CASE REPORT

Adenomatoid Odontogenic Tumour Associated with Impacted Canine in Mandible Mimicking Dentigerous Cyst: A Case Report with Literature Review.

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Abstract

Introduction: Odontogenic tumours of the jaw are group of pathologies derived from epithelium, mesenchyme or ectomesenchyme tissue remenants from the development of teeth. Adenomatoid odontogenic tumour (AOT), a tumour composed of odontogenic epithelium, is a rare and uncommon tumour of odontogenic origin accounting only 2.2- 7.1% of all odontogenic tumours as per literature. It was originally known by the rather misleading term Adenoameloblastoma. This pathological condition, also known as the "two third tumour" and the "perfect emulator of dentigerous cyst," continues to confound professionals with its diverse histomorphology and contentions regarding its origin. There has been an ongoing debate with the origin of this tumor with very few reported literature.

Case Presentation: This case report describes a unique instance of a large AOT connected to an impacted canine in mandible that resulted from a possible dentigerous cyst in a 24 years old Indian female patient.

Management: Adenomatoid odontogenic tumors have been removed surgically with little likelihood that this entity will reoccur. In our case, the recovery was uneventful with no indications of recurrence on follow up.

Conclusion: Adenomatoid odontogenic tumours are benign hamartomatous, non-invasive lesions.

Keywords: Adenomatoid odontogenic tumour, dentigerous cyst, impacted mandibular canine, two third tumour, odontogenic tumours of jaw

Introduction

Around 2.5-7.1% of all odontogenic tumours are reportedly adenomatoid odontogenic tumours (AOT), a benign odontogenic tumour of epithelial origin[1]. Dreibaldt initially identified it as pseudo-adenoameloblastoma in 1907[2]. In 1971, the World Health Organisation adopted the term "adenomatoid odontogenic tumour," or AOT, which was initially coined by Philipsen and Bien[3]. It is also known as the "Master of disguise" because it closely resembles both a dentigerous cyst and a "two-third tumour," and it typically affects young people in their second or third decade of life. In two third of cases, it is linked to missing or unerupted teeth[4].) Two-third of these entity are commonly found in anterior regions of maxilla and mandible. Presenting a preference for females throughout the second decade of life and frequently linked to impacted canines, it possesses three distinct clinical variants: follicular type (73%), extrafollicular type (24%) and peripheral adenomatoid odontogenic tumour (3%)[5]. The peripheral AOT is mistaken for gingival fibroma, while the follicular variety is frequently misdiagnosed as a dentigerous cyst. In order to effectively manage AOT, differential diagnosis is essential[6].

This case reports the anatomical and clinical features of an adenomatoid odontogenic tumour that occurred in a 24-year-old female patient's mandible and was linked to an impacted canine. Additionally, the surgical management of this entity is also covered.

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Case Report

A 24-year-old woman reported at our oral and maxillofacial surgery department complaining of pain and edema on her left lower face that had been becoming worse over the course of a month. She gave a history of multiple mobile

teeth and over-retained deciduous tooth in lower left back teeth region. The medical and family history was non-contributory. During an external examination, noticeable asymmetry of the face was observed (Figure 1A). A distinct, oval-shaped swelling was identified in the region of the left mandibular body. This swelling, which obliterated the skin fold over mentum area, stretching mediolaterally from the midpoint of the lower lip to the mandibular premolar-molar and superoinferiorly from the tip of the lower lip to the inferior border of the mandible. Its diameter was approximately 4 centimeters. The skin overlying the swelling appeared normal. The swellling was palpable, firm, and adherent to the underlying bone. There were no signs of enlarged lymph nodes.

