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Case Report

A rare entity of adenomatoid odontogenic tumor involving anterior mandibular region- A case report

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ABSTRACT

An adenomatoid odontogenic tumour (AOT) is a unique and somewhat uncommon odontogenic tumour that only arises from odontogenic epithelium and makes up between 2 and 7% of all odontogenic tumours and cysts. The lesion is categorised into three variants, the most common of which is the follicular type, which frequently presents as a dentigerous cyst clinically because it surrounds an impacted tooth. Because of the lesion's well-known ability to take on various clinical and histopathological appearances, it has been given the term "master of disguise". We report a case of AOT involving mandible in a 15-year-old male who reported to our department with a chief complaint of painless swelling in his mandibular region.

1. Introduction

Steensland originally described the adenomatoid odontogenic tumour (AOT) in 1905.[1] It is a rather uncommon unique odontogenic neoplasm. However, this tumour has been referred to by a number of other names. A list of all AOT nomenclatures recorded in the literatures was created by Neha S et al.[1,2] The disease now known as AOT has previously gone by several different names, including adenoameloblastoma, ameloblastic adenomatoid tumour, adamantinoma, epithelioma adamantinoma, and teratomatous odontoma.[1–3]

AOT is also a distinct lesion that manifests fairly consistently, which has led to its reputation as a "tumour of two-thirds," meaning that two-thirds of affected individuals are female, two-thirds develop in the second decade of life, two-thirds develop in the anterior sextant of the maxilla, two-thirds are superimposed on dentigerous cysts, and two-thirds of associated unerupted teeth are permanent canines. Additionally, in two-thirds of patients, the unilocular radiolucency exhibits sporadic dot-like (snowflake) opacities.[1,2,4]

2.7 to 7% of all odontogenic tumours are AOTs. The majority of cases occur in the maxilla in combination with an impacted permanent tooth in young females, with a male: female ratio of 1:2.[3,4] In terms of clinical and radiological appearance, AOT resembles other odontogenic unilocular cystic lesions surrounding the unerupted teeth, such as dentigerous cyst or ameloblastoma. Additionally, the lesion may occasionally be revealed to have a cystic component histopathologically. AOT typically manifests clinically as a jaw-bone tumour that is slow-growing and symptom-free. AOTs are recognised as the second or fourth most prevalent odontogenic tumour in large-series investigations of odontogenic tumours.[5]

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2. Case Report

A 15 years old male patient reported to the department of Oral Medicine and Radiology with a chief complaint of swelling in the lower front tooth region since two months. The patient was apparently asymptomatic 2 months back, then he noticed a small swelling in his lower front tooth region which was non- tender at that time. As it was small and non- tender, he didn't consult any dental physician at that time. Initially the size of the swelling was small pea-sized and it showed gradual increase in size and has attained the present size. There was no pain associated with the swelling. Past dental history and past medical history were non- contributory.

Extra oral examination revealed a diffuse swelling is present on the chin area extending from left corner of the lip till 1 cm lateral to right corner of the lip mediolaterally and from lower lip till 1 cm below the lower border of the mandible superoinferiorly. The size of the swelling is approx. 6x4 cm. The surface of the swelling appears to be normal (Figure 1). On palpation, the swelling was firm to hard in consistency and mildly tender. Right submandibular lymph nodes were palpable and tender.

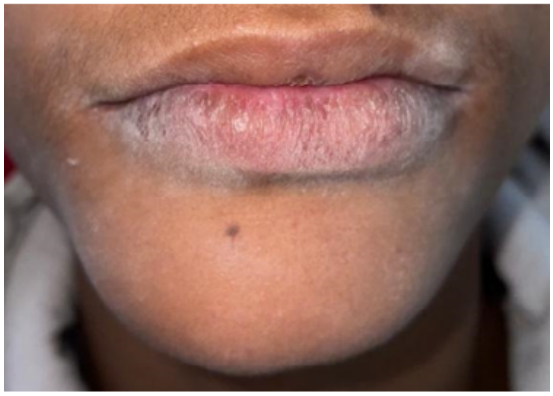

Figure 1: A diffuse swelling present on the chin region

Intra-oral examination revealed a diffused swelling is present extending from 33 to 44 region.

Clinically 43 was missing. Vestibular tenderness is present irt 33 to 44 region. On palpation, the swelling was firm in consistency and mildly tender. Retained deciduous irt 83 was also present (Figure 2).

Based on the history and clinical examination, a provisional diagnosis of Dentigerous cyst irt 43 was considered with a differential diagnosis of Adenomatoid Odontogenic Tumor irt 43.

Later, the patient was advised for OPG which revealed a well-circumscribed radiolucent lesion involving lower mandibular region extending from 33 region till distal to 45 region mediolaterally and from 1cm below the upper border of mandible till 2 cm above the lower border of mandible superoinferiorly. The border of the lesion appears

