Adenomatoid odontogenic tumor: A fluctuating tumor entity

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Abstract
Adenomatoid odontogenic tumor (AOT) is a rare, odontogenic tumor frequently misdiagnosed; nonetheless, it has three well-documented clinicopathological variants and has been described as ‘two-thirds tumor’ because of its occurrence in the maxilla in about two-third cases, predominantly in females of younger age, in association with tooth of uneruptive nature, and most frequently in canines. In the recent years, many cases of AOT have been reported in the uncommon sites such as mandible and posterior regions of maxilla with various histopathological presentations. The aim of the present review is to understand the various clinical, histopathological, and radiological features of AOT that have to be taken into consideration for arriving at accurate diagnosis of this so-called ‘fluctuating tumor.”

Keywords
Adenomatoid odontogenic tumor, benign, mandible, odontogenic adenomatoid cyst, unerupted tooth

Introduction
Adenomatoid odontogenic tumor (AOT) is considered as a benign, non-neoplastic (hamartomatous) tumor, with a slowly progressing growth. AOT is mostly infrequent, benign neoplasm and organizes around 7% of all tumors of odontogenic origin.\(^1\) It has been proposed that it may be a hamartomatous origin rather than a true neoplasm.\(^2\) The tumor presents as swelling either intraorally or extraorally in the maxilla but less common in the mandible and is referred as a two-third tumor because of its occurrence in the maxilla, two-third in young females, two-third cases associated with tooth that has not erupted, and two-third in canines.\(^3,4\) It has been suggested that the components of other odontogenic tumors are frequently witnessed in relation to AOT and most common being calcifying cystic odontogenic tumor. These tumors are termed as hybrid tumors.\(^5\) Some authors preferred to disagree this term AOT or hybrid tumor; according to Marx and Stern, the more apt term is adenomatoid odontogenic cyst (AOC) since they observed the presence of unilocular cystic lesion, fluid on aspiration, and cystic cavity on transection, hence to some extent sustenance the terminology AOC.\(^6\)

AOT History and Terminology
AOT is a rare distinct, odontogenic non-neoplasm. De Matos FR et al in his study pointed out that In 1903, Nakayama reported two cases of AOT with convincing proof based on both clinical and histopathological evidence, the name which was first termed by Steensland in 1905.\(^7\) Dreibaldt in 1907 described it as pseudoadenoameloblastoma. Philipsen HP in his recent study cited In 1909, James and Forbes from England reported a case of epithelial odontome similar to AOT.\(^8\) Harbitz in 1915 termed it as cystic adamantinoma and Wohl of Omaha in 1916 reported AOT-like feature case as tooth germ cyst of jaw.\(^9\) Ghosh in 1934 designated it as an adamantinoma of the maxilla. Stephne in 1948 first accepted AOT as a distinct pathological entity.\(^1\) Bernier and Tiecke were the pioneers by naming it as adenoameloblastoma. In 1961, Gorlin et al. presented the term adenomatoid ameloblastic tumor. Okuyama described an indisputable case of AOT calling it a “tooth cyst.” It was Philipsen and Bern in 1969, who coined the term AOT which were widely accepted. The term was accepted by the WHO in 1971 and retained in the second edition of WHO in 1992. Unal et al. in 1995 generated a list comprising all terms of
AOT reported in the literature such as adenoameloblastoma, epithelioma adamantinum, ameloblastic adamantinoma, and teratomatus odontoma. In 1998, Reichert and Philipsen presented the update on AOT based on more than 600 cases. However, in 2007, Philipsen also stated that several cases of AOT reported earlier did not fit into the terminology so-called as AOT. The terminology is still an argument from Kolaczek in 1877 to Reichart in 2016.

Clinical Presentations

AOT presents as three variants, namely, follicular, extrafollicular, and peripheral type, which was originally described by Philipson and Reichart in 1991 and 1992. In the follicular type or pericoronal type, AOT is related with unerupted tooth; however, in the extracoronal type, AOT has no association with the crown of an unerupted tooth. They also indicated that peripheral adenomatoid odontogenic tumor (PAOT) should be divided into two submodifications, namely, peripheral PAOT and erupted intraosseous PAOT. Clinical presentations of AOT in the past years show several variants ranging from a painless to painful swelling intraorally as well as extraorally.

Lee et al. presented a case of maxillary left permanent canine and painless swelling with the retention of left maxillary deciduous canine and the permanent canine missing. An impacted supernumerary tooth and the permanent canine involved which is the most common as mentioned by Philipson et al. as the follicular type accounting for 70-73% of all the cases in literature, followed by 24% of extrafollicular and 4-7% of peripheral variants. Garg et al. presented as case involving the right side with the involvement of infraorbital margin. Shreedhar et al. presented similar case with involvement of maxillary sinus which was asymptomatic. The palatal vault was malformed and the egg shell crackling was observed. All these features present the asymptomatic, slow-growing nature of this tumor.

