



Simultaneous Occurrence of Adenomatoid Odontogenic Tumor and Dentigerous Cyst- A Rare Case Report

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Abstract

Adenomatoid odontogenic tumor (AOT) is a rare odontogenic tumor with limited growth and usually associated with an unerupted/impacted permanent tooth. It is a benign lesion, with a low rate of recurrence after surgical treatment. Dentigerous cyst is an odontogenic cyst with epithelial lining that may transform into an odontogenic neoplasm- like ameloblastoma. Very few cases of Adenomatoid odontogenic tumor (AOT) developing as a complication in Dentigerous cyst have been reported in literature and it has been termed as Hybrid variant of AOT. This article contributes to literature with a rare case presentation of AOT associated with Dentigerous cyst

Keywords: Hybrid Variant; Adenomatoid Odontogenic Tumor; Dentigerous Cyst

Introduction

The adenomatoid odontogenic tumor (AOT) is a rare tumor generally found in young females (f: m = 2.3:1) [1]. It is commonly seen in maxilla (maxilla: mandible = 2.6:1) [2]. It is generally seen in the second decade of life. Dentigerous cysts are the most common type of developmental odontogenic cysts arising from the crowns of impacted, embedded, or unerupted teeth constituting the second most common cystic lesion of the jaws, after radicular cysts. They are commonly seen in the second and third decades of life and show a male predilection.³ With patient's consent, management of a case of simultaneous occurrence of these two lesions in a 19-year-old female is described in this article.

Demographic details and Clinical Findings

An 19-year-old female patient reported to Advanced surgical dental health care setup with the complaint of swelling on right upper jaw since 6-8 months that gradually increased to present size. The medical history was unremarkable. On extraoral examination, there was a diffuse swelling seen on right side of face on upper jaw.

Intraorally, patient had overretained deciduous teeth in 1st quadrant with a firm, tender swelling present in upper buccal vestibule with expansion of palatal bone, extending from 13 to 15 regions obliterating the vestibular depth.

Radiographic investigations (Figure 1-4)

In OPG (orthopantomogram) mixed areas of radiopacities and radiolucency around crown of canine (maxillary right side)13 was observed. In CBCT (cone beam CT assessment) mixed dentition was reported and in maxillary arch lesion observed was approximately 35 x 40 mm in size extending mesially from lateral wall of nose to lateral zygomatic process of maxilla. Superiorly lesion extended from floor of orbit to inferior right maxillary alveolar ridge. The shape of lesion reported was roughly oval with smooth and distinct margins. The internal structure is predominantly radiolucent with tiny flecks of radiopacities giving it a mixed radiolucent appearance in 13-14 region, 14 is seen embedded in lesion with 13 impacted giving rises to differential diagnosis of intraosseous variant of odontogenic lesion. The root of 14 was oriented medially

and inwards. The lesion caused expansion of right maxilla causing significant expansion of cortical plates; resulting in thinning of facial cortical bones. Due to lesion perforation of lateral wall of nose and upward displacement of floor of orbit was observed. Maxillary sinus appeared reduced in size and was pushed to zygomatic process of maxilla. The lesion has resulted in impaction of 14. 14 was horizontally impacted as evident in figure with root oriented medially and inwards, crown facing to palatal aspect of 54 causing resorption with 54. Lesion has also resulted in resorption of 15,16,17.



Figure 1: CBCT (cone beam CT assessment) mixed dentition was reported and in maxillary arch lesion observed was approximately 35 x 40 mm in size.



Figure 2: CBCT (cone beam CT assessment) mixed dentition was reported and in maxillary arch lesion observed was approximately 35 x 40 mm in size.

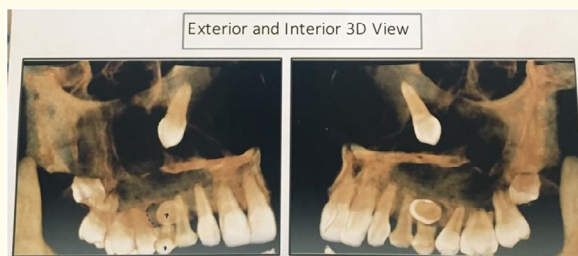


Figure 3: CBCT both exterior and interior 3 D view.



Figure 3: CBCT both exterior and interior 3 D view.

Surgical Notes (Figure 5 and 6)

Correlating clinical and radiographic findings, a diagnosis of aggressive odontogenic tumor associated with impacted 13 and supernumerary 14 was made.

