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Case Report

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Single-Step Primary Reconstruction After Complex Fronto-Orbital Brown Tumor Resection Using Computed-Designed Peek Implant

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ABSTRACT

The authors describe a patient with secondary hyperparathyroidism (HPT) who had development of a brown tumors (BTs) of the superior orbit and frontal calvarium with subsequent visual impairment. One-step surgery involving wide en-bloc resection of the entire tumor and immediate fronto-orbital reconstruction with a custom-made polyetheretherketone (PEEK) implant was planned. Using a PEEK implant a one-step reconstruction of the fronto-orbital region was achieved obtaining symmetry and good functional results, reducing operative time and avoiding donor site morbidity.

KEYWORDS: Oral squamous cell carcinoma; Management; Human papilloma virus; Sentinel node biopsy; Tissue shrinkage; Radiotherapy; Neck dissection; Salvage surgery; Prognosis, QoL; Iberic graft; Facial transplantation.

INTRODUCTION

Brown tumors are uncommon bony lesions caused by rapid osteoclastic activity and peritrabecular fibrosis. They are known to occur only in the setting of HPT, as a direct result of the effect of parathyroid hormone on bone tissue. For years, these lesions have been recognized in primary HPT. However, brown tumors have also been reported in patients with severe HPT secondary to chronic renal failure, especially those on long-term hemodialysis.[1]

The ribs, clavicles, pelvic girdle, spine, tibia, humerus and mandible are their preferred location. While the mandible is the most frequently involved bone in the head and neck region,[2-8] fronto-orbital involvement is extremely rare, with only a few cases in the literature.[9-17] Moreover, reconstruction of this region is complex and remains a challenge for maxilofacial surgeon.

We describe a patient with secondary HPT who had development of a brown tumor of the superior orbit and frontal calvarium with subsequent visual impairment. One-step surgery involving wide en-bloc resection of the entire tumor and immediate fronto-orbital reconstruction with a custom-made PEEK implant was planned.

CASE REPORT

A 27-year-old white woman with medical hystory significant for chronic renal failure on hemodialysis presented with painful progressive proptosis, downward displacement and limited upgaze of her left globe, and a palpable fronto-temporal mass beneath the left frontal scalp. The swelling was firm and painless to the touch (Figure 1).

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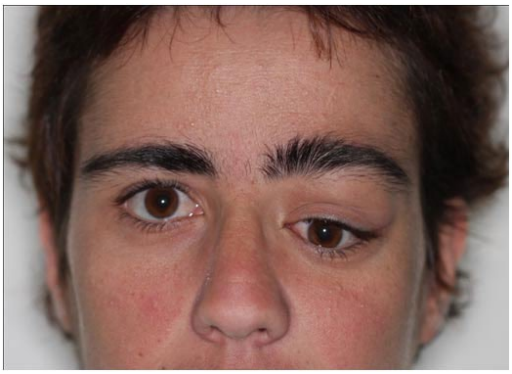

Figure 1: Pre-operative clinical photograph of patient.

On ophtalmic examination, visual acuity was 20/50 OD and 10/200 OI. Intraocular pressure was 14/16 mmHg. Left blepharoptosis and edema, proptosis of the globe, and an associated enlarging subcutaneous mass of the left frontal scalp were noted. Left hypotropia and restriction of left upgaze were present. The left macula demonstrated horizontal choroidal folds mainly in the superotemporal region. The left optic disc was pale with sharp margins more evident nasally. Pre-retinal haemorrhages secondary to compression were also evident.

Computed tomography (CT) revealed a heterogeneous mass of left frontal area with erosion of inner and outer table of the skull and roof of the orbit. The lesion extended into the superior orbit, causing depression of globe. No brain edema was present. Magnetic resonance imaging (MRI) showed a large heterogeneously enhacing extra-axial mass centered at the left frontal calvarium and roof of the left orbit. The mass showed areas of internal hemorrhage (Figure 2). CT and MRI also demostrated the presence of 2 more asymptomatic tumors with similar characteristics in maxilla and mandible.

