CASE REPORT

Ameloblastic Fibrodentinitoma: A Rarity in Odontogenic Tumors

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Abstract Ameloblastic fibrodentinitoma is a rare, benign, mixed odontogenic tumor composed of odontogenic epithelium, immature connective tissue and is characterized by the formation of dysplastic dentin. An incidence of 0.1% is reported for this benign odontogenic tumor which indicates the rarity of this lesion. In the present review, a case of ameloblastic fibrodentinitoma of the mandible in an african male is presented which was managed by surgical enucleation of the lesion along with the removal of impacted tooth associated with it.

Keywords Ameloblastic fibrodentinitoma · Mixed odontogenic tumor · Dysplastic dentin

Introduction

Ameloblastic fibrodentinitoma (AFD) is a rare, benign, mixed odontogenic tumor. It is also called as Dentinoma or Fibroameloblastic dentinoma. It is composed of

odontogenic epithelium, immature connective tissue and is characterized by the formation of dysplastic dentin. Recent review on 1088 central odontogenic tumors states that the relative frequency of Ameloblastic Fibroma and Ameloblastic Fibro-odontoma together constitutes 1.7%, which indicates the rarity of the lesion Ameloblastic fibrodentinitoma [1]. The first case of Ameloblastic Fibro dentinitoma was reported by Straith in 1936 [2].

Case Report

A 20-year-old African gentleman presented to the department of oral and maxillofacial surgery with chief complaint of swelling in the left front region of the lower jaw since 2 years (Fig. 1). Three months back, the patient noticed that the swelling which was soft initially was becoming firm. There was no history of any associated pain, fever, numbness of lower lip or any discharge from the swelling.

On clinical examination, the patient had an obvious facial asymmetry. The swelling was firm-hard extending from left body to right parasymphysis region of mandible, (approx. 8 cm × 5 cm in size). The swelling was non-compressible, non-fluctuant, non-tender, with no localized rise in temperature and with a normal color of overlying skin. A firm, mobile and non-tender left submandibular lymph node was palpable (approx. 2 × 1 cm in size). Intraorally it was found to be extending from #42 to 36 region with vestibular obliteration. There was a retained deciduous #74, missing #34, Grade 1 mobile #35 and Grade 2 mobile #74. There was no intraoral pus discharge.

An ortho-pantomogram done revealed unilocular radiolucent lesion extending from region of teeth #36–#43 and an impacted premolar #34 at the inferior border of the mandible. There was root resorption of the teeth involved

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Fig. 1 Preoperative frontal view showing an assymetry of face with a swelling in the left anterior region of lower jaw

in the lesion (#36–#42) (Fig. 2). An aspiration test done was negative for the lesion. Vitality test done for teeth in the lower jaw revealed non-vital tooth w.r.t #32, 74, 36. Dentascan (Fig. 3) showed radiolucent unilocular lesion extending from #43 to #36. Buccal cortical perforation was noticed from #33 to #37 region; however, lingual cortex was intact.

Incisional biopsy from the left lower vestibular region was performed, and it was found that the cavity was filled

Fig. 2 Ortho-pantomogram revealing unilocular radiolucent lesion associated with an impacted premolar in the left body region of mandible

Fig. 4 Incisional biopsy revealing the soft consistency mass

with soft consistency mass (Fig. 4). Based on the clinical examination and preliminary investigation differential diagnosis of ameloblastoma, chondrosarcoma was made.

Histopathological examination revealed strands, cords and islands of proliferating odontogenic epithelium associated with ectomesenchymal cell associated with ectomesenchymal cells (Fig. 5a). The strands revealed double layer of cuboidal cells, resembling dental lamina. The Islands are comprised of tall columnar ameloblast-like cells at peripheral and polyhedral stellate reticulum like cells in the center (Fig. 5b). Areas of dentinoid and osteodentin formation preceded by a zone of hyalinization (Fig. 5c). These features were suggestive of Ameloblastic fibrodentinoma. Based on histopathology enucleation of the lesion was planned under general anesthesia.

Vestibular incision was given extending from #44 to #37 region; periosteal flap was raised exposing the lesion. The buccal cortical plate was found to be perforated. A bony window was created and the content of the cystic lesion was enucleated along with the removal of impacted tooth 34 and extraction of #35, 36 (Fig. 6). Whitehead varnish dressing was given, and surgical wound closure was done

Fig. 3 Dentascan images showing the extent of the leison

Fig. 5 a–c Photomicrograph

Fig. 6 Intraoperative images showing the buccal cortical perforation, cystic cavity postenucleation and the enucleated mass

using 3–0 silk sutures. Whitehead varnish is a combination of iodoform, benzoin, balsam of tolu and storax. Its beneficial effect on tissues of wound is due to its protecting power both mechanically and in maintaining asepsis, thus promoting faster healing by secondary intention.

