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Case Report

Ameloblastic Fibro-Odontoma with Ameloblastin Immunohistochemistry

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Abstract

Ameloblastic fibro-odontoma is a subset of pediatric mixed odontogenic tumor that morphologically is related to ameloblastic fibroma and odontoma. We report a case of ameloblastic fibro-odontoma in a 3-year old boy associated with a non-specific cyst lining using ameloblastin immunohistochemistry and light microscopic comparison with normal human tooth germ.

Keywords: Ameloblastic fibroma; Odontoma; Ameloblastin; Tooth germ; Mixed odontogenic

Abbreviations

AFO: Ameloblastic Fibroma; AF: Ameloblastic Fibroma; AFD: Ameloblastic Fibrodentinoma; COC: Calcifying Odontogenic Cyst; Oe Outer Enamel Epithelium; Sr: Stellate Reticulum; SI: Stratum Intermedium; Iee: Inner Enamel Epithelium; Pa: Preameloblast; Tpsa: Transitional Presecretory Ameloblasts; Psa: Presecretory Ameloblast; Sa: Secretory Ameloblast; Cf: Cell-Free Zone; Df: Dental Follicle; Dp: Dental Papilla; Od: Odontoblast Dentin; D; Predentin; Pd; Enamel; E

Case Presentation

A 3-year old boy presented for the evaluation of a missing first right deciduous molar and associated painless swelling over the right side of the mandible of 3-months duration without any other significant findings or contributory history. Panoramic radiograph disclosed a well-defined radiolucent lesion with central radiopaque mass (Figure 1). The differential diagnosis included mixed odontogenic tumors. An excisional biopsy was planned and the lesion was removed along with the deciduous second mandibular molar and permanent first mandibular molar. The surgical specimen consisted of multiple fragments of soft and hard tissue (Figure 2). Both soft and hard tissue samples were processed for histopathological examination. On microscopy, the soft tissue specimen showed presence of epithelial and (ecto) mesenchymal components (Figure 3). On low to high power magnification, the epithelial follicles or islands were arranged in different configurations in relation to the (ecto) mesenchyme that was reminiscent of differentiation during normal odontogenesis (Figure 4). The decalcified hard tissue specimen was characterized by the presence of epithelial and (ecto) mesenchymal components with the formation of dentin, enamel and pulp tissues (Figure 5A-D). The histomorphological features are consistent with ameloblastic fibro-odontoma. Although not required for the diagnosis, immunohistochemistry with ameloblastin was performed in the decalcified section as described elsewhere [1]. Immunohistochemically, enamel matrix showed intense reaction to ameloblastin, and the central cells and peripheral columnar cells with morphology of secretory ameloblasts showed nuclear staining. The dentin matrix was negative to ameloblastin while the pulpal cells were positive with nuclear staining (Figure 5A1-D1). For comparison with normal odontogenesis archival human tooth germ routine sections from previous studies were used [1,2], as shown in (Figure 6).

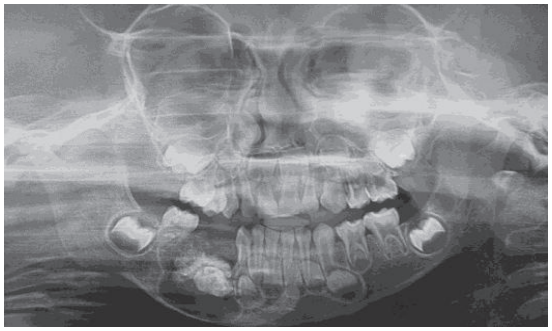

Figure 1: Panoramic radiograph shows a well-defined radiolucent lesion with central radiopaque structures of varying densities in the region of missing deciduous right first molar. The lesion extends from the lower border to the alveolar region and between mesial of right permanent first molar follicle to the distal of right permanent canine follicle. Resorption of right deciduous second molar is evident.