An intraoral examination (Figure 1B) confirmed the presence of a smooth, single, confined swelling that was obliterating the left mandibular buccal vestibule region. The swelling was around 4 centimeters by 2 centimeters supero-inferiorly and medio-laterally. Antero-posteriorly, it extended from the mesial border of the lower left first molar tooth antero-posteriorly to the lower right mandibular second incisor. The buccal vestibule was obliterated with the swelling. Grade II mobility was noted with tooth 34, 32, 31 and 41. The lower right first premolar showed a bucco-lingual rotation. Over-retained primary mandibular canine was noted with absence of permanent canine in the arch. The mucosa above it showed a typical hue. The swelling was tender on palpation along-with bony hard consistency and well- defined edges. Buccal cortical expansion was also noted.

On aspiration of the swelling with 18 guage wide bore needle, clear yellow straw coloured fluid was aspirated. Upon examination, the left mandibular deciduous canine was found to be over-retained, the left mandibular permanent canine was absent, and teeth 35, 34, 32, 31, 41, and 42 had grade 2 mobility. It also showed remnants of tooth number 36 in the form of root pieces.

Axial, coronal, and sagittal sections with a resolution of 0.015 millimeters were taken at 1 millimeter interval from the mandible of FOV (5X8) using CBCT. A well-defined, discontinuous, corticated cystic radiolucency was seen in the various radiographic sections. It extended supero-inferiorly from the alveolar crest to the inferior border of the mandible, from the 42 region upto 35 region, and it was attached to the cervical

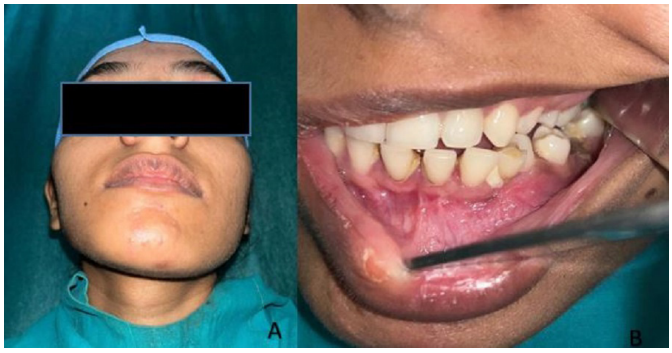

Fig. 1 A: Facial asymmetry with swelling on left lower third face. **B:** Intraoral swelling obliterating the left mandibular buccal vestibule region with missing permanent lower left canine and over-retained deciduous canine.

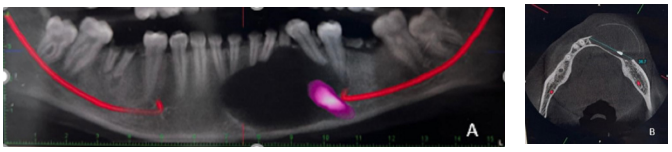

Fig. 2: CBCT image showing a well-defined, corticated cystic unilocular radiolucency associated with impacted canine with well-defined discontinuous margins in mandibular anterior region

