CASE REPORT

A Rare Case of Unusually Big Adenomatoid Odontogenic Tumor

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ABSTRACT

Of all odontogenic tumors, adenomatoid odontogenic tumors (AOT) are extremely rare, making for about 3% of cases. It is a benign, slowly developing, encapsulated, non-invasive, and non-aggressive odontogenic lesion connected to an impacted tooth. For years, these lesions could go undetected. The lesion does not return after the standard course of treatment of enucleation and curettage.[1]

Here, we report an uncommon case of an unusually large AOT in a 16-year-old female patient from northeastern India, who presented with complaints of a swelling over her left side of face for past 5 years associated with bilateral nasal blockage. Clinical examination and investigations revealed a hard swelling with some cystic regions of size 7.6 (CC) x 7.2 (AP) x 6.6 (TR) cm. The patient underwent staged removal of the tumor and the histopathological examination of the cyst wall showed features suggestive of adenomatoid odontogenic tumor. The patient remains symptom free on a follow up period of 18 months.

Keywords: Adenomatoid Tumor, Odontogenic Cyst, Odontodysplasia

INTRODUCTION

Adenomatoid odontogenic tumor (AOT) is a rare benign, non-invasive, slow growing odontogenic lesion that typically affects canine teeth which are impacted in young children. 3% to 7% of odontogenic tumors are AOTs.[1] The idea that the AOT, which was previously described in the literature under a number of different names, including drusiger Typ des Adamantinoms, ameloblastic adenomatoid tumor, adenomatoid or pseudoadenomatous ameloblastoma, adenomatoid or pleomorphic adenoma-like tumor, and epithelial tumor associated with developmental cysts, is a clinical and histologic entity among the odontogenic tumors. In their "Histological Typing of Odontogenic Tumors, Jaw Cysts, and Allied-Lesions" publication from 1971, WHO accepted the idea that the AOT should not be viewed as merely a histologic variation of ameloblastoma and embraced the nomenclature put out by Philip sen & Birn.[2]

Under a microscope, the presence of so-called distinct "duct-like structures" gives the tumor an adenomatoid, or glandular, appearance.[1] AOT's histogenesis is still unknown and is occasionally referred to as a hamartomatous lesion. The tumor is sometimes referred to as "Two Third's Tumor" because around two thirds of instances occur in the maxilla, roughly two thirds of cases are diagnosed in young females during the second decade, and roughly two thirds of cases are linked to an impacted tooth, which is canine in two third of cases.[3]

We present a case of unusually large odontogenic tumor in the maxilla in a 16-year-old female patient.

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CASE REPORT

A 16-year-old female presented to the outpatient department of a tertiary healthcare center in North eastern India with complaints of left sided facial swelling for the past 5 years associated with bilateral nasal blockage for past 2 months. The swelling has gradually progressed in size with no complaints of associated pain, fever or any sinus formation. The patient gave history of dental extraction prior to onset of the swelling. The swelling has resulted in significant deformity of the left side of the nose and face along with reduction in left eye-opening and nasal blockage which has aggravated over the course of past 2 months. On clinical examination, a single hard, non-tender, non-mobile swelling measuring approximately 10×8×5cm with smooth surface and regular margins was palpated over the left side of the face with obliteration of the left nasal cavity and the left nasolabial fold. The swelling was also visualized intraorally with involvement of the left upper alveolus and obliteration of the left upper gingivolabial and gingivobuccalsulci.

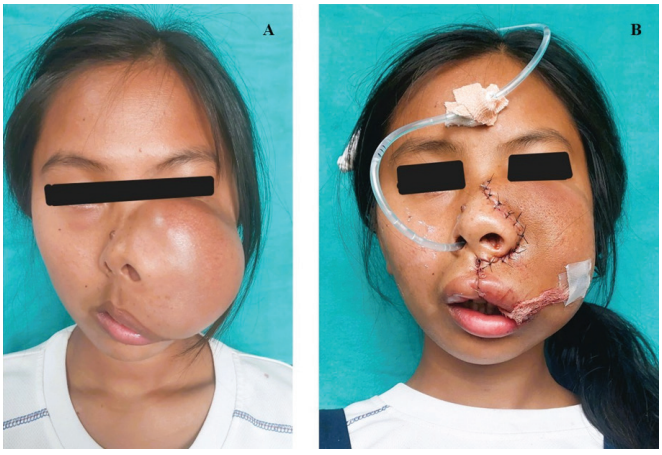

Figure 1: (A) depicts the pre-operative clinical photograph of the patient. (B) depicts post-operative photograph at day 7.