At 1-week follow-up whitehead varnish pack was changed via a secondary vestibular incision and root canal treatment was initiated for the remaining teeth involved in the lesion. The patient has been kept on regular follow-up for whitehead varnish pack change for approximately

1 year. Healing was satisfactory and uneventful as seen clinically and in subsequent radiograph (Figs. 7 and 8).

Discussion

World Health Organization publication on histological typing of odontogenic tumors defined Ameloblastic Fibrodentinoma lesion as “A very rare neoplasm composed of odontogenic epithelium and an immature odontogenic

Fig. 7 Postoperative frontal view

connective tissue, and characterized by the formation of dysplastic dentin” [3]. It is believed to be an intermediate stage between ameloblastic fibroma and ameloblastic fibro-odontoma. An incidence of 0.1% is reported for this odontogenic benign tumor, and 49 cases have thus far been reported following a search in the literature after 1936.

AFD is a controversial tumor considering its biological nature and histological diagnosis. It has been suggested that AFD is not a true neoplasm but a hamartoma. The limited growth potential of this tumor may support this suggestion. On the contrary, it has also been reported that AFD may have a significant growth potential and may enlarge to a considerable size supporting the statement that this lesion is a true neoplasm [4].

The Ameloblastic fibro-dentinoma is usually encountered in the first 2 decades of life, with a male to female predilection ratio of 2.4–3:1. The most commonly involved site is mandible than maxilla (mandible: maxilla 3:1), with the molar region being most commonly involved. The lesion is commonly asymptomatic and is discovered when radiograph is taken to determine the reason for failure of a tooth to erupt. Radiologically, it shows unilocular or multilocular radiolucency with or without radio-opaque areas.

The exact histogenesis of Ameloblastic fibro-dentinoma is unclear. Eversole et al. propose that mixed odontogenic tumors are totally dependent upon the presence of differentiation factors which are elaborated by a particular tumor [5]. Hence, there is little probability of sequential differentiating events resulting in the progression of an immature entity (ameloblastic fibroma) to a highly differentiated entity (complex odontoma). Trodahl and Gardner believe that odontomas pass through a developmental stage and that a non-calcified stage of development occurs [6]. This stage would mimic closely the histologic appearance recognized as ameloblastic fibroma. Thus, there exist two lesions that have the histologic appearance of ameloblastic fibroma—the actual ameloblastic fibroma and one stage of a developing odontoma. Gardner goes on to state that there is no evidence that the ameloblastic fibromas with dentin formation exhibit a different biologic behavior than classic ameloblastic fibromas and hence designation of AFD as a separate entity is not justified [7]. Gardner further states that these lesions should be referred to as ameloblastic fibromas with a qualifying descriptive phrase noting the presence of dentin added by the pathologist.

Fig. 8 Postoperative radiograph

McKelvy and Cherrick in support of Eversole's concept of the histogenesis of mixed tumors comments that normally amelogenesis is initiated soon after odontoblasts lay down the dentin matrix [8]. Taking into account the quantity of dentinoid found in the reported cases, it is unusual that enamel matrix has not been noted.

Philipsen et al. suggested that ameloblastic fibroma and AFD manifest in two forms. The first is a neoplastic lesion which if left in situ will not mature further. The second variant is a hamartomatous lesion that appears to have the potential to differentiate into an ameloblastic fibro-odontoma and mature further into a complex odontoma. This idea has been incorporated into the suggested modifications for the 1992 edition of the WHO histological typing of odontogenic tumors [9]. Most of the studies showed that AFD occurred in two forms mature and immature dentinoma. Immature form is thought to be an AF in which induction of mesenchymal cells by odontogenic epithelium has resulted in the formation of organic matrix of dentin or osteodentin and in mature type all the elements are similar, but with scanty or absence of an odontogenic epithelium. According to Takeda, immature dentinoma is different from AFD as both epithelial and fibrous elements may resemble those of odontogenic fibroma rather than that of AF with or without dentin formation [10].

Since the lesion is benign, the recurrence rate is very low. Occasionally ameloblastic fibrodentinosa, a very rare malignant odontogenic neoplasm, is thought to arise from the malignant transformation of the ectomesenchymal component of AFD [11]. The 2005 WHO classification of odontogenic sarcomas presented two tumors: Ameloblastic fibrosarcoma and ameloblastic fibrodentinosa. In the malignant counterpart, only the mesenchymal component undergoes a malignant transformation while the epithelial component does not show any malignant changes.

Most often Ameloblastic fibrodentinoma has a distinctive fibrous connective tissue capsule. Hence conservative surgery like simple enucleation remains treatment of choice. If the lesion is aggressive and presents with erosion of cortical plate, a radical resection should be performed [12]. In the present case, the tumor was enucleated in toto along with impacted 34 through an intraoral approach

under general anesthesia and no sign of recurrence was observed during 1 year of follow-up period.

Conclusion

Although Ameloblastic fibrodentinoma is a benign, rare and non-recurrent tumor, delayed eruption, missing or impaction of permanent teeth in pediatric patient, emphasizes the importance of early radiographic examination of these conditions for early detection, diagnosis and treatment.

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