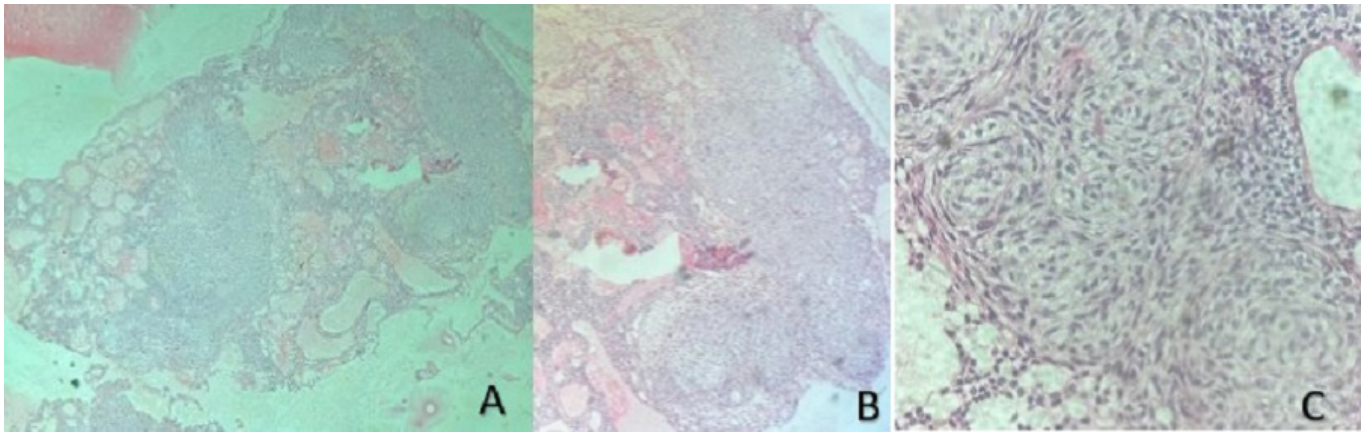

Fig. 3 (A,B): Photomicrograph with Haematoxylin and Eosin (H&E) stain) Low power: 4X and 10X magnification showing Rosette formation and ductal pattern of epithelial cells. **C:** Photomicrograph (H&E stain) High: 40X magnification showing characteristic duct like structures, lined by a single layer of cuboidal and columnar epithelial cells along with rosette formation. Several areas of tumor cells arranged in acinar or glandular pattern (Images from left to right)

TABLE 1:

SR NO.	Reference	Age/ sex	Race	Year	Site	Tooth Number
1.	Valderrama	16/F	Philippino	1988	Maxilla	Unilocular radiolucency, surrounding tooth 14 crown
2.	Warter et al.,	8/M	Nigerian	1990	Maxilla sinus	Unilocular radiolucency, surrounding tooth 13 crown
3.	Tajima et al.,	15/M	Japanese	1992	Maxilla	A well-defined radiopaque mass and crown of unerupted 28
4.	Garcia-Pola et al.,	12/M	Spanish	1998	Maxilla	Unilocular radiolucency, surrounding tooth 23
5.	Bravo et al.,	14/F	Not stated	2005	Maxilla	Unilocular radiolucency, surrounding 23 crown
6.	Nonaka et al.,	13/F	Brazil	2007	Maxilla	Unilocular radiolucency with few radiopaque areas 23 and 24
7.	Chen et al.,	15/M	Chinese	2007	Maxilla	Impacted 23
8.	Sandhu et al.,	25/F	Indian	2010	Maxilla	Impacted 13
9.	J Baby John, Reena Rachel John	38/F	Indian	2010	Maxilla	Impacted 27
10.	Khot and Vibhakar	17/F	Indian	2011	Maxilla	Impacted 33
11.	Zama Moosvi	13/F	Indian	2011	Mandible	Impacted 32
12.	Anita Dnyanoba Munde et al.,	20/F	Indian	2013	Mandible	Impacted 33
13.	Vikramjeet singh et al.,	15/F	Indian	2012	Maxilla	Impacted 13
14.	Anshita Agarwal et al.,	15/F	Indian	2012	Maxilla	Impacted 23
15.	Sushruth Nayak et al.,	32/M	Indian	2012	Mandible	Impacted 43
16.	Latti BR, Kalburge JV	15/F	Indian	2013	Mandible	Impacted 33
17.	Harish Saluja et al.,	18/F	Indian	2013	Mandible	Impacted 43
18.	Shivesh Acharya	14/F	Indian	2014	Maxilla	Impacted 13
19.	Geetha NT et al.,	14/M	Indian	2014	Maxilla	Impacted 23
20.	Ludmila De Faro Valverde et al.,	17/F	Unknown	2014	Maxilla	Impacted 23
21.	Sumit Majumdar et al.,	14/F	Indian	2015	Maxilla	Impacted 23
22.	Manjunath BS et al.,	20/F	Indian	2015	Maxilla	Impacted canine
23.	Manjunath BS et al.,	19/F	Indian	2015	Mandible	Impacted 34
24.	Gupta S et al.,	12/M	Indian	2016	Mandible	Impacted canine
25.	Nath S et al.,	13/F	Indian	2017	Maxilla	Impacted 23
26.	Neha Singla et al.,	16/F	Indian	2018	Mandible	Impacted canine
27.	Patel HB et al.,	15/M	Indian	2020	Mandible	Impacted canine
28.	Pradipta Ramgonda et al.,	18/F	Indian	2021	Mandible	Impacted 44
29.	Atsushi Fujita et al.,	31/M	Japanese	2021	Mandible	Impacted 44
30.	Pawar SR et al.,	12/F	Indian	2022	Maxilla	Impacted 22
31.	Sharma S et al.,	22/F	Indian	2022	Maxilla	Impacted 22
32.	Tanha Khan et al.,	15/M	Indian	2023	Mandible	Impacted canine
33.	Present case	24/F	Indian	2024	Mandible	Impacted canine

