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BENIGN SOLITARY AMELOBLASTIC FIBRODONTOMA IN PEDIATRIC PATIENT: NAVIGATING DIAGNOSTIC AMBIGUITIES AND SURGICAL COMPLEXITIES—A CASE REPORT

Maxillofacial Surgery

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KEYWORDS

INTRODUCTION:

Ameloblastic fibro-odontoma (AFO) is categorised as a rare mixed benign odontogenic tumour, the term was used by hooker in 1987 for the first time.[1] Originally it was called as ameloblastic odontome.[2] It is defined as a neoplasm composed of proliferating odontogenic epithelium embedded in acellular ectomesenchymal tissue that resembles dental papilla with varying degree at inductive change and dental hard tissue formation. Since it encompasses two types of odontogenic tumors that share a different histology and biologic behaviour, WHO suggested the term to be inappropriate.

Ameloblastic fibro-dentinoma (AFD) accounts for 2% of all odontogenic tumors. It occurs most frequently in younger age group in 1st two decayed.[1] AFO is very similar to ameloblastic fibroma (AF) and ameloblastic fibrodentinoma (AFD). Clinically it is slow growing, locally non-aggressive, asymptomatic and usually associated with unerupted tooth, additionally it can also cause cortical perforation.[2] There is controversy between ameloblastic fibrodontoma and ameloblastic fibro-odontoma and earlier it was thought to be between ameloblastic fibroma and odontoma. In WHO 2005 classification stated it as developing odontomas (hamartomatous process).[2,3]

According to literature, since ameloblastic fibro-odontoma is non-aggressive lesion, smaller lesions can be treated adequately with simple enucleation and curettage whereas, there is no agreement stated for extensive lesion.[4] The purpose of this paper is to discuss and to analyse the clinical radiological and treatment features of the disease in order to update and improve our knowledge and diagnostic ability of this entity.

Case 1

A 4 year old female child reported to the department of oral and maxillofacial surgery at government dental college and hospital Mumbai brought by her parents, to evaluate an indolent swelling of right cheek region of 3 months duration. The medical history and family history was insignificant.

Figure 1: Extraoral presentation of facial swelling.

Extraoral examination revealed mild facial asymmetry with diffuse asymptomatic swelling on right side of cheek region [Figure 1]. Swelling was non-tender and firm-hard on palpation with no signs of inflammation. On intraoral examination showed a bony hard bulge in the right maxilla extending from distal of canine posteriorly. The swelling was obliterating maxillary buccal vestibule and associated with mobility of maxillary molars in first quadrant [Figure 2a and 2b]. There was no history of local trauma or infection. Aspiration of the lesion was negative.

Figure 2a And 2b: Intraoral presentation of swelling obliterating the buccal vestibule.

Owing to the slow-growing nature, well-circumscribed appearance, presence of calcific structures and its presence in the tooth bearing region, a provisional clinical diagnosis of benign bony odontogenic tumour, was made. Odontogenic myxoma, developing odontoma were considered in clinical differential diagnosis.

The initial panoramic radiography revealed a well circumscribed radiolucent region, which contained a radiopaque mass in irregular size and shape. The case presented in this report had a radiolucent area with no radiopaque structures, making it difficult to differentiate it from that of other odontogenic lesions. The radiolucent area showed a tooth like radiopaque mass in it suggestive of developing tooth bud of permanent first molar [Figure 3].

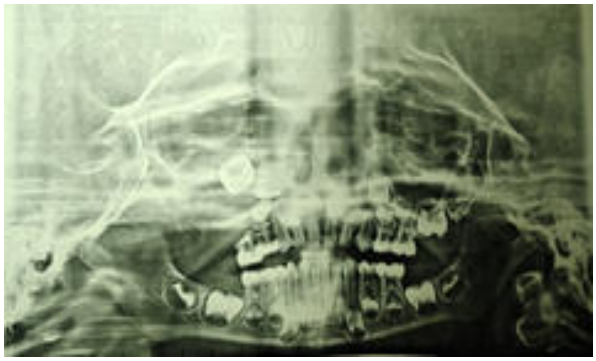

Figure 3: Orthopantomogram showing radiolucent lesion with radio-opaque mass.

Further CT scan was done which revealed a unilocular well-delineated circular radiolucent lesion with smooth borders was present. Cortical expansion, on the right side of the posterior maxilla was noted, extending from deciduous canine posteriorly and superior-inferiorly from alvelolar crest till infraorbital border. The lesion was associated with impacted permanent first molar on right side.

