right canine to the right second molar, reaching down to the lower border of the mandible (Fig. 1). Computed tomography (CT) scan of the head and neck was carried out with and without contrast, revealing a large lesion in the right side of the mandible that eroded both cortices and extended both bucally to occupy the buccal vestibule and lingually to occupy the right half of the floor of the mouth and the upper part of the neck (Fig. 2). The first incisional biopsy taken for the lesion under local anesthesia was reported as embryonal rhabdomyosarcoma.

The patient was referred to oncologist who started a course of preoperative chemotherapy aiming to shrink the size of the tumor and to control its rapid growth. The patient received 2 doses of chemotherapy within 3 months without any improvement. Quite the opposite, the tumor continued to increase in size, bulging into the oral cavity (Fig. 3). A decision of tumor radical resection was made, and the patient was referred and admitted to Nasser Institute Hospital and Research Center, Cairo, Egypt. A second incisional biopsy was taken, which confirmed the original diagnosis of embryonal rhabdomyosarcoma. Bone scan, chest CT scan, abdominal ultrasound, and upper aerodigestive endoscopy were carried out, showing no abnormality except for mild leukocytosis.

Surgical resection of the mandible was carried out, extending from the left canine and crossing the midline to the right mandibular angle together with part of floor of the mouth and the skin covering the chin. Resection margins were diagnosed free by frozen section. Immediate reconstruction was performed using titanium reconstruction plate and ascending cervical flap (Fig. 4).

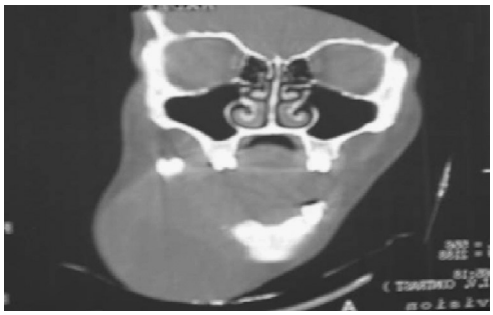

FIGURE 2. Coronal CT showing a large lesion in the right side of the mandible that eroded both cortices.

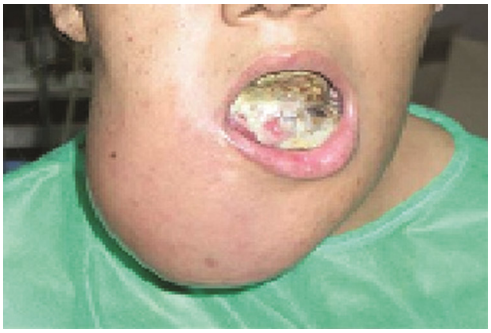

FIGURE 3. The lesion bulging into the oral cavity.

Histopathologic Examination of the Resected Tumor

Gross examination of the excised mass revealed a firm grayish white mass with focal areas of necrosis. Microscopic examination revealed infiltrating noncapsulated neoplasm composed of spindles of hyperchromatic cells with scattered mitotic figures, focal fascicular pattern with focal necrosis, and marked vascularity (Figs. 5 and 6). Tumor cells infiltrated skeletal muscles, soft tissues, and bone. Sections were treated in microwave for 20 minutes using citrate buffer for antigen retrieval. Then immunohistochemistry was carried out using antibodies for desmin, myogenin, and S100, with DAB as chromogen. Sections were examined using envision system (Dako, Carpinteria, CA). Tumor cells were negative for both desmin and myogenin and strongly positive for S100 protein (Fig. 7) and vementin (Fig. 8), which was consistent with MPNST.

Three months postoperatively, the patient presented to the outpatient department complaining of painless swelling under her tongue. Bimanual palpation revealed a large, firm, nontender, fixed lump involving the floor of the mouth. Surgical exploration was carried out through the old scar to excise the mass. Histopathology examination of the excised mass revealed recurrence. Radiotherapy and ozone injection were[29] started. Seven sessions of subcutaneous injections of 30 to 50 mL of oxygen-ozone gas mixture in the cheek and submandibular area with a flow rate of 0.5 mL/second and ozone concentration of 4 to 8 μg/mL.

Four months later, second recurrence occurred (Fig. 9). It was determined that the tumor was now inoperable. Permanent tracheotomy was performed as palliative therapy to aid the respiration. The patient died 2 months later because of respiratory problems.