The patient was admitted and planned for surgery. Routine blood investigations revealed no significant abnormality.

Contrast-enhanced computed tomography (CECT) of the paranasal sinuses and facial bones revealed a large, well-demarcated, expansile cystic lesion originating from the left maxilla. The lesion measured approximately 8 cm craniocaudally (CC), 7.2 cm anteroposteriorly (AP), and 7 cm transversely (TR), occupying a significant portion of the left midfacial skeleton. The cyst exhibited thinning of the surrounding cortical bone, indicating chronic pressure-induced remodeling without frank cortical breach.

Within the lesion, an unerupted or impacted tooth was visualized along its superior aspect, with the crown protruding into the cystic cavity. A second unerupted/impacted tooth was identified superior and posterior to the first, positioned just outside the cystic boundary. These findings are consistent with the follicular origin commonly seen in odontogenic lesions.

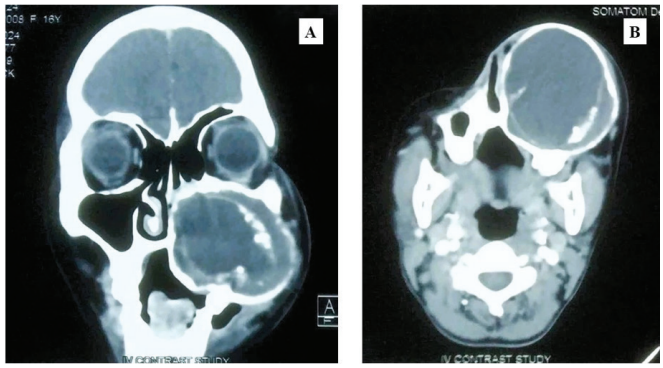

Figure 2: Contrast enhanced computer tomography scan image depicting the tumor in coronal view (A) and axial view (B).

The lesion extended medially into the left nasal cavity, causing marked deviation and near-complete obliteration of the nasal airway on that side. Superiorly, the cyst abutted and displaced the inferior orbital wall, which formed the superior boundary of the lesion. Anteriorly, there was significant thinning and outward bulging of the anterolateral wall of the maxilla, with stretching of the overlying facial skin, suggesting a longstanding expansile process. The anterior wall of the left maxillary sinus was bowed outward, and the sinus cavity itself was nearly entirely obliterated.

Notably, the lesion exhibited an inner rim with scattered foci of calcifications, a radiographic feature that may suggest the presence of odontogenic epithelium or dystrophic calcification within a neoplastic process.

Fine-needle aspiration cytology (FNAC) of the lesion yielded hemorrhagic fluid containing scattered inflammatory cells, predominantly neutrophils and histiocytes, in a background of red blood cells. No epithelial cells or atypical features were noted. The cytological findings were suggestive of a benign odontogenic cyst, with a differential diagnosis favoring a dentigerous cyst at that stage. However, the presence of calcifications and associated impacted teeth warranted further histopathological confirmation.

DIFFERENTIAL DIAGNOSIS

Because adenomatoid odontogenic tumors (AOT) frequently exhibit painless swelling and absent permanent teeth, they can be mistakenly identified as dentigerous cysts or ameloblastomas.

Odontogenic keratocyst, calcifying odontogenic cyst, calcifying odontogenic tumor, unicystic ameloblastoma, and keratocystic odontogenic tumor are further differential diagnosis for AOT.

TREATMENT AND OUTCOME

The patient was operated in a two staged procedure. In the first stage, marsupialisation and evacuation of the cyst contents was done using a canine fossa antrostomy. Fluid aspirated

from cystic lesion. Cyst wall was incised. A tooth was found in the superior wall and extracted. The 2nd stage surgery of partial maxillectomy with excision was done. Weber Ferguson incision was made. Flap was elevated. The entire anterolateral wall of maxilla was pathological as it was bulging out. Diseased bone removed. Inner cyst wall excised in piecemeal from the maxilla. Bony edge smoothened using diamond burr drill. After achieving haemostasis, maxilla packed with betadine-soaked ribbon gauze. Wound was closed in 2 layers using 3-0 vicryl, 3-0 ethilon. In the sublabial region the wound was left partially open to facilitate regular cleaning and dressing of the maxillary sinus. The opening was closed after 10days with secondary suturing using 3-0 vicryl. All sutures were removed and the patient discharged on post-operative day 10.

