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Giant craniofacial osteoma with orbital invasion

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Abstract:

Craniofacial osteomas, which involve orbits, can cause several problems, such as displacement of the globe, diplopia, and refractive changes. We report the case of a young man with diplopia and blurred vision, with a giant osteoma occupying right frontal and ethmoid sinuses, encroaching into his right orbit. The symptoms resolved 3 months after surgery. Our case highlights the successful surgery for a symptomatic craniofacial osteoma with orbital invasion.

Keywords:

Craniofacial, orbital tumors, osteoma

Introduction

Craniofacial osteomas are benign, slow-growing bone-forming neoplasms which mainly involve unilateral frontal and ethmoid sinuses and nasal cavity.^[1] Less than 10% of patients were found to have bilateral craniofacial osteomas. They are usually asymptomatic with no sex predilection. The most common clinical features are a headache, nasal obstruction, and facial asymmetry, including globe displacement and visual loss.^[2] So far, very few cases of craniofacial osteomas involving orbits have been reported. Larrea-Oyarbide *et al.*^[3] reported a review that included 106 patients with 132 craniofacial osteomas between 1986 and 2003. Most of their craniofacial osteomas were found over the mandible area. In a study conducted by Gundewar *et al.*,^[4] 12 cases were diagnosed with craniofacial osteomas in 8 years. Only 4 out of 12 patients had orbit involvement. Surgery is mandatory when complications happen due to enlargement of the craniofacial osteomas, which include proptosis, epiphora, diplopia, or intracerebral invasion.^[5] Previous studies^[4,6,7] reported the transcranial or transnasal approaches

to excise the cranial osteomas. Herein, we describe a successful case of the removal of orbital craniofacial osteoma by anterior orbitotomy without the need for transnasal endoscopy or craniotomy.

Case Report

A 19-year-old male presented with binocular diplopia for half a year and refractive change over his right eye for the past 1 year. There was no known systemic medical illness or ocular diseases. At our clinic, we observed downward and outward deviation and protrusion of his right eye with a difference of 2 mm by Hertel exophthalmometer and limitation of upgaze [Figure 1]. The pupils were round without relative afferent pupillary defect signs. The funduscopy examination revealed pinkish disc, normal vessels, but certain retinal folds over the posterior pole of his right eye. The visual acuity was 20/20 for both eyes under full correction. The Hess chart examination revealed upgaze limitation in his right eye [Figure 2]. The computed tomography (CT) of the orbit showed a giant osteoma (34 mm × 20 mm × 3.5 mm) partially occupying right fronto-ethmoid sinus, extending the two-third depth of the medial orbital wall, and pushing away the

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optic nerve [Figure 3a and b]. The patient underwent surgery for tumor removal.

We used the transcaruncular approach of anterior orbitotomy for tumor removal. Under general anesthesia, we removed the lateral orbital rim to gain enough space for the medial wall approach. The caruncle was incised, and we found pedunculated mushroom-shaped hard radiopaque ivory-like masses, along with a broad-based part, attached to the cortical plates of fronto-ethmoid bone. The tumor [Figure 3d] was separated from the cortical plate of ethmoid-frontal sinus by a chisel and a hammer, followed by sculpting the surface with a burr. Pathology confirmed the diagnosis of osteoma [Figure 3c].



Figure 1: Mild proptosis, exotropia, and inferolateral displacement of patient's right globe

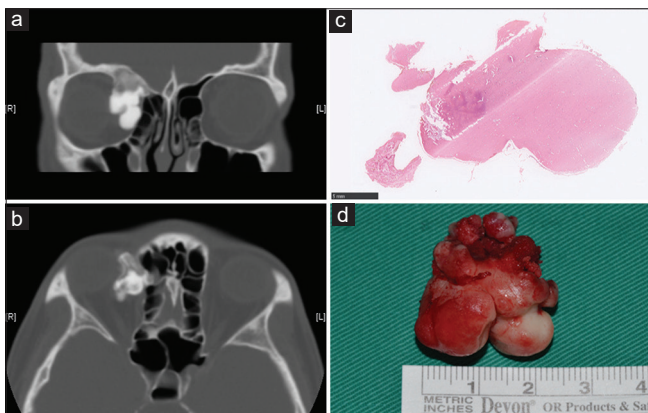


Figure 3: (a and b) A bony lesion over right frontal and ethmoid sinus, encroaching into right orbit. (c) On microscopic examination, the osteoma consisted of dense and lamellar cortical bone. (d) The specimen obtained is a well delineated neoplasm composed of mature compact bone and broad trabecular bone measuring up to 32 mm × 25 mm × 15 mm