The initial treatment involved the correction of hyperparathyroidism, which usually leads to tumor regression. The patient underwent a total parathyroidectomy identified in the usual location by scintigraphy, with auto-transplantation of parathyroid tissue. However, the orbital lesion progressively increased in size and there was not regression in the proptosis of the right eye, as confirmed by a CT performed 6 months after parathyroidectomy. We did not observe changes in maxilla and mandible lesions and they remained asymptomatic.

The resection of the fronto-orbital mass was planned with virtual pre-operative surgery that allowed manufacturing of a specific implant to accurately fit to the defect during the one stage surgery (Figure 3).

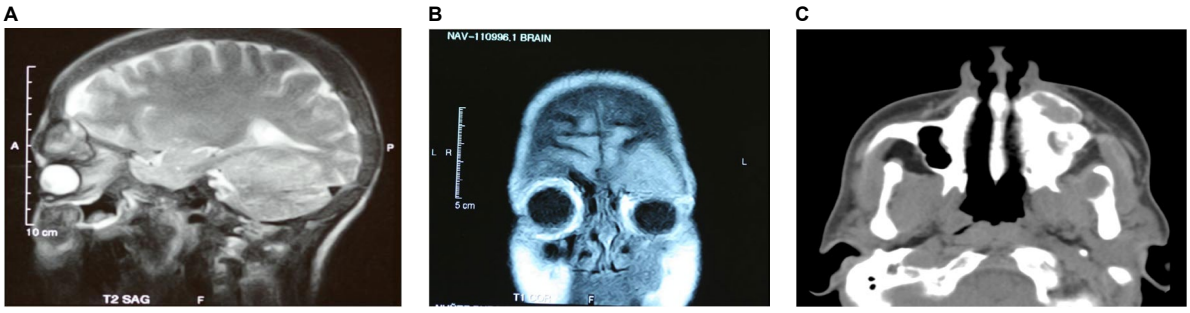

Figure 2: Sagital (A) and coronal (B) MRI that shows a heterogeneous mass of the left frontal calvarium with erosion of inner and outer table of the skull and (C) roof of the orbit.

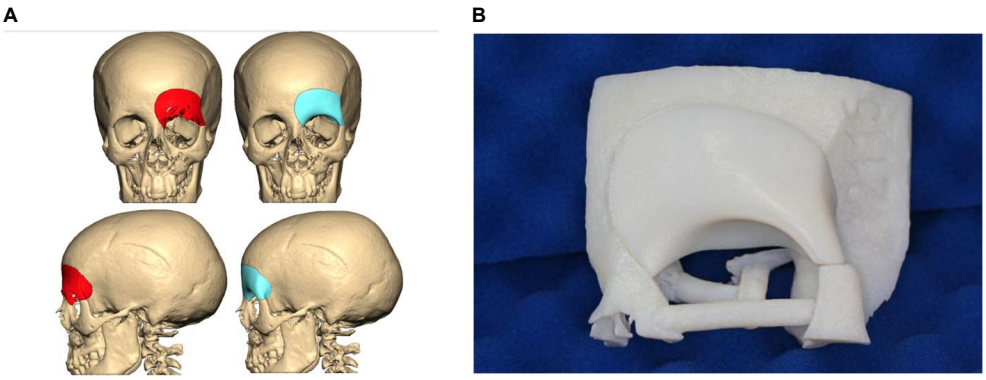

Figure 3: Planning of the resection (A) and implant design (B) sent by the manufacturer for approval.

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A pre-operative high-resolution 3-D CT scan was first obtained from axial images. The images (DICOM format) were sent by CD to the manufacturer. An accurate delineation of the lesion was performed on the software, and based in our measurements, a PEEK prosthesis was manufactured. Implant models were sent to the surgeon for review, mark up and/or approval. The resulting implant had an accuracy to within 0.5 mm.

Surgery was performed *via* a coronal approach. A cranio-orbitotomy allowed to completely excise the large, necrotic, heterogeneous mass. As planned, we performed primary reconstruction of the fronto-orbital defect with a custom-made PEEK prostheis, that was fixed with titanium plates and screws (Figures 4 and 5).