Histopathological examination of the cyst wall revealed a predominantly cystic swelling with thick fibrous capsule. Focal areas showed nodular proliferation of odontogenic epithelium involving duct like structures which are lined by columnar to cuboidal epithelium. Areas of calcification were also noted. The features were suggestive of adenomatoid odontogenic tumor.

The patient was doing well on 18 months follow-up and was advised regular follow up.

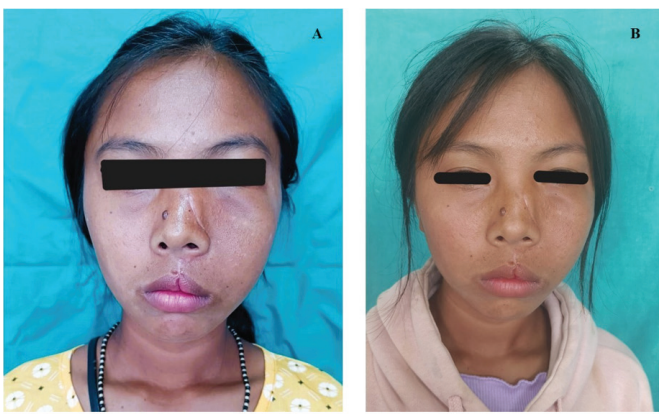

Figure 3: (A) depicts follow up at 1 month of surgery and (B) depicts at 18 months of surgery

DISCUSSION

Different titles have been used in the past in the literature for cases that are comparable to AOT.

The English researchers James and Forbes reported a case of epithelial odontome, a condition related to AOT, in 1909. A case of cystic adamantoma was reported by Harbitz of Norway in 1915. Wohl of Omaha described an AOT-like case in 1916 with a mandibular tooth germ cyst (also known as chorioblastoma). The first series of AOT was reported by Stafne in 1948 and was titled "epithelial tumors associated with developments cyst of maxilla."

The term "adeno ameloblastoma" was first used in a case published by Bernier and Tiecke.

Abrams et al. proposed the name "odontogenic adenomatoid tumor" in 1968. Philipson and Birn first proposed the term "adenomatoid odontogenic tumor" in 1969.[1] AOT occurs in three variants: 1) extrafollicular, 2) follicular, and 3) peripheral. A central intraosseous lesion connected to an impacted tooth is known as the follicular form. Approximately 75% of all documented cases of AOT are thought to be of this prevalent variety. The extrafollicular kind is an intraosseous lesion, usually found between the roots of neighbouring erupted teeth, that is unrelated to an unerupted tooth. Rarely do peripheral kinds develop in the gingival tissue. One of the two lobes of the tumor, which was partially in soft tissue and partially in bone, was situated in the maxillary bone in an uncommon case that has been documented.[4]

According to reports, radiographically, two-thirds of intraosseous AOTs show up as tiny radiopaque foci within well-defined unilocular radiolucencies. However, a greater frequency of central AOTs presenting as unilocular radiolucencies without radiopaque foci was noted by Arotiba et al. and Leon et al. It's still unclear how AOT originates. According to Philipsen et al., AOT might originate from the dental lamina's intricate structure or its leftovers. Nonetheless, the derivation of these tumors from the diminished enamel epithelium may be supported by the findings of ultrastructural and immunohistochemical investigations.[5]

Under a microscope, an extensive fibrous capsule typically envelops AOT. The tumor is made up of solid nodules of spindle-shaped, polygonal, or cuboidal epithelial cells that form nests, rosette-like structures, duct-like spaces, and strands of epithelium with a trabecular or craniform configuration. There is eosinophilic amorphous material (often referred to as "tumor droplets") between the epithelial cells and in the core of the rosette-like forms. In addition, the majority of lesions contain varying degrees of calcified material.[5]

Very few cases have been reported in literature of large AOTs. In our case the unusual large size of the lesion may be attributed to delayed treatment seeking due to remote areas in Northeast India as well as higher growth rate being in a young girl. Though the treatment usually consists of enucleation and curettage, in our case due to the large size and expansile nature of the tumor with facial disfigurement, a partial maxillectomy with excision was opted for.

CONCLUSION

To prevent needless mutilating surgery, a correct histological diagnosis is essential, even if enucleation and curettage is the most often used therapy technique for AOT sometimes necessitating excision. Although usually small and slow-growing, delayed diagnosis can result in unusually large lesions requiring more extensive surgical intervention. Multistage surgical approaches are usually required for giant lesions causing facial disfigurement and sinus obstruction. Early intervention and regular follow-up are critical to prevent complications and improve patient outcomes.

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