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Case Report

Cemento-ossifying fibroma: an extensive fibro-osseous lesion involving paranasal sinuses

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ABSTRACT

Cemento-ossifying fibroma (COF) is a fibro-osseous lesion arising from cells of the periodontal ligament. This tumor typically occurs in individuals during their third and fourth decades of life, with a predilection for women. COF primarily affects the maxillofacial region, most commonly the mandible. Its localization in the paranasal sinuses and skull base is unusual. However, we present a rare case of COF involving the right paranasal areas of the maxillofacial region in an 18-year-old female. Computed tomography (CT) revealed an expansile lesion measuring 52.45×32×74.29 mm involving the right maxillary sinus, expanding into both the anterior and posterior choanae. The patient underwent endoscopic sinus surgery (ESS) with preservation of adjoining vascular structures. Clinical, radiographic, and histologic features are presented, and differential diagnosis is discussed.

Keywords: OF, COF, Paranasal sinus, ESS, CT

INTRODUCTION

Cemento-ossifying fibroma (COF) is a benign mesenchymal odontogenic lesion characterized by well-circumscribed, unilocular radiolucency mixed with radiopacity based on presence of mineralized tissue either cementum or bone histologically.[1-3] COF is believed to derive from the cells of the periodontal ligament and multipotential cells capable of forming cementum, lamellar bone and fibrous tissue.[1-4] Though the global prevalence is only 3.1%.[5] The aggressive nature of lesion necessitates the need for its early intervention. This case report highlights a case of COF fibroma in the paranasal areas of the maxillofacial region, accompanied by clinical, radiological, and histopathological findings, and its management through ESS.

CASE REPORT

An 18-year-old female patient, convoyed by her mother and with no relevant medical history, reported to the ENT department with a swelling on the right side of her facial region for the past one year, with an occasional pain. The patient also complained of occasional blood in her sputum, along with intermittent breathing difficulty. The mass had been gradually increasing in size, reaching its current dimensions over the last six months.

Physical examination revealed enlargement in the ethmoid and maxillary area, facial asymmetry (Figure 1 A and B) and right eye proptosis. The oral mucosa appeared normal, with no erythema or surface ulceration of the swelling. The neurological examination findings and visual acuity were normal. CT of PNS + orbit revealed a large heterogeneous expansile lesion in the right paranasal sinuses extending superiorly into the frontal sinus via ethmoidal cells and inferiorly into the right nostril and maxillary sinus (Figure 2 A and B). Posteriorly it extended into the sphenoid sinus. A significant mass effect was seen on the right frontal lobe. There was a remodelling of the right orbit due to the expansile mass-effect by the intranasal mass causing

significant mass effect on the lamina papyracea. The overall size of the lesion measured 52.45×32×74.29 mm (Figure 3 A and B). The patient underwent an ESS for the said mass involving the paranasal sinuses under general anaesthesia. Significant component of the mass present in the right frontal sinus had to be removed with an angled endoscope (70-degree scope). As mass had remarkable vascularity. Bipolar suction cautery and surgical were needed for haemostasis. The anterior ethmoidal artery was bipolarized as it was engulfed in the mass (to prevent its retraction into orbit and its associated complications). Intraoperative findings included a tan-white, gritty mass (Figure 1 C) that distorted the surrounding anatomy followed by placement of an anterior nasal pack. Postoperatively, patient received an antibiotic coverage and a dose of tranexamic acid. Blood parameters at the time of discharge were normal and follow up visit was uneventful. However, HPE showed fibro-osseous lesion with hyper and hypocellular areas, composed of whorls of spindle to oval cells. Numerous rounded to irregular cementum like/psammatoid calcific foci and few osteoclast like multinucleated giant cells were also noted in the stroma (Figure 2 A and B). Thus, co-relation with clinical, radiological and histopathological findings lead to a diagnosis of COF in the right facial region.