DISCUSSION

Unlike schwannoma, MPNSTs are rare in head and neck region,[23] totaling about 246 cases reported worldwide.[1,30] Intraosseous

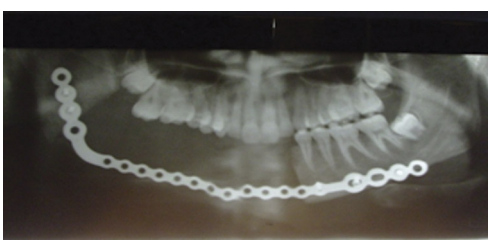

FIGURE 4. Orthopantomogram showing reconstruction plate in place after resection.

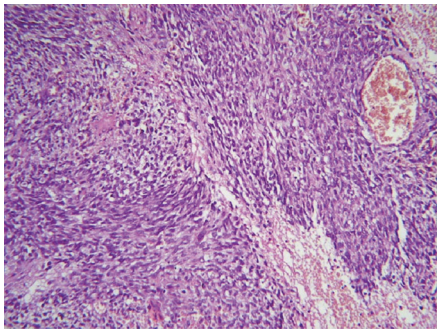

FIGURE 5. Markedly cellular tumor tissue composed of spindle hyperchromatic cells in fascicular pattern with focal necrosis and marked vascularity (hematoxylin-eosin stain, original magnification ×200).

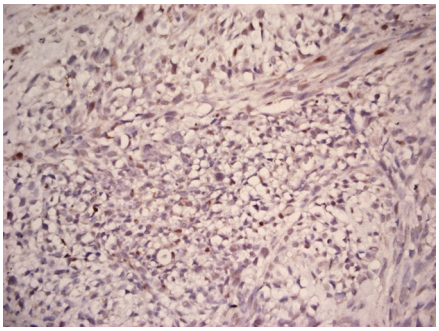

FIGURE 7. Cellular tumor with moderate number of strongly positive tumor cells for S100, mostly cytoplasmic staining (immunohistochemistry, original magnification ×400, S100).

MPNSTs are rare compared with soft-tissue MPNSTs. Nevertheless, 13 of 18 intraosseous cases of MPNST reported in the literature took place in the mandible and maxilla.[19] Low incidence of MPNST in the bone may be attributed to the lack of tendency of intrabony nerve sheath to malignant behavior.[31]

Diagnosis of MPNST

Imaging with CT, magnetic resonance imaging, and angiography may be used in the diagnosis of MPNST, with the CT being better in visualizing bone destruction, whereas magnetic resonance imaging is superior in showing the soft-tissue extension and demarcation of tumor tissue from normal tissue.[1] Clinically, the gross appearance of MPNSTs is that typical of sarcoma as they are usually solitary, pseudocapsulated, hard, fleshy tumors, usually larger than 5 cm in diameter, and often show necrosis and infiltrate the surrounding tissues.[24,32]

Diagnosis of MPNST is mainly based on microscopic detection of nerve fibers within the tumor mass or structural features that display multiple-branched cytoplasmic processes covered by basal lamina.[33] However, because of divergent differentiation of this highly pleomorphic tumor, there may be considerable difficulty in its diagnosis based on histopathologic examination. Most cases show the typical alternating fascicles of spindle-shaped cells and areas of myxoid or hyalinized tissue.[1] However, they can also have a variety of features that can render diagnosis difficult, including sarcoma-like mesenchymal elements,[7,34] glandular elements, and squamous epithelial cells.[16,35,36]

Diagnosis should be confirmed by immunohistochemical analysis, showing tumor cells negative for desmin and myogenin and positive for neuron-specific enolase, which demonstrates neural or neuroendocrine differentiation, in addition to S100 protein to support the diagnosis of neural origin.[6,7,20,21] Some tumors may also be positive for melanotic marker HMB-45.[1] Additional confirmation can be obtained with electron microscopy that shows cellular ultrastructure and early cell differentaiation.[37,38]

Detection of chromosomal abnormalities has been suggested for the diagnosis of soft-tissue tumors.[7,22] In cases of MPNST, however, some alterations may be detected, which are not consistent in all cases.[39,40] These include structural abnormalities of the NF1 and tumor suppressor gene (TP53) loci[41] in chromosome 17 or point mutation[42] of exon 4 or exons 5 to 8 of P53 gene.[43] In some cases, there was inactivation of both alleles of the NF1 gene.[44] In addition, evidence suggests that p27 nuclear expression is absent, and more recently, p16 deletion is present in most cases.[45]