Histopathologic examination demonstrated groups of osteoclast type multinucleated giant cells in a well vascularized, cellular fibrous stroma. There was hemorrhage and cluster of hemosiderin laden macrophages. Reactive woven bone, which displayed osteoblastic activity, was seen in some áreas (Figure 6). Based on the thorough diagnostic work-up including medical history, clinical manifestations, radiographic findings and consecutive routine laboratory findings, the patient was diagnosed as having hyperparathyroidism with brown tumors of facial bones as a result of long-standing renal disease.

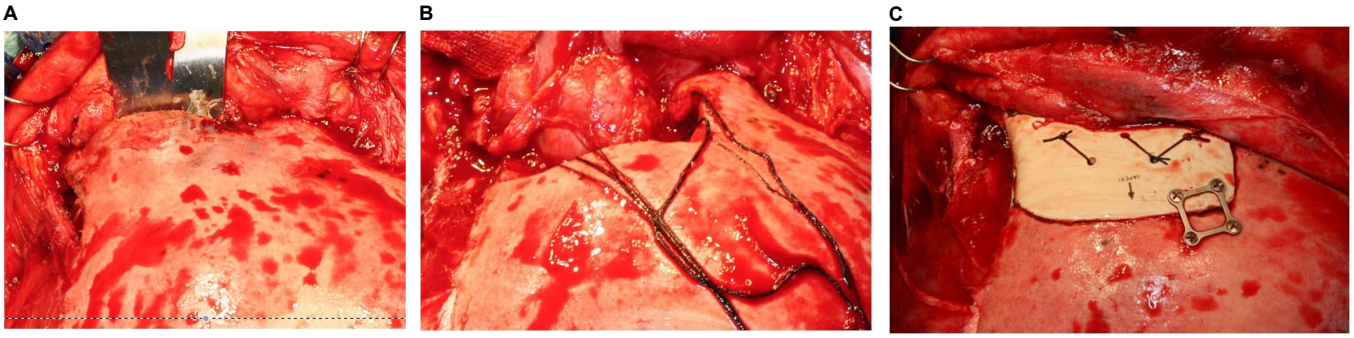

Figure 4: Intraoperative view showing the brown tumor (A), resection (B) and PEEK patient specific implant (PSI) perfectly fitting the bony defect (4C).