aspect of impacted 33 (Figure 2). The lesion was roughly 36.7 millimeters in the anterior direction, 21.3 millimeters in the supero-inferior direction, and 24.6 millimeters in the labiolingual direction. It was observed that adjacent teeth's roots had migrated and that there was external root resorption. There was buccal and lingual cortical plate expansion along with partial loss in few areas.

According to the aspiration of contents from the swelling in clinical examination and radiological presentation, the differential diagnosis of dentigerous cyst, calcifying odontogenic cyst, adenomatoid odontogenic tumour, unicystic ameloblastoma, and odontogenic keratocyst were made.

Under local anaesthesia, an incisional biopsy was obtained and submitted for histolopathological analysis. The microscopic picture (Figures 3A, 3B, 3C) revealed sheets of epithelium in a whorled pattern, the cells of which are round to oval, arranged in a close proximation in the form of rosette bud pattern.) Homogenous eosinophilic areas interspred within proliferating tumor cells suggestive of cystic spaces along with areas of calcification. The overall picture was suggestive of "Dentigerous cyst undergoing Adenomatoid Odontogenic Tumor". The histological characteristics of the lining of the dentigerous cyst and the AOT, which manifest as rosettes and nodules, confirm that the cystic lining has transformed into a tumour in this case. This characteristic was also covered in case studies by Nonaka et al., Sandhu et al., Warter et al., and others[6,7,8].

Management

The procedure was performed under general anaesthesia. From tooth number 36 to tooth number 43, a crevicular incision was made, and a mucoperiosteal flap was elevated. The impacted mandibular canine, which was attached to the well-encapsulated tumour mass, was removed along with the lesion, which was completely excised in toto (Figures 4A, 4B, 4C). The surrounding bones showed no signs of invasion. Teeth 36, 35, 34, 32, 31, 41, and 42 were extracted, in addition to the over-retained deciduous canine. Resorbable suture material was used to close the surgical site. Following surgery, there was excellent post-operative healing (Figures 5A, 5B).

Discussion

An uncommon benign odontogenic epithelial tumour that develops from the complex or residual dental lamina system

Fig. 4 A: Intraoperative image showing intraosseous tumor mass in mandibular anterior region **B:** mandibular defect with thin lower cortical border remaining after removal of the pathologic tissue **C:** Enucleated pathologic tissue in toto along-with impacted mandibular canine.