Classification and Grading

According to the World Health Organization, MPNSTs are classified into epithelioid MPNST, MPNST with divergent mesenchymal and/or epithelial differentiation, melanotic MPNST, and melanotic psammomatous MPNST.[46] There is no universally accepted histologic grading system for MPNST. However, it has been suggested that tumors that show crowded, hyperchromatic, and mitotically active cells, with or without necrosis, and those with sarcomatous differentiation could be considered high grade, whereas those with perineural cell differentiation could be considered low grade.[47]

Others classified high and low grades according to the World Health Organization grading system, based on degree of cellularity, anaplasia, vascular proliferation, nuclear atypia and enlargement,

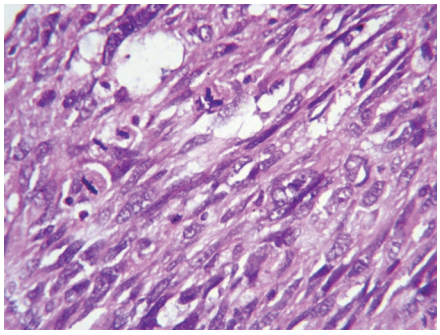

FIGURE 6. Spindle cell sarcomatous lesion with marked pleomorphism and multiple mitotic figures in 1 high-power field (hematoxylin-eosin stain, original magnification ×400).

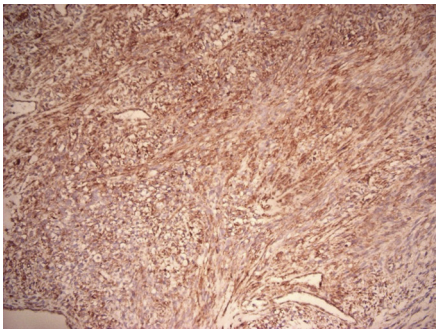

FIGURE 8. Tumor cells are markedly positive for vimentin (immunohistochemistry, original magnification ×100, vimentin).

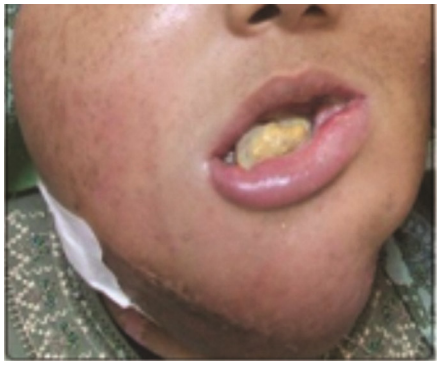

FIGURE 9. Second recurrence of tumor mass protruding in the oral cavity.

hyperchromatism, mitotic rates, and existence of necrosis.[48] Accordingly, high-grade tumors would be those that are formed of fasciculated cells with hyperchromatic nuclei and mitotic figures of more than 4 in 10 high-power field, whereas low grade would have lower cellularity, with less hyperchromatism and less mitotic figures.[25]

Treatment

Because of its aggressive and rapid local invasion and high recurrence rate, which ranges from 38% to 68%,[3,24] MPNSTs are treated with very wide resection, in addition to postoperative radiotherapy[2,3,23] and/or chemotherapy.[49,50] Brachytherapy can be used as a complementary,[1,6,15,25] or standalone treatment.[1,11] Several investigators recommend the use of radiotherapy for preventing local recurrence.[20,25,51] Others rejected the value of chemotherapy,[48] and radiotherapy, whether preoperative or postoperative, in improving the prognosis.[6,24,52] Lymph node dissection is not required.[9,13,51,53] Wide surgical margin is the only reliable procedure for controlling local recurrence.[48]

Prognosis

In this review about the 273 reported head and neck cases in the period from 1970 to 2008,[1,30] 142 cases (52.01%) were treated with surgery and radiotherapy, with 42% with 5-year survival rate. Another 66 patients (24.1%) were treated with surgery alone and had 35% survival rate, and 61 cases (22.3%) treated with surgery and received both radiotherapy and chemotherapy with 38.5% survival rate. Only 2 cases (0.7%) treated with surgery and chemotherapy had a survival rate of 22%. Studies that correlated the clinicopathologic features of PMNST to treatment outcome reported that older patients with large tumor size require radiotherapy and existence of NF1 increases recurrence rate.[3,24] Others reported a relationship between increased mitotic rate and local recurrence[24,52] or inadequate surgical excision and recurrence.[45,51]