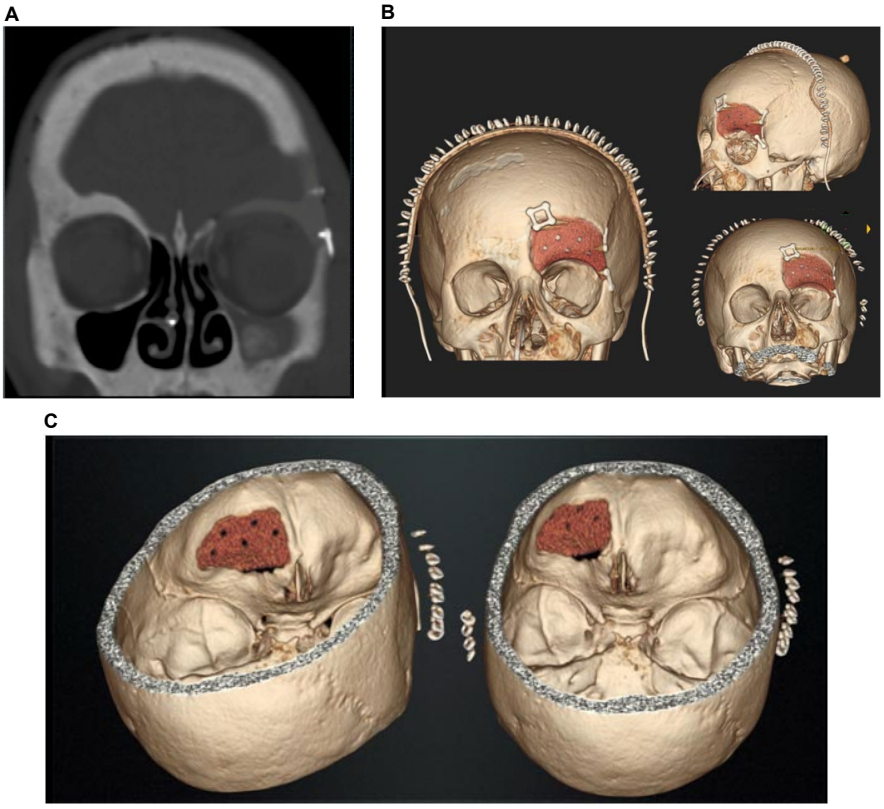

Figure 5: Immediate post-operative CT scan (A) and tridimensional reconstruction of the prosthesis (B and C).

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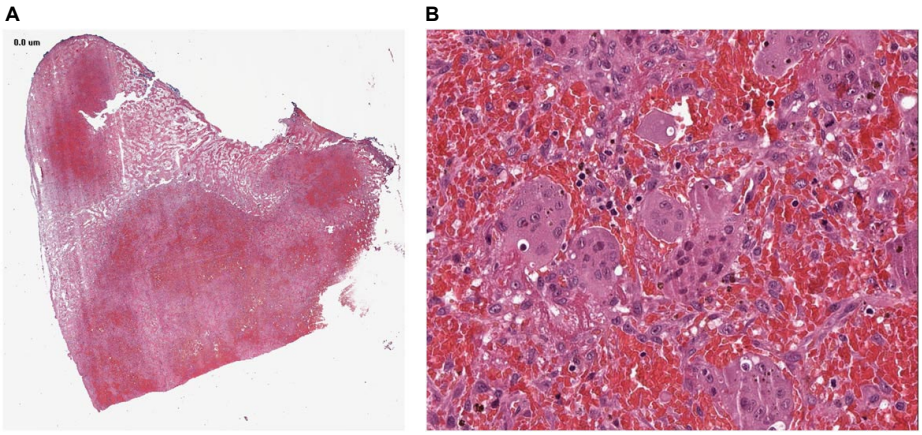

Figure 6: Histopathologic examination revealed a tumor composed of a uniform distribution of multinucleated giant cells in a background of oval to short-spindled stromal cells. These findings were consistent with a giant cell tumor and, in the setting of secondary HPT, the diagnosis of Brown tumor was made. (Figure 6A: H&E x1, Figure 6B: H&E x 200)

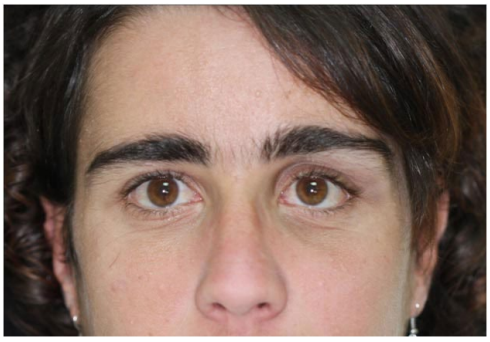

Figure 7: Post-operative image, 2 years after surgery, with good cosmetic and functional results.

Two years later, there was no recurrence of the brown tumor, exophthalmos correction and orbital contour symmetry (Figure 7).

DISCUSSION

Brown tumor or osteoclastomas are histologically benign focal lytic bone tumors caused by primary or secondary hyperparathyroidism.[18] They are called brown tumors due to the high hemosiderin level of localized accumulation of osteoclasts, vascularity and hemorrhage, that gives characteristic brown color.[19]

These tumors arise secondary to both primary and secondary hyperparathyroidism. They have been reported to ocurr in 4,5% of patients with primary hyperparathyroidism and 1,5 to 1,7% of those with secondary disease.[20] Secondary HPT is an adaptive response to chronic kidney disease (CKD) as a result of a disruption in serum phosphorus, calcium, and vitamin D homeostasis. Chronic renal disease with secondary retention of phosphates leads to hyperphosphatemia. Deficient 1,25 (OH) vitamin D3 related with renal failure and the resulting hypocalcemia induces to secondary hyperparathyroidism.[21-23] The increase of secretion of parathyroid hormone stimulates the osteoclastic activity with cortical thinning, subperiosteal resorption, bone cysts, and rarely, brown tumors. These tumors represent a reparative cellular process rather than a true neoplasia.[21]