Discussion

Ameloblastic Fibro-Odontoma (AFO) presents as a slow growing, painless tumor, which is most commonly found in children of 1-6 years age and usually before 13-years of age, that is frequently detected by routine roentgenogram. It may occur in any part of the jaw but frequently found in the premolar and molar regions of the mandible [3]. Radiographically, AFO appear as an expansile cystic radiolucency containing small bodies of a radiopaque material and occasionally a larger, centrally located mass of hard tissue [3]. The histological appearance of Ameloblastic Fibroma (AF), Ameloblastic

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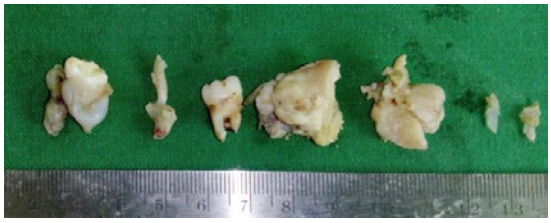

Figure 2: Formalin fixed surgical specimen shows root resorption in the right deciduous second molar, follicle attachment in the right permanent first molar with malformed crown, fragments of soft tissue and large specimen corresponding to the radiopaque structure seen in panoramic radiograph.

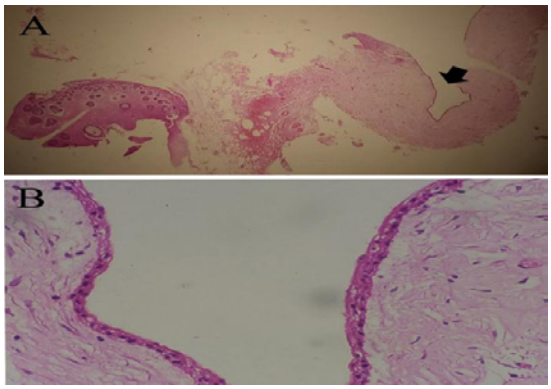

Figure 3: A: Scanner view of the soft tissue specimen shows epithelial and (ecto) mesenchymal components and a cyst epithelial lining (arrow head). **B:** Higher magnification of the epithelial lining appears reduced enamel epithelium-like as normally seen in dentigerous cyst. H & E, x20, x400.

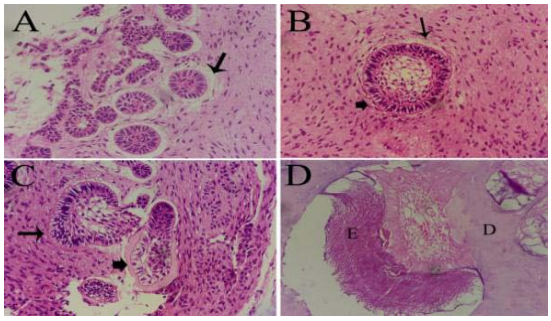

Figure 4: A-C are higher magnification of Figure-3A that shows different configurations of epithelial and mesenchymal components corresponding to normal odontogenesis as shown in (Figure 6). H & E, x400.

Fibrodentinoma (AFD) and AFO are more or less similar and characterized by epithelial and mesenchymal components [4]. The latter two tumors are defined by the formation of either dentin or enamel, dentin, pulp and cementum while the former is considered as the most primitive form with no accompanied tooth matrix materials. AFO microscopically exhibit soft epithelial and hard mesenchymal components. The soft epithelial component simulates AF with strands, buds, islands or follicles [3,5]. The strands and buds are only few cells thick that resembles dental lamina [3]. Whereas the islands or follicles demonstrate an inner stellate reticulum – like cells surrounded by an outer layer of columnar cells with variable differentiation [3]. The epithelial strand or island/follicle rests upon