Many other variables have been studied, including age, sex, duration of symptoms, site and size of the tumor, lung metastasis at presentation, S100 expression, and adequacy of surgical margins.[25,48] Large tumor size,[5,25] histologic grading,[25] and lung metastasis at presentation are all associated with poor prognosis.[48]

Tumor site is a significant factor for survival, which may be due to the early manifestation of the disease in exposed areas such as extremities versus the tumors in the body cavities, and also the ease and accessibility of wide surgical resection in the extremities.[25] Tumor necrosis of more than 25% was associated with poorer prognosis.[4] Existence of NF1 is an adverse prognostic factor.[2,5,25] Radiation-induced[5,25] MPNSTs also have a poor prognosis.[3,25]

Unfortunately, our patient died 12 months after the first presentation regardless of radical treatment, radiotherapy, and ozone therapy. The tumor recurred twice, both within a short time of excision, emphasizing what other authors reported regarding the aggressive nature of this type of tumors.[54,55]

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Patients' Satisfaction After Surgical Facial Reconstruction or After Rehabilitation With Maxillofacial Prosthesis

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Abstract: The matching of the aesthetic, functional, and psychosocial results of a facial deformity may produce devastating effects in its carriers, especially if the lesion is extensive or the treatment is aggressive. Because of this, the objective of the present article was

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to evaluate patient's satisfaction rating after surgical facial reconstruction or rehabilitation with oral and maxillofacial prosthesis, by means of reviewing the literature.

Key Words: Maxillofacial prosthesis, patient satisfaction, surgical facial reconstruction

The face permits man to express his feelings, needs, and aims and, essentially, to communicate with other people. The face is unique regarding its structure, function, and specificity of its tissues and functional subunits such as nose, lips, and eyelids, none of which can be borrowed from other body parts.

The oncologic surgeries, facial trauma, and congenital or pathologic deformities may be causes of facial deformation. The patient who has such deformations, for the most part, does not succeed to absorb the problem on account of being totally exposed. This raises difficulties to his/her integration in the social ambient, affects the self-esteem, and weakens his/her quality of life.[1]

Plastic surgery is a treatment of choice because, undoubtedly, the autoplastic reparation is much more desirable if compared with the alloplastic or artificial treatments.[2] However, in some situations, for instance, the surgical reconstruction is contraindicated: absence of donor area, too much extensive lesion, patient who does not intend to be submitted to surgical procedures, poor financial condition, failing health, and oncologic patients (due to risk of relapse). In the mentioned cases, the oral and maxillofacial prostheses, which are restorative prostheses, are an excellent option of treatment. These prostheses artificially reconstruct the loss of substances of the various regions of the face, restore the aesthetics and function, protect the blood-stained and exposed tissues, and, in addition, act as a psychologic therapy.[3]

The aim of this article was to evaluate patients' satisfaction after surgical facial reconstruction or after rehabilitation with maxillofacial prosthesis by means of a literature review.

MATERIALS AND METHODS

To compose the present review, the MEDLINE database and SciELO indexers were consulted. Studies published between the years 1967 and 2009 were considered. The terms selected for research were "maxillofacial prosthesis," "patient satisfaction," "health-related quality of life," "facial prosthesis," "head and neck cancer," "maxillofacial surgery," and "positive life evolution," "maxillofacial rehabilitation." These terms were individually researched and arranged among themselves. Each article was individually selected by its abstract and analyzed by the following inclusion criteria: (1) The articles should be in English or in Latin language and (2) should be prospective studies; longitudinal, retrospective, and literature review were included.

DISCUSSION

The ideal therapy for reconstructing maxillary defects remains controversial. Several therapeutic approaches have been published, including obturator prosthesis, nonvascularized grafts, and local, regional, and free flaps. The reconstruction of maxillary defects by surgical or prosthetic means depends on the characteristics of the patient, such as age, medical history, and the dimension of the defect.[4]

Facial surgical reconstruction is still a challenge for the surgeon because of trauma, malformation, or oncologic causes. Nowadays, it is recommended for patients with smaller facial defects, for whom the available techniques are effective. The authors suggest that not only is surgical treatment valid regarding the objective of surveillance but also is psychologically and functionally relevant.[5]