The reported prevalence of brown tumor is 0.1%.[24] They usually affect young people, particularly females. The ribs, clavicle, pelvic girdle, hand, and mandible are their preferred location. The most important complications of this neoformation are related to its position and size and the possible effects on nearby structures.[22] Only few cases have been reported in the fronto-orbital region.[9-17] When the orbit is affected, the presenting symptoms include a palpable mass, pain, proptosis, diplopia, impaired extrinsic ocular motility or decreased visual acuity.

Diagnosis of BTs depends on clinical, biochemical, radiological, and pathological factors.[25] These tumors do not have specific imaging characteristics; sarcomas as well as giant cell reparative granulomas, langerhans cell histiocytosis, aneurysmal bone cysts, metastases, multiple myeloma and non-ossifying fibromas are the other lesions that should be kept in mind as the differential diagnosis.[25-27]

Histologically there is no difference between a brown tumor and a central giant cell granuloma. They contain a mixed population of multinuclear giant cells, mononuclear cells, and osteoblasts. There is high nuclear activity without nuclear

atypia. Extravasated red blood cells (RBC), areas of hemorrhage and hemosiderin-loaded macrophages usually appear.[28]

The treatment is initially based on the correction of hyperparathyroidism, which usually leads to tumor regression. Surgical excision of the diseased parathyroid gland to control parathyroid hormone (PTH) is the first choice of treatment for a brown tumor because the normalization of parathyroid function and should lead to a reduction in size or disappearance of the tumor.[29]

However, persistent disease is defined and accepted by the authors as the reappearance of typical symptoms, laboratory abnormalities and radiological signs.[30] Surgical resection is indicated in cases of encroachment on neural structures, and when the mass continues for more than 6 months or even grows despite adequate metabolic control.[10,20] In this case, the patient returned with an enlargement of the fronto-orbital lesion 6 months after parathyroidectomy.

Bony lesions in the fronto-orbital region often require extensive bone resections. Reconstructive cranioplasty of such defects remains a challenge for craniofacial surgeons and neurosurgeons. The anatomical complexity of this area requires restoration of the forehead and orbital walls with perfect symmetry to provide good functional and aesthetic results.

Several techniques for fronto-orbital reconstruction in this setting have been described. Autologous bone graft is the standard technique for craniofacial reconstruction in many settings. Donor site morbidity and increased operative time must be the considerations in this approach, and the rigidity of this material may increase the technical challenges of creating a conformational reconstruction of the bony orbital skeleton.[31] Due in part to these limitations, some surgeons have advocated orbital reconstruction with the use of preoperative conformation of biomaterials.[32]

In our case, a 3D CT-scan reconstruction andvirtual surgery allowed planning of the appropriate resection, and helped design the implant. Consequently, an optimal custom-made implant could be used immediately following the surgical resection. We choose to use PEEK biomaterial for this type of craniofacial reconstruction because of numerous advantages: excellent mechanical and chemical properties, biocompatibility and radiographic translucency.[33] Moreover, thin parts can be manufactured on the implant to reconstruct orbital walls for example, and PEEK can be drilled for rigid plate fixation to the maxillofacial skeleton. These implants have the advantage of being preoperatively tailored to the exact size of the cranial defect, thus allowing a shortening of the operative time as well as the amount of the intraoperative modifications, hence guaranteeing post-operative stability and incomparable cosmetic results.[34] In our case, PEEK implant allowed one-step reconstruction of the fronto-orbital region achieving symmetry and functional results, reducing operative time and avoiding donor site morbidity.

CONFLICTS OF INTEREST

The authors declare that they have no conflicts of interest.

CONSENT

The patient has provided written permission for publication of the case details.

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