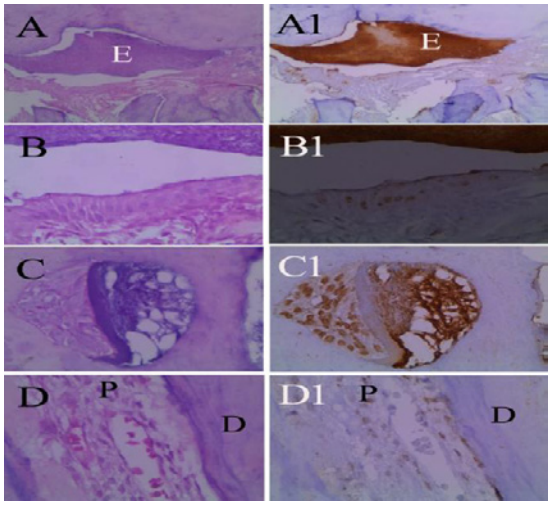

Figure 5: A-D & A1-A4 are serial routine and ameloblastin stained sections. A & A1 shows positive reaction in the secretory ameloblasts (nuclear) and enamel matrix (E). B & B1 are corresponding higher magnification of A & A1. C & C1 shows positive reaction in the enamel matrix and reduced enamel epithelium with negative reaction in dentin/osteodentin. D & D1 shows positive reaction in the nuclei of pulpal cells (P) but negative reaction in the dentin/osteodentin (D). x100, 400.

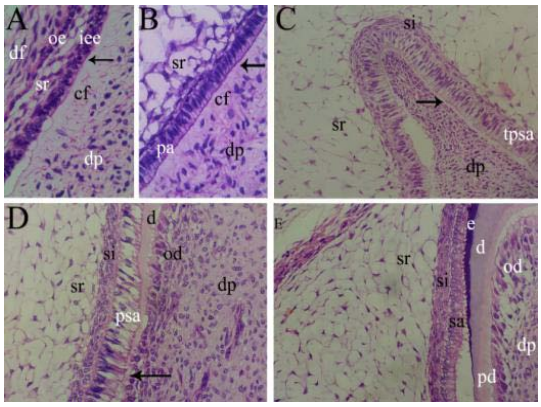

Figure 6: A-E are illustrative fields of epithelial and ectomsenchymal (dental papilla) interface during late (A, B, D & E) and early bell (C) stages of odontogenesis.

A: Shows inner enamel epithelium with cuboidal to short columnar shaped and nuclei placed centrally or toward dental papilla with prominent cell-free zone.

B: Shows preameloblasts with tall columnar shaped with palisading and polarization away from the basement membrane (arrow), and relatively narrow cell-free zone with the peripheral cells of dental papilla begins to show condensation.

C: Shows transitional presecretory ameloblasts with tall columnar shaped with the nuclei arranged in a pseudostratified pattern with mostly obliterated cell-free zone and the peripheral cells of dental papilla shows visible condensation.

D: Shows presecretory ameloblasts with slender, high, tall columnar in shape with the nuclei showing palisading and polarization away from the basement membrane which appear disrupted and obliterated cell-free zone (arrow and compare it with arrows in A-C).

E: Shows secretory ameloblasts with tall columnar in shape, cytoplasm is granular to clear, and the nuclei shows palisading and polarization from the enamel/dentin, predentin and odontoblast complex. H & E, x400.

a thin delicate basement membrane surrounded by either pauci-or cellular stroma but may exhibit a cellular zone at one and a cell-free

zone at the other [5]. In addition, a narrow band of connective tissue hyalinization may rim the epithelial component [4,5]. The hyalinized material have been variedly interpreted such as basal lamina, dentin, atubular dentin and dentinoid [6,7]. The presence of this material is believed to negatively impact reciprocal epithelial – mesenchymal interactions for the elaboration of differentiated products in pathological situations [6]. Apart from the soft ameloblastic fibroma component, AFO exhibits a hard tissue odontoma like component with enamel, dentin/dentinoid/atubular/osteodentin, bone and pulp [4,6]. Although it is believed that odontoma may mimic AF and/or AFO during their natural developmental history [6-8], there are no histological criteria to distinguish with certainty [4,6-8]. However, unlike odontoma, AFO exhibits preponderance of dental papilla-like tissue [6]. Nevertheless, in routine practice, it is prudent to rely on clinical and radiological findings such as age, size and location of the lesion in association with unerupted tooth and bone destruction [7].