Patients endure complex problems after the surgery, among which the sociocultural factors outstand considerably. Those who have higher risks of suffering are people with low quality of life related to their health, those with low level of social support, and particularly women. Not only the patients but also their intimate relationships are affected, and these problems are seldom mentioned in the medical registers. Psychosocial problems have been related by patients of all ages and even in cases of those with only small lacerations.[6]

Almost 5% of the cases of cancer affect the mouth structures, tongue, oropharynx, nasopharynx, and larynx. After the excision of these lesions, it is possible that some problems appear during mastication, deglutition, and speech. In addition, after surgical intervention, the changes in the appearance, in psychosocial function, and professional performance may affect the quality of life of those patients.[4]

Oral tumors and their treatments cause obvious changes in the patient's facial area. People with visible disfiguration reported higher levels of distress when compared with the regular values. The potential hazard caused by facial disfiguration has been laid to the importance of the facial region to personal identity, self-image, communication capacity, and success in interpersonal relationships. Specifically, 41% of cancer patients are worried about their appearance. Nevertheless, the patients submitted to resection of the oral tumor and reported a variety of emotional reactions related to their personal disfiguration.[7]

The focus of facial reconstruction, besides aesthetics, is reestablishment of the function. The facial reconstruction techniques were developed, assisted by the detailed anatomic knowledge, which permits to bring tissue from distant sites. In the last decades, the concept of aesthetic facial reconstruction was popularized. This concept honors the units of facial aesthetics as the boundary composed of transition areas from light to shade on the face and the facial surface, which varies from concave to convex. These boundaries are ideal sites to place the scars. Central units of facial aesthetics such as nose or mouth may be divided in subunits to improve facial reconstruction. Intending to have a discreet reconstructed surface, the facial central subunits are totally replaced, if possible, whenever the majority of the units are lost.[8]

Facial aesthetic reconstruction is an artistic challenge. The reproducible and good quality results can be reached only through a detailed preoperative plan with various options for solution. The appropriate planning is fundamental to any acceptable result. In many cases, consecutive phases need to be executed as part of an initial plan viewing to attain a perfect final result.[9]

Technological progress in computation and image processing, especially in the last 10 years, has permitted the adoption of three-dimensional image protocols in health arena. Moreover, the accessibility and facility for using these modalities allow their extensive adoption and employment in diagnosis and treatment planning. This is particularly important, considering complex deformities, which comprehend both function and aesthetics.[10]

Considering the prosthetics rehabilitation of facial mutilation, the oral and maxillofacial prosthesis is an odontologic specialty that basically aims to recuperate aesthetics, function, and tissue protection and to assist in psychologic therapy. These prosthesis can be internal or external facial restorers, which include ocular, oculopalpebral, nasal, auricular, and extensive facial.

The prosthetic restoration presents various advantages over the surgical reconstruction: it is a new alternative treatment for the patients who carry surgical contraindications. The process is

relatively inexpensive and affords a periodical evaluation and cleanliness of the surgical site. The manufacture process is proportionally short, and the maxillofacial physician has complete control over the prosthesis color, shape, and position. However, the disadvantages include possibility of local irritation of the tissue, necessity of periodically remanufacturing, and the dependency on adhesives and other means of retention. Moreover, the patient may see the prosthesis as a mask and not a part of his/her body. There are reports mentioning that 12% of the patients who received auriculars, nasal, oculars, and facial silicone prosthesis did not use them. There are still uncountable reports about dissatisfaction related to prosthesis aesthetics, color stability, function, and longevity.[11]

The loss of a part of the face causes diverse behavior modification in the person such as low self-esteem, shyness, anxiety, fear, and lack of social and familiar interaction. The prosthesis contributes to the patient to restore his/her concept of a corporal image more satisfactorily.[12]

For the person with mutilation, to accept an artificial substitution of a lost and/or mutilated organ, a psychologic attendance is necessary. Oral and maxillofacial rehabilitation fulfills its aesthetic, functional, and psychosocial objectives within a multidisciplinary context.[13]

Many are the applied means of retention, such as anatomic, which uses the internal shapes of the surgical defects; mechanical, such as glass frames, bases of acrylic resins for silicone prosthesis, where the magnetos and clips are attached; and adhesives and osseointegrated implants.[14]

