Case Report

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A Case of Adenomatoid Odontogenic Tumour in a 14-year-old Female

Dentistry Section

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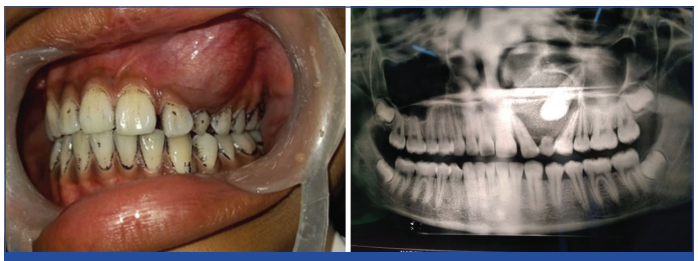
ABSTRACT

Adenomatoid Odontogenic Tumour (AOT) is a rare odontogenic neoplasm that primarily affects adolescents and young adults. The present case report discusses the presentation, diagnosis, and successful surgical management of AOT in a 14-year-old patient with a six-month history of upper left jaw swelling. Radiological investigations confirmed the lesion's characteristics, leading to a provisional diagnosis of AOT. Subsequently, surgical enucleation and histopathological examination {Haematoxylin and Eosin (H&E)} confirmed the diagnosis. The patient showed no recurrence during follow-up at three weeks and six months postsurgery, with excellent functional and aesthetic outcomes. Postoperative follow-up is essential for optimal patient care and long-term outcomes. The present case report contributes to the existing literature on AOT, providing real-world clinical insights and data, which aids in a better understanding and management of this rare condition.

Keywords: Management, Odontogenic neoplasm, Surgical enucleation

CASE REPORT

A 14-year-old female patient presented with a chief complaint of swelling in the left maxillary jaw region persisting for the past six months. The patient reported that the swelling had initially been small in size when it first appeared but had gradually increased in size over the months. No associated symptoms, such as pain or discomfort, were reported. The patient's past medical history was unremarkable, and there were no specific habits or relevant family history. Upon clinical examination, a firm, non tender swelling in the left maxillary region measuring 3 cm (length)×3.5 cm (breadth)×2 cm (width) was observed, particularly associated with teeth 22 and 63 [Table/Fig-1]. Radiographically, a well-defined radiolucent lesion with a corticated border was identified in the left maxillary anterior region, extending from tooth 22 to 63 [Table/Fig-2]. The lesion exhibited displacement of adjacent teeth but did not show root resorption or cortical perforation. Importantly, the unerupted permanent successor of tooth 63 was also noted.

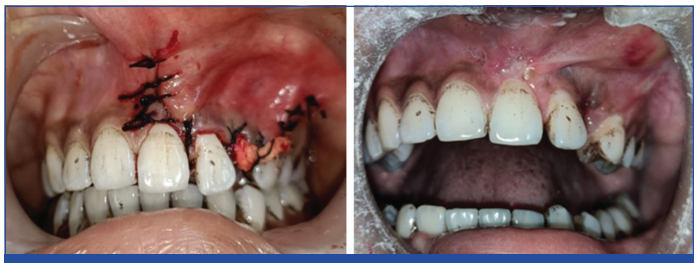


[Table/Fig-1]: Preoperative lesion.
[Table/Fig-2]: Preoperative Orthopantomogram (OPG). (Images from left to right)

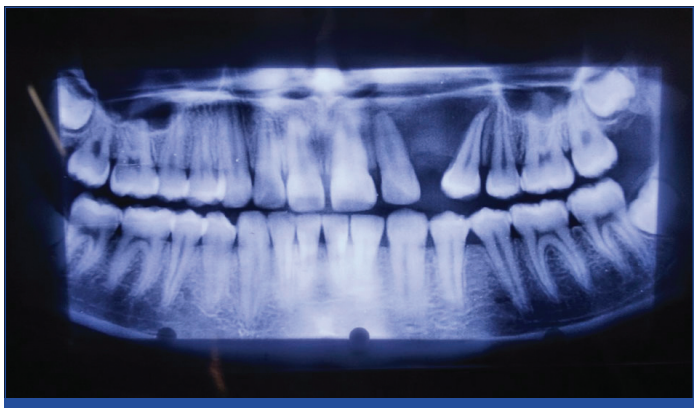
Based on the clinical and radiological findings, a differential diagnosis of unicystic ameloblastoma, dentigerous cyst, aneurysmal bony cyst, and AOT was made, with a provisional diagnosis of AOT. Subsequently, the patient underwent laboratory investigations including complete blood count, bleeding time, clotting time, prothrombin time with international normalised ratio, and serology tests for human immunodeficiency virus 1, 2, Hepatitis C, and Hepatitis B Surface Antigen, all of which were within normal limits. The aspirated fluid for total protein examination revealed a total protein content of 5.34 g/dL.