The mixed odontogenic tumor such as AF, AFD, AFO and odontoma has the potential to demonstrate epithelial and ectomesenchymal interactions similar to tooth germs during odontogenesis [3,5,6,9-11], which can be visualized on light microscopy as amorphous eosinophilic or hyaline bands around the epithelial follicles or islands [3,5,7,9].

The light microscopic findings noted in the present report is, however, than in accordance with the previous observation in AFO or immature complex odontoma [10,11]. Therefore, it is pertinent to draw attention to the context of light and/or electron microscopic observations by previous studies in relation to the relevant findings in normal human tooth germs [1,2,9-11]. According to previous studies [10,11], AFO mirror tooth development up to the level of bud to cap stage of odontogenesis but does not differentiate further due to an arrest in histodifferentiation as result of an incapacity of the epithelial cells to exert an organizing influence over the ectomesenchymal cells [10]. In contrast, the arrest in histodifferentiation in immature complex odontoma has been attributed to failure of ectomesenchymal cells to respond to the normal epithelial cues [11]. This implies that although there is defective histodifferentiaion in immature complex odontoma, the epithelium (preameloblasts) is still capable of further differentiation into a more advanced stage than the ectomesenchyme, even without an apparent physical contact up to the stage (secretory ameloblast) of enamel deposition [11]. Therefore, the differences in cytodifferentiation between cells of AFO and immature complex odontoma overtly conveys differences in their genesis [10,11]. However, regardless of whether they pursue different differentiation pathways, the differentiation of ectomesenchyme is restricted to the development of fibroblast-like cells that do not acquire the morphology of odontoblasts [10,11]. Therefore, the formed dentin matrix is defective which explain the lack of tubular dentin in these entities [10,11]. By immunohistochemistry, the literature reveals expression of terminal differentiation specific markers of ameloblast and odontoblasts such as amelogenin and osteocalcin (common phenotypic marker for osteoblasts, odontoblasts and cementoblasts) in the epithelial zones (keratin positive peripheral columnar cells and central stellate reticulum or stratum intermedium like cells) of AFO and complex odontoma, which not only indicates the presence of mixed epithelial cells that have phenotypic features of both epithelial and mesenchymal cells but also suggests that the tumor cells may have reached an advanced stage of maturity than they correspond to the cap and/or early bell stage [12]. In light of the foregoing observations [2,10,11], the routine section and immunohistochemical findings of the present case illustrates that at least in some microscopic fields the epithelial and ectomesenchymal cells do come in contact as normally occurs in human tooth germ during odontogenesis [2]. The positive reaction of ameloblastin in both the enamel matrix and the peripheral tall columnar cells of epithelial nests/follicles lends further support to our light microscopic observations. However, unlike human tooth germs, ameloblastin stained peripheral columnar and central cells of the epithelial islands and as well as pulpal cells with a nuclear pattern as against our previous observation of cytoplasmic staining restricted to the peripheral columnar cells in human tooth germs [1]. The shift in the specific cytoplasmic staining pattern of the peripheral columnar cells (presecretory and secretory ameloblasts) in human tooth germ to non-specific nuclear staining of both epithelial and mesenchymal cells is likely to be related to the acquisition of tumorigenic process.

The literature reveals documented cases of calcifying odontogenic cyst (COC) or dentigerous cyst with the presence of ameloblastic fibroma component in the wall [13,14]. The literature also reveals rare reports of ameloblastic fibro-odontoma in the wall of COC [15-17]. In the present case, the apparent continuity of AFO from a cyst lining that lacked features suggestive of COC would well exclude the latter as the primary lesion. However, the presence of reduced enamel epithelium like lining would suggest dentigerous cyst, but there is no radiological or gross microscopic context to support it. Therefore, the AFO in the present case would have either evolved from an initial non-specific cystic process or from reduced enamel epithelium. In conclusion, we report a rare case of AFO associated with a non-specific cyst lining with reference to human tooth germ using ameloblastin immunohistochemistry.

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