Rehabilitation of individuals who have craniofacial deformities, resulting from traumas, congenital malformation, or tumors, is an inciting and complex purpose. Most resources and treatment options brought progress to patients and afforded them to outlive their expectations, turning rehabilitation into an urgent and necessary subject. With the advent of craniofacial osseointegrated implants and the improvement of prosthetic materials, the disadvantages are being overcome. This occurs on account of the fact that it is possible to succeed with more retention and stability when implant-supported prosthesis retained by bars, clips, or magnetos are used. The prosthesis becomes lighter with more refined borders without the deleterious effects of the solvent used to remove the adhesive present in the chemical retention. The result of this advance is the evident functional, psychologic, and social improvement, based on the reestablishment of the trustworthiness and aesthetic acceptance, which generates well-being and quality of life.[15]

Research by Chan et al[16] in 2005 evaluated patients' perception over their satisfaction with treatments with adhesive facial and implant-retained prosthesis. An inquiry with 28 questions was conducted evaluating the perception regarding appearance, comfort, fitting and irritation, trustworthiness of retention, usage frequency, application or removal facility, and the treatment financial value. The implant-retained group reported more positive answers in the whole questionnaire, compared with the adhesive group. The authors concluded that the implant-retained facial prosthesis was rated much better, considering the facility of usage and conservation. They were more used during daily activities. This research corroborates the results met with Toljanic et al,[17] where the authors agree that the implant-retained prosthesis offers more benefits to the prosthetic rehabilitation than the adhesive systems (Fig. 1).

Another research, through specific inquiry and clinical investigation, analyzed the results of treatment with implant-retained prosthetic rehabilitation. The involved patients who were submitted to throat tumor removal answered questions regarding the impact in their quality of life. The authors concluded that the implant-retained prosthesis significantly improved the patients' quality of life. It is relevant to mention that the satisfaction rate was considered greater than in the nonirradiated compared with the irradiated cases,[18] although a research[19] performed in 2008 reported the placement of oral and maxillofacial implants, which were placed in irradiated and in nonirradiated patients. A success rate of 98.2% was obtained for the irradiated and 90% for the nonirradiated patients.

The knowledge retained by the professionals in the health arena and, especially, by dental surgeons over the importance of the facial-prosthetic rehabilitation is relevant because of facial mutilations that cause morphofunctional and psychosocial alterations, leading the patients to social and familiar isolation. Nevertheless, these individuals acquire a status of stigmatized and a carrier of a sign that distinguishes them from the others. Through facial prosthetic reconstitutions, they will be able to be reintegrated to the society and strengthen their quality of life.[18,19]

The oral and maxillofacial prosthesis perhaps is the greatest challenge of dental surgeons, which in the extent of dentistry specialties devotes itself to restore parts and regions of the human face, considering not only the anatomic and functional aspects, but also the fact of having to reproduce them naturally.[20]

The problems experienced by maxillofacial prostheses wearers are reduced, because a specialist team applies all the practice and experience and maintains a regular follow-up. The patients' prosthetic rehabilitation allows for satisfactory aesthetic recuperation and well-being, whereas psychologic therapy allows for patients' reinsertion in social and familiar environment to make them feel happier and safer.[4]

CONCLUSIONS

Facial rehabilitation, to some extent, promotes a psychologic restructuring in the patient who has a mutilation. The reaction before the mutilation is different for each patient, and it may cause significant psychologic traumas; thus, it is indispensable to have a psychologic and individualized attendance, until a small inconvenience that may be overcome only through facial rehabilitation.

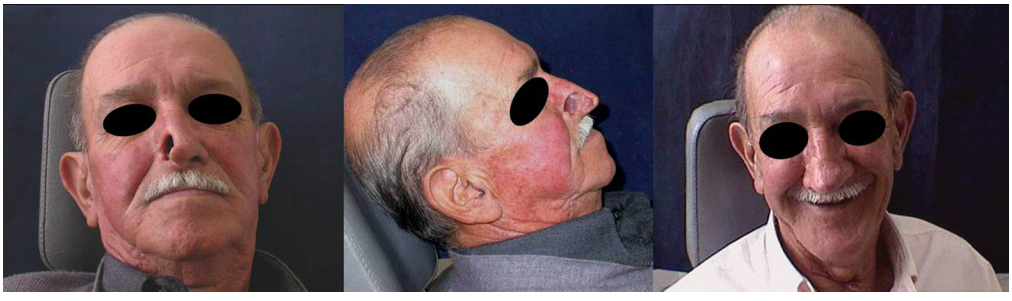

FIGURE 1. Patients' satisfaction after rehabilitation with maxillofacial prosthesis.

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