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Giant Frontal Sinus Osteoma Presenting as Abducens Nerve Palsy with a Local Granulomatous Inflammation: Case Report with a Literature Review

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Abstract

Osteoma is one of the rare benign bone tumors and often presented asymptomatic. The current article will present a case report of giant frontal sinus osteoma and successful surgical treatment. The size of the tumor described is remarkable and among the largest currently reported. In a discussion section, we try to provide the helpful groundwork for this disease's etiology and describe the surgical approach performed. The purpose of this study is to present the clinical symptoms, radiological findings, and surgical indications of a giant frontal sinus osteoma with an orbital cavity expansion and secondary VI cranial nerve palsy. Moreover, histological findings of the tumor showed granulomatous inflammation and tumor-infiltrating lymphocytes with single multinucleated giant cells, leading us to provide additional antibacterial treatment and perform an osteoplastic operation 6 months later.

Keywords: giant osteoma, local inflammation, osteoplasty.

Introduction

Osteomas are benign, slow-growing bone tumors primarily developed from the paranasal and craniofacial sinuses [1]. Only 0.03% of primary bone tumors that have undergone biopsy are osteomas [2]. Osteomas in the orbit can present with various symptoms depending on their location and size. Due to the limited space within the orbit, the growth of an osteoma can lead to compression of the surrounding structures, resulting in symptoms such as proptosis (bulging of the eye), restricted eye movement, diplopia (double vision), and visual disturbances. Furthermore, in cases where the osteoma extends into the sinus cavity, it can cause sinus congestion, facial pain, and deformity [1, 3]. Recurrence is quite rare: according to one literature analysis that gathered data on 477 cases, recurrence or persistent residual disease of osteoma only occurred in 12 (2.5%) individuals with a mean follow-up of 29.7 months [3]. Primary bony tumors of the skull make up only 1% of all bone tumors, and only 25% of all craniofacial osteomas were found in the orbit, whereas 17% implicated the frontal bone [4]. The cranial osteomas can be classified into skull base osteomas, skull vault osteomas, dural osteomas, and intraparenchymal osteomas, with skull base osteomas being the most prevalent among these groups [5]. Orbital extensions may be present in frontal sinus osteomas. The cranial nerves III, IV, and VI, the ciliary ganglion, and the ophthalmic sympathetic nerve fibers are all located within the relatively small cavity of the orbit, which is a bony pyramid with an approximate volume of 30cm3 [6]. An orbital extension that exceeds 50% of the orbit may cause irreparable harm, such as permanent vision loss [6]. In this case, the right eye's proptosis and VI cranial nerve palsy were caused by the protrusion of the tumor into the orbital region. This case report aims to provide a successful case of resected frontal sinus osteoma that expanded to the roof and lateral walls of the orbit.

Case description

A 31-year-old female with a history of minor head trauma at the age of 2 and hepatitis B arrived at the National Center of Neurosurgery in April 2023 with bulging in her right eye. For the previous five years, the patient had been suffering from terrible widespread headaches. Other complaints included swelling, pain in the right eye, and proptosis that progressed during the last 2 years. During the neurological examination, VI CN palsy was observed in the form of horizontal diplopia.

The CT and MRI scans were performed, revealing signs of the osteoid formation of the right frontal sinus and the orbit's destruction. Periorbital edema and proptosis (Figure 1 C, D) were seen, and a performed MRI revealed that the right eye was dislocated to approximately 6.6 mm from orbit and the signs of a tumor formation in the intracranial space.