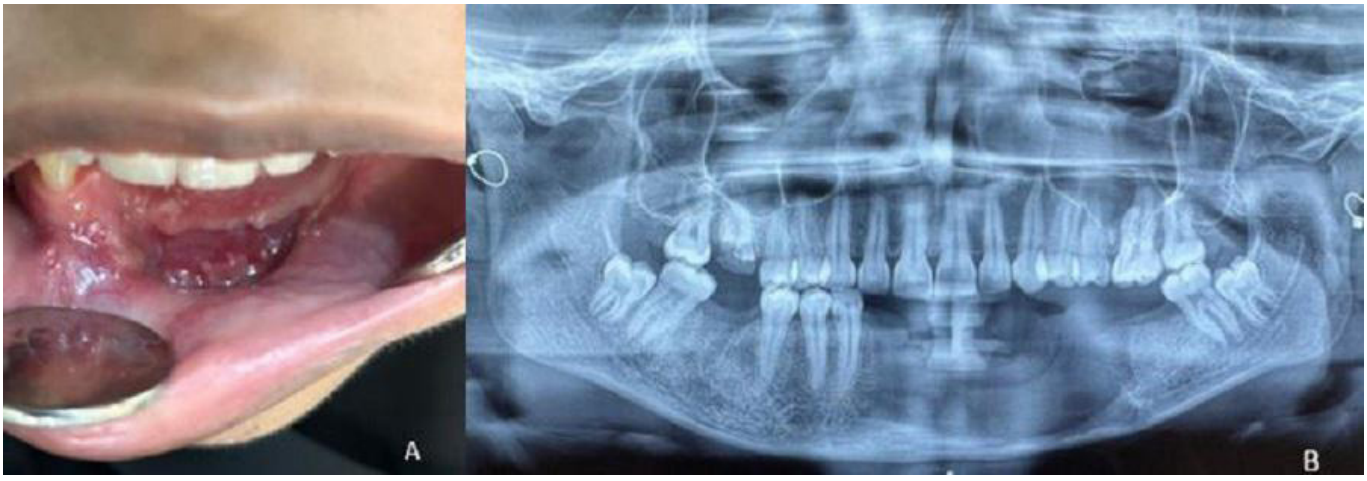

Fig. 5 A: Post-operative healing after 3 weeks **B:** Post-operative orthopantomogram

is called an adenomatoid odontogenic tumour. There are cases where odontogenic tumours and certain odontogenic cysts co-exist[9]. It is not clear whether the lining of an associated cyst is a true dentigerous cyst, a cystic change within an AOT, or some other entity all together. Because the majority of central AOTs originate in a pericoronal connection with an associated tooth, it is impossible to distinguish between the lining of an associated cyst that is a true dentigerous cyst and a subsequent cystic change inside the AOT. A comprehensive analysis of the English-language medical literature yielded only 32 cases that were similar, of which only 12 involved AOT involvement and a dentigerous cyst in the jaw. Table 1 lists the age, location, radiographic findings, and clinical manifestation of all previous AOT studies, including the current case, that histologically demonstrated a cystic lining[10].

The aforementioned case report serves as an example of the distinctive radiographic and clinical characteristics of the follicular variety of the AOT, which mimics a dentigerous cyst. The likelihood of calcifying odontogenic cyst, odontogenic keratocyst, and unicystic ameloblastoma was ruled out in this case due to a well-demarcated radiolucent lesion associated with an impacted tooth[11,12]. Since calcifying odontogenic cysts and odontogenic keratocysts are typically multilocular and unrelated to the crown of an impacted tooth, they were excluded from consideration as well. In the majority of cases, dentigerous cysts and unicystic ameloblastomas are found in the posterior jaw and are connected to the third molars. In contrast, adenomatoid odontogenic tumours mostly affect the maxillary anterior region and exhibit a 60% propensity to affect canines[13].

Under both microscopic and visual inspection, AOT associated with dentigerous cysts may reveal one or more cystic cavities lined by non-keratinized stratified squamous epithelium. In the present case, the cyst lining and the tumour component were continuous. In light of this, we think that a dentigerous cyst containing an unerupted tooth forms initially, maybe as a result of an unknown stimulus; AOT would then emerge from the dental lamina's epithelial remains within the lining of the odontogenic cyst. Further studies by Manjunatha BS et al., Sandhu SV et al., and Pradipta Ramgonda et al.[14,15] supported these results, suggesting that the dentigerous cyst undergoes a neoplastic transformation. Nevertheless, Chen YK et al. and Agrawal A et al., in their study, consider the idea that this type of lesion is a unique hybrid variation[16,17].

Conclusion

Adenomatoid odontogenic tumours grow slowly but steadily and are benign (hamartomatous), non-invasive lesions. The majority of these types of tumors have been removed surgically with precision and caution. There is very little likelihood that this diseased entity will reoccur. In our case, the recovery was uneventful with no indications of recurrence on follow up.

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