Patient was operated under General anaesthesia with nasal Endotracheal Intubation. Patient was prepped and draped in usual surgical fashion. After local infiltration in the right maxillary vestibule, right vestibular incision was taken to expose the anterolateral wall of right maxilla. Care was taken to identify right infraorbital nerve and preserved under the retractor and gauze piece. Bony window was created with the inferior cut horizontal to the occlusal plate and at least 5mm above the root apex of premolar teeth. Maxillary sinus was found to be obliterated with cystic lining and the canine was found to be impacted with its root pointing toward the lateral wall of nose approaching the supero-medial wall. The impacted canine with the cystic lining was carefully dissected out and removed in toto. Another hard tissue tumor like tissue was found to be filling the sinus and more towards the floor of sinus causing expansion of palatal bone with impacted supernumerary teeth. The tumor was dissected free from the palatal mucosa and removed in one piece and submitted for histopathological examination. Closure of the maxillary sinus was done after open packing with betadine gauze which was subsequently removed after 48 hrs. Routine medication was given through IV route.



Figure 5: Intraoperative photographs- Before removal of lesion.



Figure 7: Intraoperative photographs- Before removal of lesion.

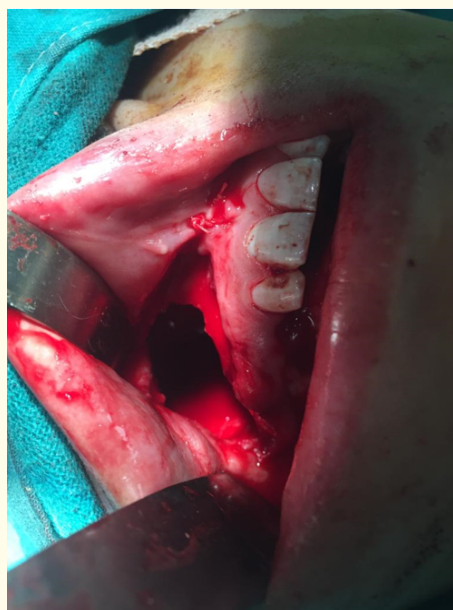


Figure 6: Intraoperative photographs- After removal of lesion.

lular changes were seen . Hence the final diagnosis of Adenomatoid odontogenic tumour with simultaneous occurrence of dentigerous cyst was given. Post-operative healing was eventful, and the patient is being followed up for last 12 months.

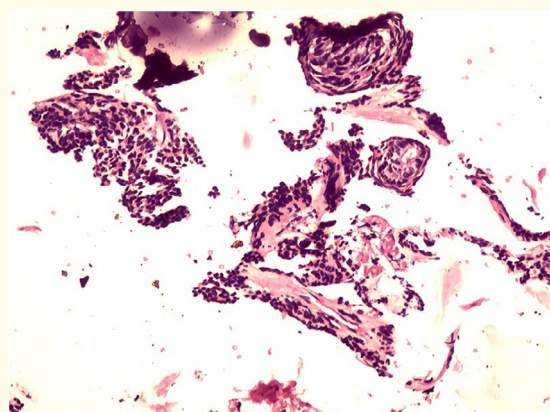


Figure 8: Whorls, duct/microcyst with small lumina (thin arrow) and stands of basaloid cells.

Gross Specimen for microscopy (Figure 7)

Two pieces are received one as an impacted 13 and supernumerary 14 with attached cyst and mass associated with posterior and palatal wall together measuring 4 x 3.5 x 3 cm in size.

Microscopic findings (Figure 8-11)

Multiple sections examined from both specimens show markedly fibrotic cystic wall lined focally thin non keratinized stratified squamous epithelium Resembling reduced enamel epithelium . small fragments of fibrous cyst were lined by basaloid cells with hyaline pink deposits in pseudo glandular spaces. Some areas showed amyloplastic epithelium arranged in whorl and formation of ductular spaces. Focal calcification is observed. No atypical cel-

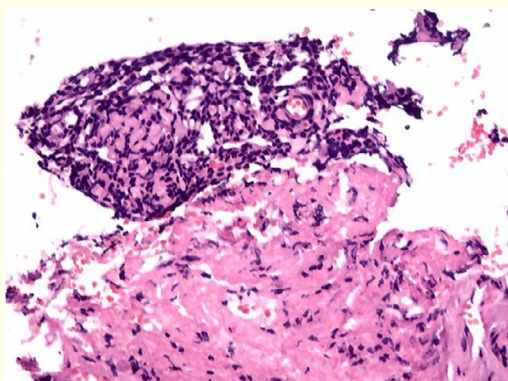


Figure 9: 10x – basaloid cell cluster arranged as cribriform configuration with pink eosinophilic material.