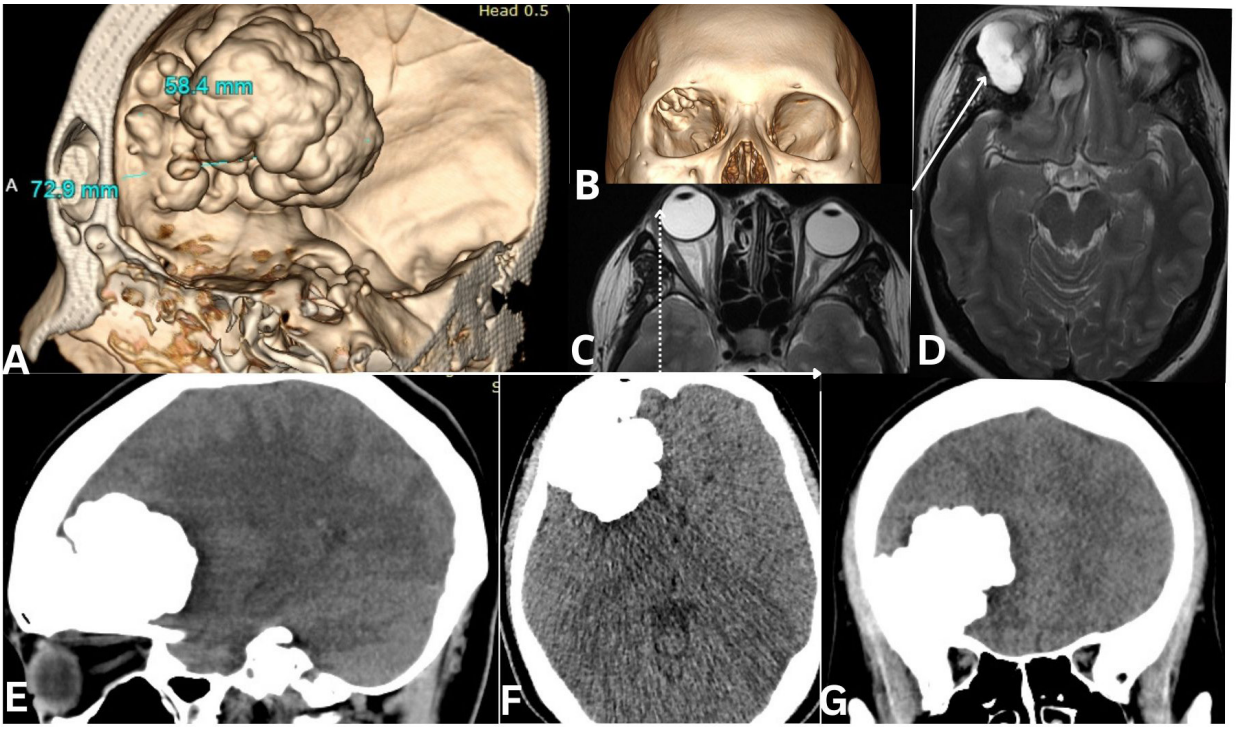

Figure 1 – Initial presentation of the patient's osteoma with measurements on CT (Figure 1a); figure 1B demonstrates invasion to the frontal bone; the dotted line on figure 1C demonstrates the proptosis, and an arrow on figure 1 D shows sign of granulomatous inflammation. There are signs of the orbital destruction (Figure 1E, 1F, 1G)