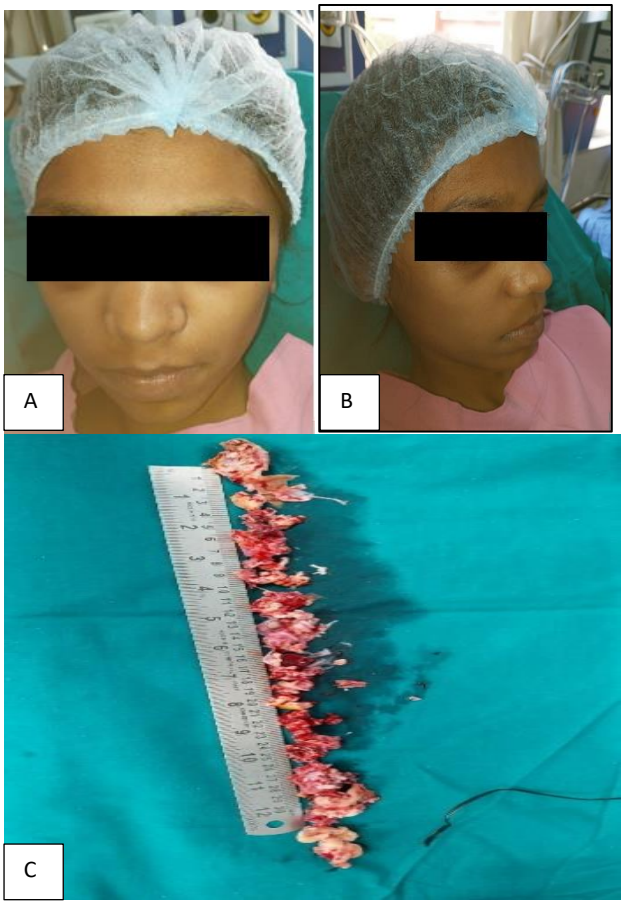

Figure 1 (A-C): Clinical images of the patient preoperatively and post-operative biopsy specimen.
Frontal view (Pre-op. Lateral view swelling extending upto the orbital region. (Pre-op) and biopsy specimen (Post-op).

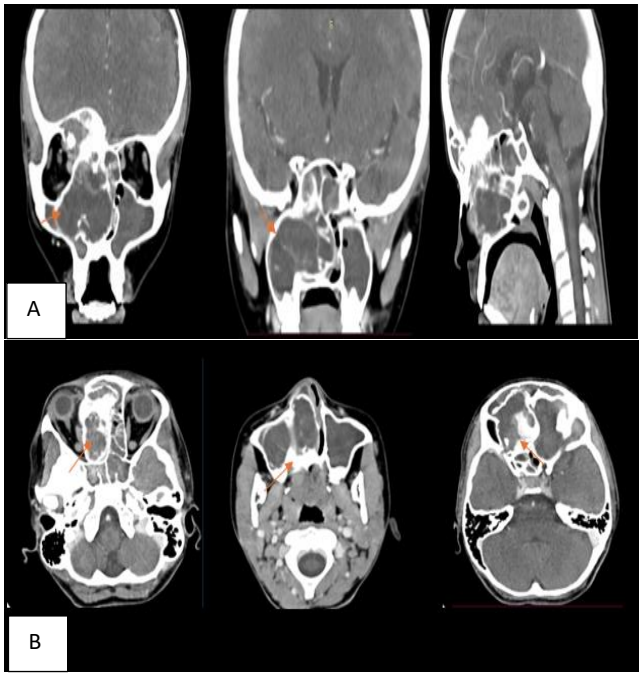

Figure 2 (A and B): CT of (Para nasal sinus (PNS)+ orbit).
Fibro-osseous lesion involving the paranasal sinuses viz. maxillary sinus and frontal sinus (coronal view and sagittal view). Fibro osseous involving the paranasal sinus viz. sphenoid sinus and ethmoid sinus (axial view).