Subsequently, surgical enucleation of the cystic lining was performed under local anaesthesia [Table/Fig-3-5]. The unerupted permanent successor of tooth 63 was extracted along with the cystic lining due to the lack of bony support [Table/Fig-4]. The patient was diligently followed-up at three weeks and six months after the surgery. Clinical examination and radiographic evaluation during these follow-up appointments revealed no signs of recurrence or complications [Table/Fig-6,7].

[Table/Fig-3]: Surgical enucleation done.
[Table/Fig-4]: Excised tissue. (Images from left to right)

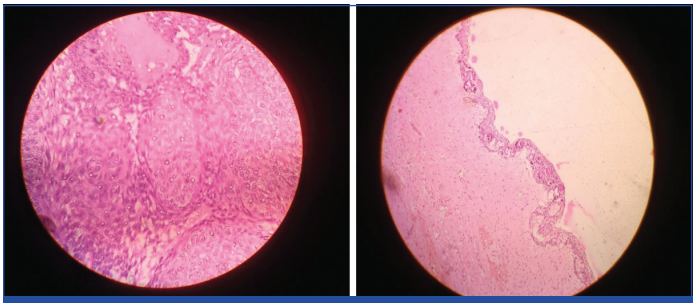


[Table/Fig-5]: Suturing done.
[Table/Fig-6]: 3-week follow-up. (Images from left to right)

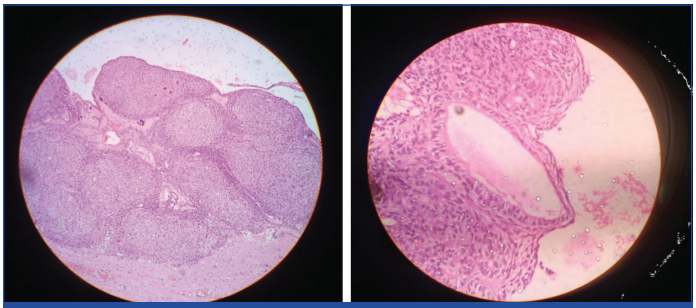


[Table/Fig-7]: Postopertaive OPG (6-month follow-up).

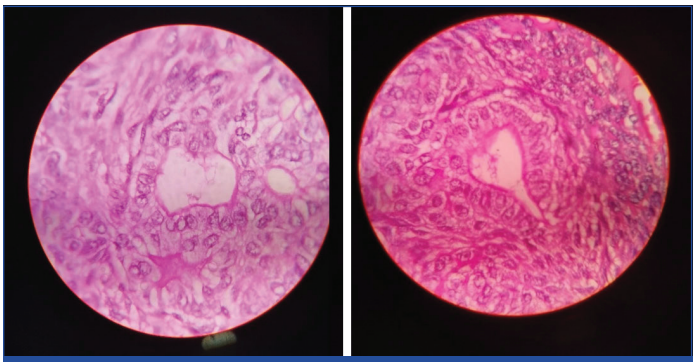
A histopathological examination was conducted on the excised tissue. Microscopic evaluation of the tissue sections revealed characteristic features of AOT, such as cellular multinodular proliferation and the presence of tumour cells arranged in duct-like structures and rosette patterns [Table/Fig-8-13]. Importantly, the histopathological examination ruled out any evidence of malignancy, confirming the diagnosis of AOT.



[Table/Fig-8]: (40X; H&E stain): **[Table/Fig-9]:** (10X, H&E stain): Images showing multiple areas of foci of calcifications observed between the tumour cells. (Images from left to right)



[Table/Fig-10]: (10X, H&E stain): Image showing sheets of spindle-shaped cells arranged in a ductal pattern. **[Table/Fig-11]:** (40X, H&E stain): Image showing tumour cells in ductal and rosette patterns. (Images from left to right)



[Table/Fig-12,13]: (100X, H&E stain): Image showing tumour cells in ductal and rosette patterns. (Images from left to right)

The patient remained asymptomatic throughout the follow-up period, and both functional and aesthetic outcomes were excellent. The present case emphasises the significance of timely diagnosis and surgical intervention in effectively managing AOT, ensuring optimal patient care while considering the status of adjacent teeth, including unerupted permanent successors.

