

Frontal lobe mucocoele secondary to an extensive frontal sinus osteoma

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ABSTRACT

Osteomas are benign slow growing tumours of the paranasal sinuses. The frontal and ethmoidal sinuses are most commonly affected. We present an unusual case of a 42-year-old Malay lady who had a frontal lobe mucocoele, secondary to an extensive frontal sinus osteoma with orbital and intracranial extension, and presenting primarily with orbital proptosis. She was successfully managed by a multi specialty team.

Keywords: Frontal sinus, mucocoele, osteoma, proptosis, intra-cranial extension

INTRODUCTION

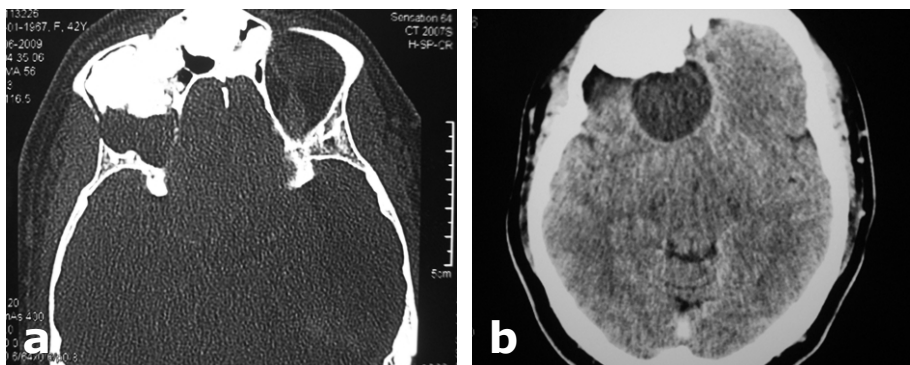
Osteomas are benign slow growing tumours of the paranasal sinuses, most commonly found in the frontal and ethmoid sinuses, and are often asymptomatic until in the advanced stages. Due to the anatomical relationship, clinical features secondary to orbital or intracranial extension is frequently the initial presentation. Radiological investigations usually confirm the nature and extent of the pathology, and may prove the first diagnostic clue in many cases. A combined management involving a multi-disciplinary team including the Oromaxillofacial, Otolaryngology, Ophthalmology and Neurosurgical specialties may be required, as extensive craniofacial expo-

sure may be needed. We report an unusual case report of a frontal lobe mucocoele, that was secondary to large frontal sinus osteoma with orbital and intracranial extension, and that was successfully managed by a craniofacial approach involving a multidisciplinary team.

CASE REPORT

A 42-year-old Malay housewife presented to the Ophthalmology clinic with a gradually increasing proptosis of the right eye of more than eight months, with restricted right eye movements, and some degree of diplopia. She also occasionally experienced a mild headache, mainly in the right frontal area. Examination revealed proptosis of the right eye, with infero-lateral displacement of the right globe and restricted movements in the superior and medial direction. There was no redness

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Figs. 1: a) A non-contrast axial computed tomography (CT) image showing a mass in the frontal lobe that is better visualised on contrast imaging (b).

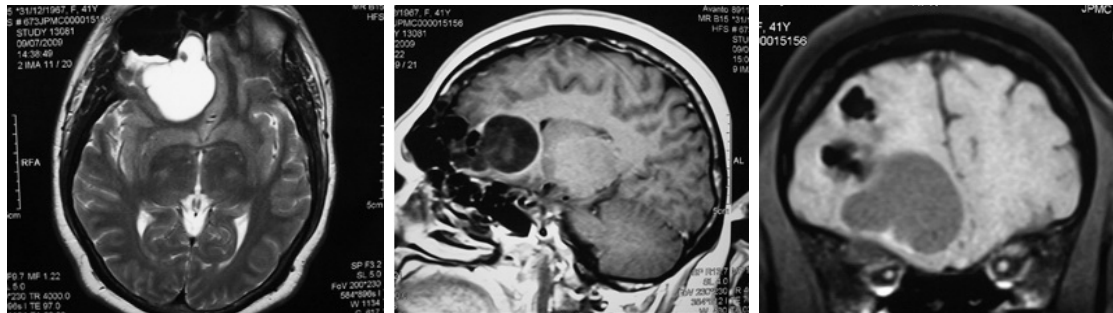
of the eye or papilloedema. She did not show any disturbance in her visual acuity.