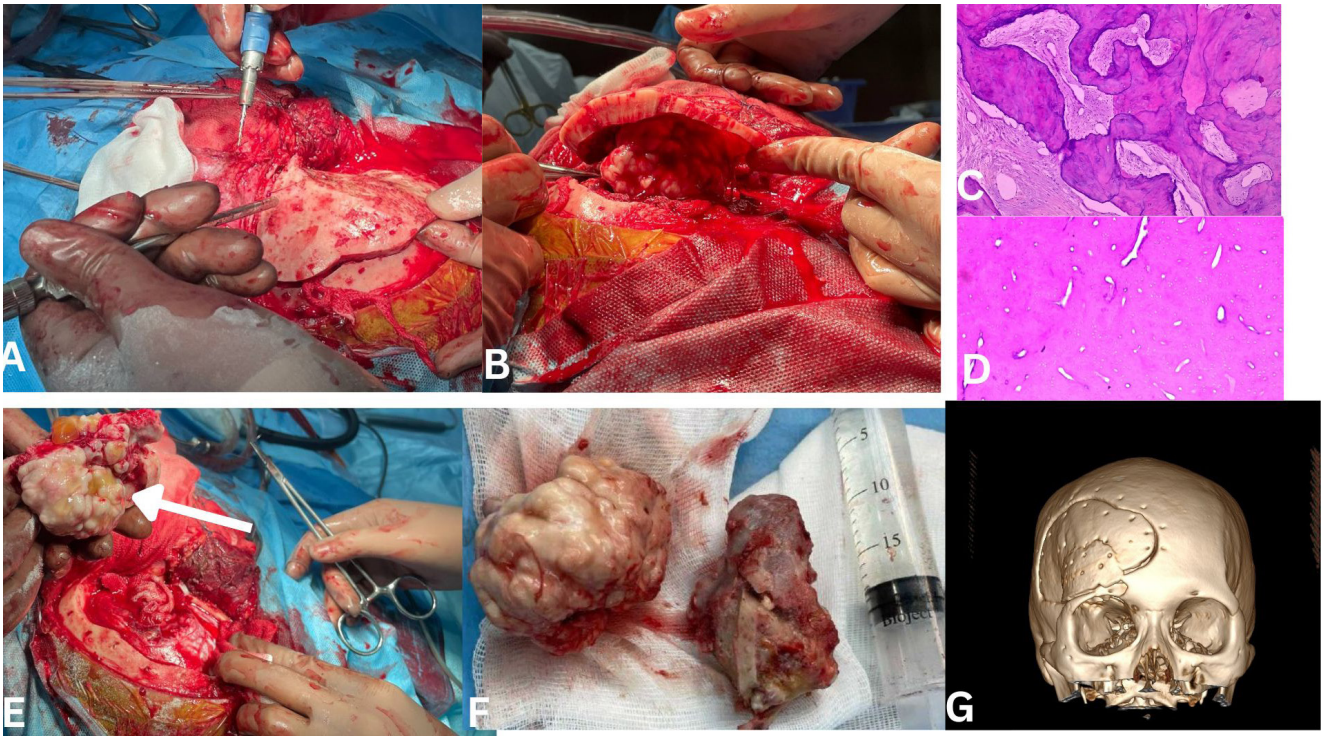

Figure 2 – Intraoperative images demonstrating modified frontotemporal approach (figure 2A), bone elevation with detachment of the osteoma from dura mater by a dissector (figure 2B), removed part of the bone with the tumor attached to it, local inflammatory reaction on the surface of the osteoma was seen as yellow membranous part (arrow on the figure 2E), post-resection images of the tumor (figure 2F). Histopathological evaluation with H&E stain revealed the presence of osteocytes within the dense bone matrix, with minimal fibrous stroma noted (C, D). Figure 2G demonstrates a post-operative CT demonstrated total removal with osteoplasty.

A firm mass was palpated at the angle of the right orbit. A cerebral computed tomography (CT) scan (Fig. 1E, F, G) showed a polypoid and irregularly shaped solid bony mass with well-defined borders and intracavitary growths occupying the anterior part of the frontal sinus and extending toward the intracranial surface of the orbit with thinning of the lateral wall of the frontal sinus and causing 7 mm frontal lobe displacement to the left. CT of the head revealed a 7 cm × 5.8 cm × 6 cm well-defined, lobulated, irregularly shaped solids with bony consistency markedly radio-dense lesion arising from the frontal bone and right frontal sinus. The tumor had extended upward to the right orbit's superior wall, showing significant intracranial extension. The mass had displaced the globe and the orbital soft tissues inferiorly and laterally. Supra-lateral part of the orbit had T2 hyperintensity signals (Fig. 2 D), indicating an inflammatory process beneath.

The mass was responsible for irreducible and non-pulsatile right eye proptosis of 6.6 mm with superolateral globe displacement and lateral rectus paralysis. Visual acuity and sharpness were preserved.

Operation

The patient has undergone a pterional craniotomy (Figure 2A, B), where the orbital roof destruction and dura erosion were distinguished. The right frontal sinus was opened and sealed with a hemostatic sponge after thorough washing with chlorhexidine. Further work on the olfactory fossa provided a layered reconstruction of anterior cranial fossa defects with periosteum. Another muscle flap was superimposed on top to ensure total sealing. The optic nerve was intact. The dura mater was sutured using an artificial dura. The part of the frontal lobe was partially removed and modified, the osteoma adjacent to the bone was cut out completely, and the remaining bone, not damaged by the tumor, was left in place. Because of the signs of the inflammation during the operation (fig 2E), the decision was made to perform an osteoplastic operation 6 months later. The compressed part of the right frontal lobe had parenchymal bleeding and signs of necrosis; the bleeding was consequently removed with hemostasis provided. The operation was performed without complication, and the patient regained consciousness after a few hours.