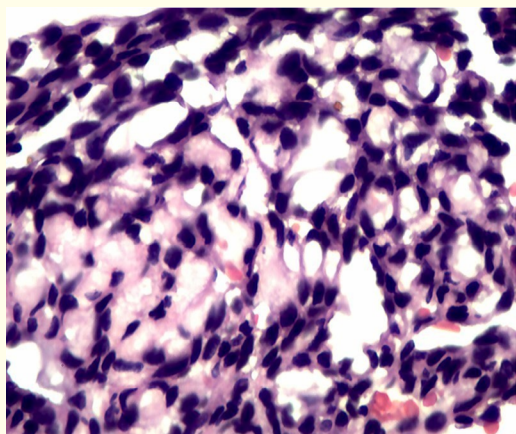


Figure 10: 40 x – reticulum like spindle cells with some round to oval basaloid epithelial cell.

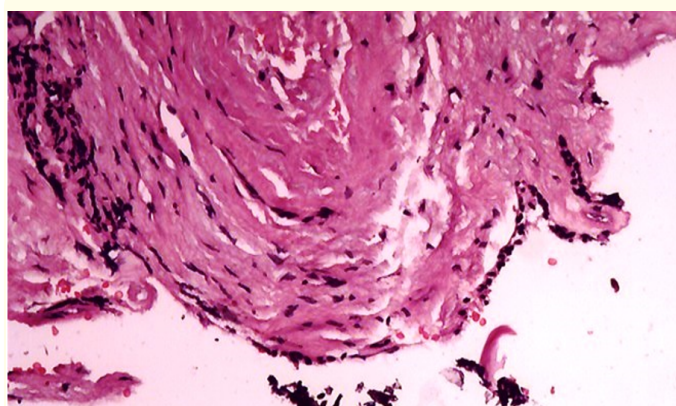


Figure 11: 10x cyst lining.

Discussion

The odontogenic tissue surrounding an impacted tooth is prone to develop into odontogenic cysts and tumors. The most common odontogenic cyst arising from pericoronal tissue is dentigerous cyst (DC). The DC is defined as, “developmental odontogenic cyst associated with the crown of an impacted tooth mostly a permanent tooth.” The most common sites are the mandibular third molar and maxillary canine areas. Rare but secondary development of neoplastic lesions from DC such as adenomatoid odontogenic tumor (AOT), complex odontome (CO), ameloblastoma (AB), mucoepidermoid carcinoma (MEC), and squamous cell carcinoma (SCC) have been documented in various case reports and series of cases [2].

Our case showed mural nodules of AOT developing from the epithelial lining of DC. This case was clinically and radiographically diagnosed as aggressive odontogenic tumor. The true nature of these lesions was only identified after thorough histopathologic evaluation of tissue. The affected patient is a teenaged female with involvement of maxillary impacted premolar. There is an uncertainty whether the lining of an associated cyst represents a true DC, cystic change within an AOT or may represent a distinct entity with aggressive potential. The hypothesis that follicular AOTs arise from the reduced enamel epithelium (REE) that lines the follicles of unerupted teeth is fairly conclusive. They surround the crowns and are attached to the necks of unerupted teeth in a true follicular relationship. It cannot be ruled out that the DC with an impacted tooth developed first followed by development of AOT in the cyst wall or vice versa opening the debate for entertaining new hybrid variant in classification. Odontogenic tumors and odontogenic cysts are two distinct pathologies seen in tooth-bearing areas of the jaws. Association of these two pathologies is rarely noticed [3]. There is reported case of AOT being developed from the fibrous capsule of the dentigerous cyst [4]. AOT and dentigerous cyst are both benign, encapsulated lesions, and conservative surgical enucleation or curettage is the treatment of choice.

Conclusion

The present case appeared to be a usual and uncommon Hybrid variant of AOT that is simultaneous occurrence of AOT and dentigerous cyst around the impacted tooth, but extensive sampling and careful microscopic examination is required which could contribute to the precise diagnosis and appropriate management. Regular follow up is required in these cases to study their behavioral pattern.

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