A computed tomography (CT) scan showed a poorly enhanced calcified mass, that appeared to have originated from the roof of the orbit, filling the entire right frontal sinus and extending to the opposite side with erosion and extension intracranially in right suprasellar region in the frontal lobe area. A secondary smooth swelling was also found extending into the frontal lobe area which appeared to be a mucocoele (Figures 1). The bony mass had all the radiological features of an osteoma. In view of the clinical picture and the CT findings, the patient was referred to the Oromaxillofacial (OMF) and Ear, Nose and Throat (ENT) departments for further management.

On further questioning, there were no noticeable neurological or specific past or present ophthalmic and ENT complaints. She had no past history of ophthalmic complaints, and with the absence of colorectal or gastrointestinal complaints, the possibility of Gardner's Syndrome was ruled out. There was no visible facial swelling or nasal mass, and the scalp

area was normal. A magnetic resonance imaging (MRI) was done to further confirm the extent and nature of intracranial swelling as seen in the CT imaging. Based on the T2 weighted images, the intracranial swelling in the frontal lobe was consistent with features of a mucocoele (Figures 2). A three-dimensional reconstruction of the skull was also done to better conceptualise the anatomy and surgical trajectory.

In view of the clinical and radiological findings that were suggestive of an extensive frontal sinus osteoma with a secondary frontal lobe mucocoele, a combined external fronto-ethmoidal approach with osteoplastic frontal sinusotomy was planned to remove the osteoma and the mucocoele at the same time. This was planned as a combined multi-disciplinary surgical team approach that included the OMF, neurosurgery, ENT and Ophthalmology. The surgical approach was done via a bicoronal forehead flap with reconstruction of defects: appropriate bone grafting and customised titanium plate cover to prevent any cosmetic defect, and fascia lata to cover base skull defect in the posterior plate of frontal sinus after mucocoele removal



Figs. 2: Pre-operative magnetic resonance imaging (MRI) showing the osteoma and mucocoele in a) axial (T2 weighted image), b) sagittal, and c) coronal views.

(Figures 3).

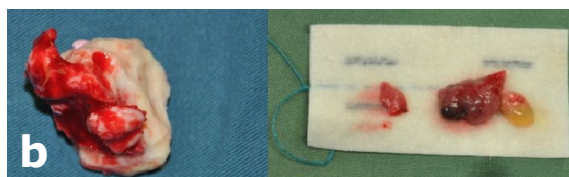
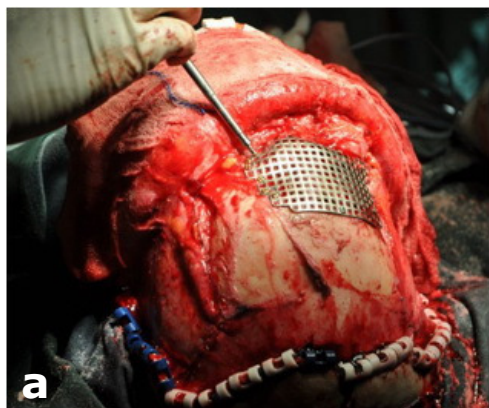
The patient had an uneventful recovery in the postoperative phase, with no neurological deficits or visible cosmetic deformity, and improvement of eye movements. Follow-up in the past 28 months has so far been unremarkable, and serial follow-up CT scans showed no evidence of residual or recurrent disease.

DISCUSSION

Osteoma is the commonest primary bone tumour in the craniofacial skeleton that not only involves the paranasal sinuses but also the mandible, external auditory canal and mastoid air cells.^{1,4,5} It is the most common benign tumour arising in the paranasal sinuses.

The frontal sinus is most commonly affected with incidences ranging from 37% to 80% in various reports.^{1,6} Buyuklu *et al.* reviewed 17,154 CT scans of the paranasal sinuses and found 243 cases of osteomas with 75.3% located in the frontal sinus, followed by the ethmoid (20.5%), sphenoid (2.9%) and maxillary sinus (2.1%).⁴ Erdogan *et al.* found the ethmoidal sinuses (55%) to be the most frequent location for osteoma, followed by the frontal sinus (37.5%), maxillary sinus (6%) and sphenoid sinus (1.5%).¹ However, this study only included 1,889 CT scans of paranasal sinuses.