Although common in the maxilla, many cases are described in the mandible both in anterior as well as posterior regions. Batra et al. in their study presented a case of obliterated labial vestibule by buccal cortical plate expansion from lower right canine to left first premolar with the incisors displaced labially and missing permanent canine. In a study Zhang et al presented a case of hybrid tumors with a painless swelling in the anterior mandibular region extending from left first molar till right first premolar as a massive swelling with associated lower lip paresthesia, unerupted lateral incisors in a 64 years female which is a very rare observation of this age group. Panjwani et al. in 2010 reported a patient with unilaterial swelling in relation to 32 and 34 presenting an extrafollicular peripheral variant on the attached gingiva with evidence of egg shell crackling. Sharma et al. in 2012 showed an unusual variant with swelling extraorally extending from midline till anterior border of ramus of the mandible with retained 83, 84, 85 and impacted 44, 45, 46 teeth.

Aggarwal reported the first case in the permanent mandibular molar region as in the literature available with tenderness present and impacted first premolar with displacement of roots of the canine and second premolar. Shresta reported a case in association of left mandibular canine and later with the eruption presented as an extrafollicular variant, leading to root dilaceration of left mandibular canine. Many other case reports by Jain et al. and Gomez et al. reported in the left posterior mandible and Vasudevan reported in the right anterior mandibular region.

It is also noticed that AOT of the mandible often associated with large lesion more than 1-3 cm in diameter with displacement of the adjacent tooth and root resorption. Cases have shown nonvital tooth that has created a dilemma considering maxillary as a predominant site without aggressiveness of this tumor rather than just a progressive growth.

Radiographic Features

AOT presents as a well-demarcated, round, well-circumscribed radiolucent lesion with distinct radiopaque margin associated with the supernumery or the impacted tooth inside the cystic area, but major difference being in dentigerous cyst; only coronal part is involved not the entire tooth. In case of extrafollicular variant unilocular, well-demarcated radiolucency was noted between or above, covering the roots of erupted tooth often resembling a radicular cyst, presence of discrete foci, flocculent pattern showing mixed radiopaque and radiolucency. About 27% of cases show radiolucency with no internal calcification. This has to be taken into consideration in the differential diagnosis of corticated radiolucency with small radiopaque foci, exclusively in teenagers and young adults.

Histopathological Features

Reichart and Philipsen proposed first characteristic pattern of AOT irrespective of tumor variants. Tumor shows various-sized solid nodules of cuboidal/columnar epithelial cells resembling rosette-like pattern or nests with scanty connective tissue stroma between the nodular-like epithelial cells and at the center of rosette pattern eosiophilic amorphous material known as tumor droplets with calcified bodies are present. Though odontogenic in nature, the presence of tubular or duct-like configuration gives the tumor, a glandular-adenomatoid appearance. Third typical cellular pattern is nodules composed of polyhedral, eosiophilic epithelial cells resembling squamous cells with amyloid-like material present inside. Occurrence of one or several nodules in this cellular arrangement had led a number of authors to suggest the existence of calcifying epithelial odontogenic tumor. Areas mimicking calcifying ghost cell tumor, developing odontomas, or other tumors should be considered as a variant of AOT. Lee et al. showed a cystic mass in which lining epithelium proliferated into the lumen in sheets and ball-like rests. Nonaka et al. observed fragments of odontogenic lesion characterized by the proliferation of fusiform/globular pattern, arranged as large islands and solid sheets and also numerous...
duct-like structures which are resemblance of dentigerous cyst as well as AOT in few areas. Another epithelial pattern presenting as a trabecular or cribriform pattern has also been described. The connective tissue is very loosely arranged with thin-walled congested vessels, characterized by marked changes in the endothelial lining. Gang et al. observed fibrous capsule is absent around the tumor which in contrast with the study done by Philipsen et al. Several authors exhibited the presence of hyaline, disorganized material, or calcified osteodentin with associated unproductive enamel which has been infrequently reported. Dentin or dentinoid containing tubules are rare. According to Philipsen and Nikai, these materials are believed to be consequence of degenerative process as odontogenic ectomesenchyme is not evident in AOTs and therefore should not be inferred as an induction phenomenon.

**Conclusion**

AOT though occur in anterior maxilla, here we have reviewed few cases of mandible based on available literature, many unreported cases should also have to be considered, yet it is assumed to have a low incidence. This variant clinical representation should be kept in mind while examining asymptomatic mandibular swelling. Irrespective of the location of this tumor, since most variants show benign nature and more or less all are encapsulated, surgical enucleation or curettage is the most preferred choice of treatment. Recurrences involving mandible is still an uncommon finding. The debate on being a hamartoma has to be considered. It should be noted that AOT also behaved as a unique heterogeneous lesion that ranges from a hamartomas nature to benign and malignant neoplasm of various aggressive behaviors and also that few cases of follicular variants in the literature have shown extreme involvement of the face with perforation. The extrafollicular is very rare, but if present, it is more frequent in mandible than maxilla. Hence, by careful diagnostic procedures along with sufficient understanding of clinical, radiological, and histopathological suppositions can aid in obtaining a precise diagnosis of AOT which mimics various cystic lesions or other lesions of jaws.

**References**