Considering the patient's age, the indolent growth pattern of the lesion, and the desire to minimize repeated exposure to general anaesthesia, a definitive excisional biopsy under general anaesthesia was deemed the most appropriate approach.

Figure 4, 5, 6: surgical excision of the lesion.

The surgical therapy based on enucleation and curettage with healthy tissue margins was carried out. After divulging and displacing the facial tissues, the surgical access revealed glistening well encapsulated lesion [Figure 4]. During the surgical procedure, we detected that the lesion was easily detached and separated from the adjacent bone, Therefore the lesion was removed in toto [Figure 5, 6]. The mobile primary molars along with the developing bud of permanent molar embedded with the lesion was removed. The flap from cheek and mucosa was made to close the defect. Intraoral closure was done with resorbable suture. No specific technique was performed to promote the bone regeneration. The surgical specimen measuring 45×43×32 mm was sent for histopathological evaluation. The immediate postoperative period proceeded without any complications and the patient was discharged 5 days after the surgery ensuring satisfactory healing and recovery.

Given the limited extent of the lesion and the absence of features suggestive of aggressive behavior, a conservative treatment approach was chosen, a decision that ultimately proved to be appropriate.

The microscopic findings revealed the lesion was composed of pieces of loose cellular connective tissue reminiscent of dental papilla with cords, nests and islands of cuboidal to columnar odontogenic epithelial cells. Cystic and microcystic degeneration in the nests and islands of the epithelial cells was observed. The overall findings were favouring the diagnosis of ameloblastic fibro-odontoma.

The case is under follow-up, in order to minimize the chances of recurrence and provide early intervention in case of recurrence [Figure 7].

Figure 7: Follow up after 2 years.

DISCUSSION:

The AFO belongs group of lesion referred to as mixed odontogenic tumors, according to literature these includes Ameloblastic Fibroma, AFO, Odontoma and Odontoameloblastoma.[6] The lesion is defined as neoplasms composed of proliferating odontogenic epithelium embedded in cellular ectomesenchymal tissue that resembles dental papilla, with varying degrees of inductive changes and dental hard tissue formation.[5]

The dispute is whether these lesions, often grouped as Ameloblastic Fibroma and AFO, are in fact neoplasms, or an odontoma in different development statges.[2] Slootweg et al believed that ameloblastic fibroma and fibrodontoma are various stages of the same lesion and over time it matures to form odontoma. However, Ameloblastic fibro-odontoma is reconsidered as a distinct entity after the latest WHO classification.[5] Although the presence of BRAF and V600E mutations in AFD and AFO is similar to AF but absent in odontome, supported the arguments that at least some of these lesions are in fact neoplastic, particularly those with locally aggressive biological behaviour, large size, and recurrence.[1]

Histologically it represents combination of ameloblastic fibroma and odontoma. Ameloblastic fibro-odontoma is relatively rare slow growing, asymptomatic locally non-aggressive lesion. Etiology of which is unclear yet.[2] The tumour usually occurs in young patients with average age of 3.3 years, with no gender and site predilection in particular.[2,3]

The commonest presenting complaint is usually painless swelling with unerrupted teeth.[4] Sometime the lesion is accidentally diagnosed upon radiographic examination during evaluation of unerrpted tooth.

Microscopically, the lesion is composed of strands, cords and islands of odontogenic epithelium embedded in a cell-rich, primitive ectomesenchyme resembling the dental papilla with variable amounts of irregular formation of enamel, dentin and a cementum-like material, as was seen in this case. Surprisingly in this case, cystic and microcystic degeneration was seen in nests and islands of odontogenic epithelium, this being in fact a common feature of ameloblastoma.[5-7,10, 12-13.]

Literature suggests of conservative surgical excision as a treatment for this lesion. In most cases, the impacted tooth associated with the tumour is removed at the same time. There is a low potential for recurrence. Despite the low potential recurrence of the tumour, a 5 year-long follow-up is recommended.[5-12] Fredrich et al stated that the recurrence is associated with inadequate surgical removal, and may occur if tumour remnants persist in the resection margins and in the tooth involved, especially for large tumours.[13] It is recommended to extract the involved tooth to avoid reincidence which was followed in our case.

CONCLUSION:

Ameloblastic fibro-odontomas appear as relatively rare and benign-like lesions; however, they do require special attention in order not to become larger lesions. Like most authors, we recommend enucleation of AFO as the treatment of choice, followed by a long preservation.

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