DISCUSSION

Odontogenic tumours encompass a heterogeneous group of rare lesions primarily affecting the jawbones. Among these, AOT has historically been recognised as the fourth most prevalent odontogenic tumour. It is noteworthy that since 2017, the nomenclature of AOT has evolved, with the tumour being reclassified as an epithelial odontogenic tumour. This reclassification was guided by the principles of simplicity in communication and considerations of similar clinical aspects [1]. In India, AOT represented 7.6% of all odontogenic tumours, a range similar to that seen in other parts of the world [2].

One noteworthy aspect of AOT is its predilection for young individuals, with a higher incidence among females. This observation aligns with a previous retrospective study that revealed a female predominance for AOT, with global female-to-male ratios ranging from 1.9:1 [3].

The clinical presentation of AOT typically manifests as a painless swelling, often incidentally discovered during routine radiographic examinations [4]. Such asymptomatic growth is consistent with the present case report, where the patient exhibited gradual jaw expansion over a six-month duration. Radiographically, AOT is characterised by well-circumscribed, unilocular, or multinodular radiolucent lesions with corticated borders. These findings align with previous descriptions, and importantly, root resorption and cortical perforation are infrequent, serving as distinguishing features from other odontogenic tumours [5-7]. Histopathologically, AOT exhibits distinctive characteristics, including cellular multinodular proliferation and the presence of duct-like structures. The columnar cells lining these structures may display palisaded nuclei and cuboidal to columnar cytoplasm, giving rise to a stellate reticulum-like appearance [8,9]. This histopathological profile is consistent across all subtypes of AOT, which are uniformly well-encapsulated.

Regarding management, surgical enucleation and curettage are the preferred treatment modalities for AOT, consistently demonstrating effectiveness with a low recurrence rate, as supported by Woo VI et al., and Díaz Castillejos R et al., [10,11]. In cases where AOT leads to periodontal intra-bone defects, guided tissue regeneration using a membrane technique is recommended after complete tumour removal [12]. A summary of similar case reports has been presented in [Table/Fig-14] [2,6,9,11,13-18]. None of the cases had a recurrence.

S. No.	Study	Age (years)	Sex	Location	Treatment
1.	Bansal SP et al., 2022 [2]	19.8	F	Anterior maxilla	Enucleation
2.	Costa KB et al., 2021 [9]	16	F	Maxillary premolar region (right)	Decompression followed by enucleation with curettage
3.	Johny J et al., 2021 [13]	27	M	Anterior mandible	Deep curettage with enucleation
4.	Vanessa E, Roberto O 2021 [6]	17	F	Posterior maxilla	Enucleation and curettage
5.	Sangalette B et al., 2020 [14]	16	M	Anterior maxilla	Enucleation
6.	Díaz Castillejos R et al., 2015 [11]	13	M	Anterior maxilla	Enucleation and curettage
7.	Virupakshappa D et al., 2014 [15]	20	F	Anterior maxilla	Surgical management
8.	Prakasam M et al., 2013 [16]	19	F	Anterior maxilla	Surgical enucleation
9.	John JB et al., 2010 [17]	39	F	Anterior maxilla	Surgical enucleation
10.	Shetty K et al., 2005 [18]	12	M	Maxillary Cuspid and biscuspid area	Surgical enucleation

[Table/Fig-14]: Summary of recent studies on AOT [2,6,9,11,13-18].

CONCLUSION(S)

The present case report discusses the diagnosis and management of an AOT in a 14-year-old female patient. AOT is a rare odontogenic neoplasm primarily affecting young individuals, with a notable predilection for females. The patient's clinical presentation is characterised by a gradually enlarging swelling in the left maxillary jaw region over six months. The preferred management strategy is conservative surgical enucleation or curettage, which has a low recurrence rate. The case highlights the importance of timely diagnosis, surgical intervention, and postoperative follow-up in AOT management.

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