The aetiology of osteomas remains debatable. Commonly accepted theories include those that are embryologic, traumatic or infectious in origin.¹ Given that many osteomas appear to rise at the junction of the



Figs. 3: a) Intraoperative image showing placement of a metal mesh, and b) resected specimen showing the osteomas and the mucocoele.

ethmoid and frontal sinuses, a location where membranous and cartilaginous tissues meet during embryonic life, the embryonic origin theory is favoured. Trauma in early life until puberty, when the bone is still developing, has also been implicated.⁶ Sinusitis stimulating osteoblastic proliferation in the mucoperiosteal lining of the sinuses has also been postulated as a possible cause of osteoma. However, it is widely believed that sinusitis occurs later, secondary to the osteoma occluding the sinus ostium.⁶

Osteomas are generally detected in the second to fifth decades of life.³ The ratio of males to females is about 2 to 1. The greater preponderance of sinus osteomas in males is attributed to their greater exposure to trauma, and the larger size of their sinuses.

Paranasal sinus osteoma is a slow-growing, encapsulated bony tumour that is commonly asymptomatic. It may be first detected when it has outgrown the confines of the sinus to invade the surrounding structures such as the orbit or into the intracranial plane. This may result in nonspecific symptoms such as headache or facial pain or even facial asymmetry. Frontal sinusitis, secondary to blockage of the sinus ostia is another common presentation. Presentations such as our patient are rare. A predominant intraorbital extension can lead to proptosis, diplopia and global displacement with restricted eyeball movements. On the other hand, intracranial complications (pneumatocoele, mucocoele, abscess formation, meningitis and cerebrospinal fluid rhinorrhoea) may also occur as a result of extensive destruction of the anterior cranial fossa.⁶ Cranial nerve palsies, raised

intracranial pressure, and obstruction of the nasolacrimal duct and nasal passage can occur in longstanding cases, albeit rare. Most cases are diagnosed only after radiological investigations such as plain radiography or with more advanced imaging such as CT scan or MRI.^{3, 4}

Characteristic CT findings of osteomas include high density mass with well defined borders that lack contrast enhancement, and some lytic bone destruction. CT scan is distinctly more advantageous over MRI for evaluating bony extent and destruction.⁶ However, MRI helps to delineate the extension outside the sinuses, including intracranial complications, as was seen in our case.¹¹

The occurrence of osteomas in the setting of Gardner's syndrome, a rare autosomal dominant disorder (associated with high-risk intestinal polyps, pigmented skin lesions and multiple osteomas) must be considered.^{1, 6} Investigations of the gastrointestinal tract may need to be considered. In our case, as our patient did not have any gastrointestinal symptoms at presentation and on follow-up, we have not yet carried out any evaluation.

In the management of such cases, three surgical approaches have been described for the treatment of frontal sinus osteoma: the frontoethmoidal supraciliary approach, the endonasal (possibly endoscopic) approach, and the bicoronal approach.^{2, 10} Even though the growth rate of osteomas are relatively slow, surgical management becomes mandatory when it obstructs sinus ostia producing symptoms, or extends to the adjacent bones, intracranial plane or orbit. The choice of operative approach and extent

of the osteotomy depend on the size and extent of the tumour, its relation to the anterior and posterior walls of the frontal sinus, the dimensions of the affected sinus, and also the extent of tumour outside the confines of the sinus cavity. Extension into the orbit and cranial cavities are also important considerations. For large osteomas of the frontal sinus, particularly with orbital and/or intracranial extension, as is our present case, a bicoronal approach is preferred.⁸ This approach provides an excellent surgical exposure for complete removal of the neoplasm. It allows approach to any intracranial extension, or deal with secondary complications like frontal mucocele through the same exposure. It also limits intra- and postoperative complications, reducing the chance of recurrence, and gives an acceptable aesthetic result.⁹

In conclusion, our case highlights a rare manifestation of osteomas of the frontal sinus that presented with an extensive secondary mucocele in the brain. In the management of such conditions, it is important to be cognisant of the fact that a multidisciplinary team approach is required for the best treatment outcome.

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