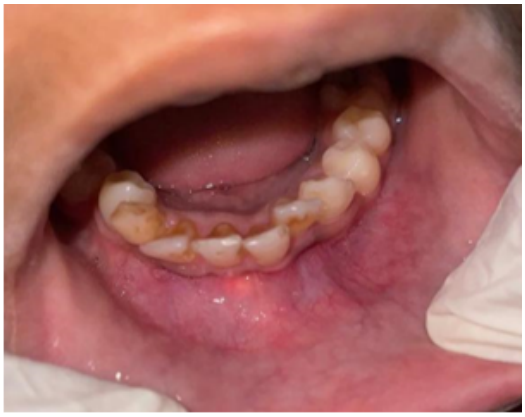

Figure 2: A diffuse swelling present extending from 33 to 44 region

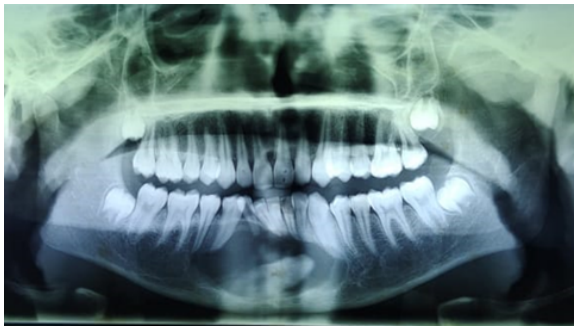

Figure 3: OPG revealing a well-circumscribed radiolucent lesion involving lower mandibular region

to be well- defined and corticated. The radiolucent lesion shows impacted 43 with well-defined radiopaque border. The attachment of the lesion appears to be apical to the CEJ along the root portion of impacted 43. Displacement of 31,32,33 is also seen (Figure 3).

The patient was then advised for histopathological investigation. The lesion was excised and submitted for the histopathological report. After the report, final diagnosis of "Adenomatoid Odontogenic Tumor" was made based on the histopathological report.

3. Discussion

Adenomatoid odontogenic tumours are a very rare neoplasm that make up around 2-7% of all odontogenic cysts and tumours.[5,6] It occupies a unique position as a result of the relatively lengthy discussion surrounding the nature of this pathology, with few authors considering it to be a developmental hamartomatous odontogenic growth in opposition to the current classification it holds in the WHO of odontogenic tumours and allied lesion as an odontogenic tumour. The WHO currently classifies AOT as a "benign tumour consisting of odontogenic epithelium in a variety of histoarchitectural patterns, embedded in mature connective

tissue stroma, and characterised by slow but progressive growth."[6,7]

Due to its close association with areas of the jaw that bear teeth as well as impacted teeth, as well as its cytological similarity to the remnants of enamel organ epithelium, reduced enamel epithelium, and rests of Malassez, AOT has traditionally been thought to be derived from the odontogenic apparatus.[8–10]

AOT is primarily seen in younger people, peaking in occurrence in the second decade,[9–14] and is very rare in patients over the age of 30. The maxilla has a striking preference for the anterior jaw and it occurs twice as frequently there as in the mandible. The usual male: female ratio is 2:1. With a few documented big lesions as exceptions, most AOT rarely exceed 3 cm in maximum diameter.[10–12]

AOT can be split into various variants according to the clinical and radiologic findings: 1) Central (or intraosseus) type: Follicular kind, resembling a dentigerous cyst because it encloses the crown of an immature tooth. 2) Extrafollicular type: The crown of an unerupted tooth is not associated with the extrafollicular type. Depending on the lesion's real intraosseous location, the preliminary diagnosis for this variety may be a "residual," a "globulomaxillary," or a lateral periodontal cyst. 3) Peripheral (or extraosseous) type: Variant that is peripheral (or extraosseous) and resembles a fibrous epulis or gingival fibroma.[13]

Though uncommon, the peripheral or extraosseous type of the tumour typically manifests as tiny, sessile lumps on the face gingiva of the maxilla. These lesions cannot be distinguished clinically from the typical gingival fibrous lesions. They are typically asymptomatic and are found through routine radiography examinations or when trying to figure out why a tooth isn't erupting, with the maxillary canine being the most common culprit. Greater lesions cause the bone to expand painlessly.[12,14]

The tumour appears radiographically as a well-defined, unilocular radiolucency that affects the crown of an immature tooth, most frequently a canine. The radiolucency associated with the follicular form of AOT occasionally extends apically along the root past the cementoenamel junction, providing as a helpful differentiating factor. However, the radiolucency associated with the follicular type of AOT is impossible to distinguish radiographically from the more common dentigerous cyst. Rarely, AOT may appear as an extrafollicular type, well-defined unilocular radiolucency between the roots of erupting teeth.[11,12] The lesion may be totally radiolucent, although it frequently has little calcifications (snowflakes), which helps to distinguish it from dentigerous cysts and ameloblastomas. In the proximity of tumours, teeth displacement is a frequent occurrence. There may or may not be root resorption. The nearby cortical bone may have eroded due to the peripheral lesions.[3,6,14]

The central AOT typically manifests macroscopically as a well-defined round-oval mass that is usually encircled by a thick, fibrous capsule. When examined under a microscope, the surface may look solid, tan, or show the existence of numerous cystic regions that are different sizes and contain a yellowish-brown fluid-like substance. While grossing, it's possible to come across calcified masses that have a characteristically grittier consistency.[6,8,10]

The preferred treatment method is conservative surgical enucleation. Bone grafts, directed tissue regeneration using membrane technology, or waiting for the minor intrabony defects to heal can all be employed to treat big intrabony defects brought on by AOT. Because of its capsule, the adenomatoid odontogenic tumour separates easily from the bone and is totally benign. Aggressive behaviour and recurrence of AOT is rarely seen following enucleation.[1,12,14]

4. Conclusion

Even though AOT is uncommon in the mandible, we can infer that dental practitioner can encounter these cases often. Their remarkable characteristic features makes them simple to diagnose. The best course of treatment for these lesions is still surgical curettage followed by enucleation.

5. Ethical Clearance

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6. Source of Funding

None.

7. Conflict of Interest

None.

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