The first month after operation, the patient had upgaze restriction and slight enophthalmos, which resolved spontaneously after 3 months. Postoperative orbital CT studies showed a static residual osteoma inside the frontal sinus without evidence of tumor recurrence in the following 2 years [Figure 4a and b]. The patient had continued clinical stabilization at the 5-year follow-up.

Discussion

Most of the craniofacial osteomas are detected incidentally due to the small tumor size and the slow-growing nature. The pathological picture in our case showed an ivory-hard, dense, mature bone with the total absence of Haversian canals, which was in line with the slow-growing osteoma described by Earwaker.^[8] Osteomas of paranasal sinuses >30 mm in size or weighing more than 110 g are called giant osteomas.^[2] Giant osteomas have a higher incidence of intraorbital extension, which might cause ocular symptoms such as globe displacement, diplopia,

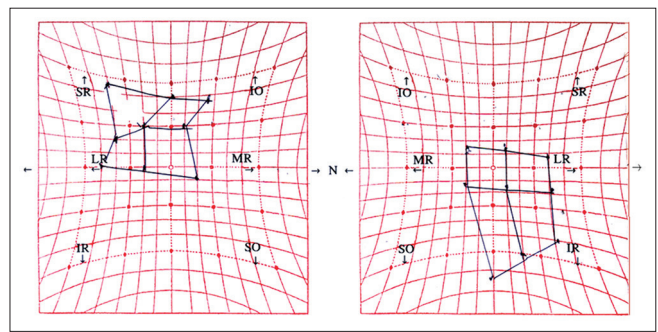


Figure 2: Hess chart examination revealed upgaze limitation of patient's right eye



Figure 4: Orbital computed tomography: Stabilized residual osteoma inside the frontal sinus without recurrence of tumor was noticed 2 years after surgery. No recurrence of tumor in ethmoid sinus. (a) Coronal view. (b) Axial view

visual disturbance, epiphora, or proptosis, and should be treated surgically. Some studies suggest surgical removal of osteoma if it has a rapid growth, defined as the growth of more than 1 mm/year, more than 50% involvement of the frontal sinus volume, or location at the frontal recess in asymptomatic cases.^[2,5] Others suggest close observation with repeat CT scans every 12 months.^[9] A review of the literature to compare transnasal endoscopic removal or external excision of osteoma had found that the average size and the location of osteomas were not different using various approaches.^[6] The choice of surgical method was at the discretion of the surgeon's experience. Craniofacial osteomas were removed endoscopically from sinuses or through craniotomy commonly. In the presented case, the osteoma is extensive and involves the orbit with lateral extension making an endoscopic approach difficult.

We introduced a novel and feasible method to excise the tumor by anterior orbitotomy. In this case, we removed the osteoma through anterior orbitotomy, through an incision over the caruncular area. The space between the medial orbital wall and orbital tissue was created by removing lateral orbital rim in advance, which enables us to excise the intraorbital part of the osteoma with a clear view. We removed the tumor easily from the ethmoid side. On the frontal side, we carefully separated it from the orbital roof by using surgical chisel and burr. The reason that we did not attempt to remove the entire osteoma was the risk of postoperative sinusitis or intracranial infection. Recurrence of osteomas is rare after surgical removal. Only 10% of recurrence had been reported due to incomplete removal of the osteomas.^[10]

Most literature reporting giant orbitoethmoidal osteomas are case reports or small series. Therefore, the superiority of one surgical approach over the other is not yet established. However, in the presence of gigantic lesions with lateral extension, as observed in our case, the transorbital approach is feasible and effective. Complete surgical removal is not mandatory, and partial sculpting may relieve symptoms such as diplopia, reduce surgical morbidity, and preserve the visual acuity.

Declaration of patient consent

The authors certify that they have obtained the required patient consent forms. The patient and his parents have

provided informed consent for publication, for his images and other clinical information to be reported in the journal. The patient understands that his name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

The authors declare that there are no conflicts of interests of this paper.

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