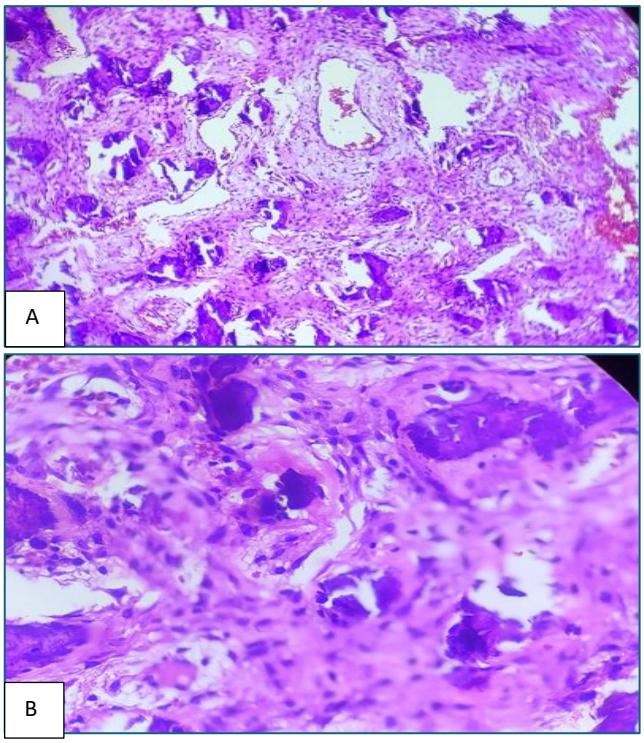

Figure 3 (A and B): Histopathological image of the excised mass (Haematoxylin-eosin stain).
Histopathology image: Fibro-osseous lesion revealing hypercellular to hypocellular areas showing whorls of spindle to oval cells. Histopathology image: cementum like rounded structures amidst fibrous stroma.

DISCUSSION

Ossifying fibroma is a circumscribed, indolent, non-malignant fibro-osseous neoplasm comprising fibrous stroma intermingled with varying quantities of metaplastic mature osseous tissue.[6] The world health organization (WHO) initially classified fibro-osseous lesions (FOL) in 1971 under lesions containing cementum, including fibrous dysplasia (FD), OF, and COF.[7] Various theories have been put forward regarding the origin of COF but current interests are oriented toward traumatic and developmental etiologies.[8,9]

The clinical manifestations of COF are diverse and contingent upon the lesion's location. The mandible is more frequently affected (93% of COF cases) than the maxilla, particularly in the premolar and molar regions. The COF may also be present in sphenoid, ethmoid, parietal, and temporal bones suggesting the presence of ectopic periodontal membrane tissue, with a female predilection.[11] The lesion may remain asymptomatic for an extended period, with patients often presenting with a painless facial swelling that can persist for several years.[10] Classically, this tumor appears ovoid in shape owing to a centrifugal pattern of growth, that is, growth from the center toward periphery.[12] These lesions are diagnosed using CT, while magnetic resonance imaging (MRI) is utilized for preoperative surgical planning. The differential diagnosis mainly includes cement-osseous dysplasia, calcifying epithelial odontogenic tumor (CEOT), calcifying odontogenic cyst, cementoblastoma, and Paget's disease.[13]

However, COF typically presents as a well-demarcated unilocular lesion containing variable amounts of radiopaque material. The density of fibrous and osseous tissues primarily determines the radiological appearance of these lesions. In advanced stages, the center of the lesion comprises mature bone tissue, whereas in the earlier stages, the center is typically soft and fibrous.[14] The treatment plan mainly includes surgical removal of the affected area, in some cases accompanied by bone grafting.[15] The first documented case of COF in the ethmoid sinus was in 1977, necessitating a radical maxillectomy to control the tumor while preserving the orbital contents.[1] Due to its resistance to radiation, radiotherapy poses a challenge.[16] Complete excision is crucial to prevent recurrence, as central COF s typically "shell out" effortlessly during surgery.[17,18]

Endoscopic resection of sinonasal COF is a highly effective therapeutic option when performed by an expert surgeon, as successfully demonstrated in this case. Advantages of this technique include direct visualization, magnification, absence of external deformity, and reduced morbidity. A potential complication is skull base injury with resultant cerebrospinal fluid leakage, which can be repaired endoscopically in the same session.[19]

In this study, a minimally invasive technique i: e ESS, was employed to excise the lesion involving the paranasal region, thereby preventing facial scars and other related injuries. A similar case reported the complete endoscopic resection of an 18 mm ossifying fibroma without complications or recurrence after a five-year follow-up.[20]

CONCLUSION

With the lesion's progressive nature and extensive involvement in the paranasal sinuses, effective management remains crucial. Therefore, meticulous surgical planning and expertise, supported by thorough investigations and careful interpretation of radiographs, are essential for managing FOL effectively.

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