Histology

The tumor, divided into two parts, exhibited features of osteoma on histological examination. The first part consisted mainly of mature lamellar bone tissue, while the second part showed infiltration by lymphocytes and multinucleated cells resembling foreign bodies. The tumor mass, discovered post-surgery, had a polypoid shape with a smooth white and yellow surface composed of mature lamellar bone tissue with osteons (fig 2 C, D). Microscopic analysis revealed active granulomatous inflammation, and the case was classified as an ivory-type osteoma based on these findings. It is recognized that the granulomatous inflammation surrounding a tumor is an indication of the host immune system reacting to the tumor.

Postoperative Course and Follow-Up

The clinical course following the craniotomy was successful: headaches were gone, and proptosis regressed consequently. The patient was able to regain lateral gaze ocular movements. No neurological deficits were identified. There were no incisional infections, meningitis symptoms, or neurological deficits. However, the cranioplasty could not be performed immediately during the excision of the tumor because of granulomatous inflammation detected in surgery. Based on the type of surgery, the location of the osteoma with extension into the sinus, the high risk of neurological complications, and the microbial environment of the medical facility, the clinical team, in collaboration with the hospital's clinical pharmacologists, decided to implement antimicrobial therapy as part of the patient's treatment plan. Following the surgical removal of the osteoma, the patient was prescribed a course of antimicrobial therapy. The treatment plan included the administration of 1000 mg of Cefepime twice a day and 500 mg of Metronidazole once a day for 7 days after the procedure. Acetazolamide was recommended for the prevention of irregular liquorrhea. The patient was discharged with no symptoms on post-operative day 7. The post-operative 6th-month MRI confirmed no residual tumor. Six months later, the patient was hospitalized to provide a cranioplasty of the defect by a Synicem Cranioplasty (Fig. 2G).

Discussion

Osteomas are benign, slow-growing bone tumors that primarily originate from the paranasal and craniofacial sinuses [1]. These tumors account for a small proportion of primary bone tumors, with only 0.03% of biopsied primary bone tumors being identified as osteomas [2]. Our institution has encountered approximately 1-2 osteoma cases annually, but this case represents the first time we have performed surgical treatment of a sizeable osteoma. The surgery helped to normalize the patient’s proptosis and lateral gaze palsy. The traditional procedure for removing huge osteomas was the open approach, and the presence of orbital extension and the need for bone reconstructing had a big impact on the preoperative choice. The tumor was a massive 7x4x3 cm, with a smaller portion measuring just 2x1x1 cm. In addition, the osteoma expanded from the frontal sinus to the orbital cavity. The complaints about the 5-year-long headache of the patient could be associated with chronic frontal sinusitis. The enormous osteoma, which had an extensive intracranial extension and orbital bone involvement, was indicated for the pterional incision. The protection of the underlying brain and the restoration of cranial form required the reconstruction of bone abnormalities. Autologous bone grafts, titanium mesh, polyethylene sheets, and hydroxyapatite cement may be utilized [7] if an enormous part of the bone has been destroyed. In our case, we used an acrylic cranioplasty to repair the bone defect of the anterior wall of the frontal sinus.

The osteoma is a slow-growing benign tumor with a mean speed of growing 1.66mm/ per year [8]. It is hard to define the etiology of the disease and whether it could be derived from environmental, embryological, and genetic factors. Several case reports mention that the patients had head trauma long before the diagnosis of osteoma [9]. In frontal sinus osteoma reviews, 5-7% of the patients who had osteoma responded that they had head trauma earlier in their life [10]. Our patient also stated that she had head trauma at the age of 2.

Conclusions

The extended tumor with intracranial or intraorbital involvement impacted diffuse headaches proptosis of the

patient's right eye. The open surgery performed on the giant osteoma was a successful example of removing bone tumors and reconstructing the frontal sinus and frontal lobe bone. The masses were removed, and the patient no longer had partial limitation of eye movement, which was the main consequence of the expanded tumor. The lateral rectus palsy regenerated, and the ability to lateral ocular movement was regained. The damaged supraorbital part of the frontal bone was dissected and reconstructed. Surgical approaches to the orbit require a multidisciplinary approach involving neurosurgeons, ophthalmologists, clinical pharmacologists, and otolaryngology-head and neck surgeons.

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Ethic approval and consent to participate: The photographs are completely unidentified and personal details are not mentioned in the text. The authors are accountable for all aspects of the work and for ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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