

A case of swelling in the upper front tooth region

Sanjay Chikkarasinakere Jogigowda ¹, Sathish Radhakrishna ², Karthikeya Patil ²

1. Department of Oral Medicine and Radiology, JSS Dental College and Hospital, JSS Academy of Higher Education and Research, Mysuru - 570015, Karnataka, India

2. Department of Oral and Maxillofacial Surgery, JSS Dental College and Hospital, JSS Academy of Higher Education and Research, Mysuru - 570015, Karnataka, India

SUBMISSION: 12/07/2024 | ACCEPTANCE: 05/09/2024

PART A

A 17-year-old female presents with a 1-year history of painless, slow-growing swelling in the right upper front teeth region. The patient reports a history of trauma to the upper front tooth region at age 12-13. Clinical examination reveals a well-defined swelling extending labio-palatally from the 21 to 14 regions, measuring approximately 4x5 cm, with obliteration of the labial vestibule. There are clinically missing 11, 13, and lingually placed 12, as well as fractured and retained 51, 52, and

53 (Figure 1). On palpation, the swelling is firm in consistency with crepitus or eggshell crackling. Teeth associated with the swelling are non-tender on percussion. The patient was subjected to CBCT (Cone Beam Computed Tomography) (Figure 2) and FNAC (Fine Needle Aspiration Cytology) (Figure 3) and later to surgical removal and histopathological evaluation of the same (Figure 4 and 5). Based on these clinical findings and investigations, what is the most likely diagnosis?



CORRESPONDING
AUTHOR,
GUARANTOR

Professor, Department of Oral and Maxillofacial Surgery
JSS Dental College and Hospital
JSS Academy of Higher Education and Research
Mysuru - 570015, Karnataka, India, E-mail- sathishr@jssuni.edu.in
Phone: +91 9845310737, ORCID ID: 0000-0001-5577-7210



Figure 1: A - Extraoral Frontal view; B - Intraoral view of the lesion

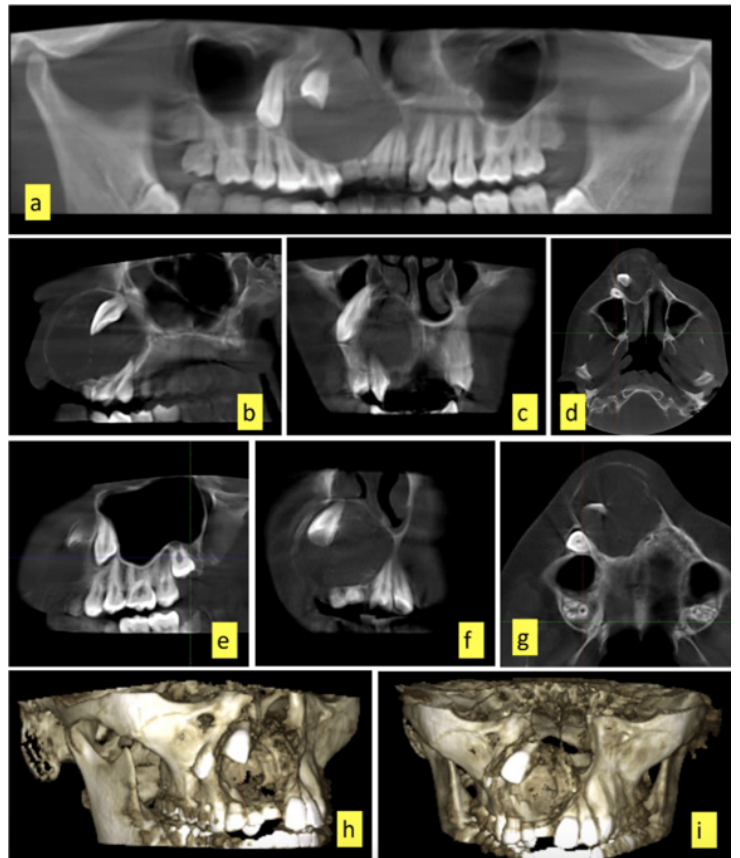


Figure 2: a - Orthopantomogram; b to g - CBCT images in axial, coronal and sagittal sections; h and i - 3D reconstruction images

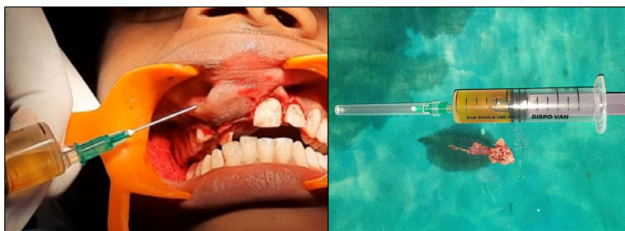


Figure 3: Fine needle aspiration and incisional biopsy

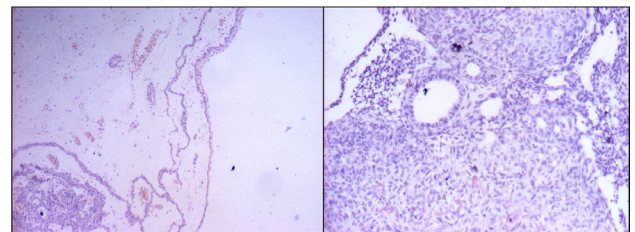


Figure 4: Histopathology of the FNAC and the incisional biopsy

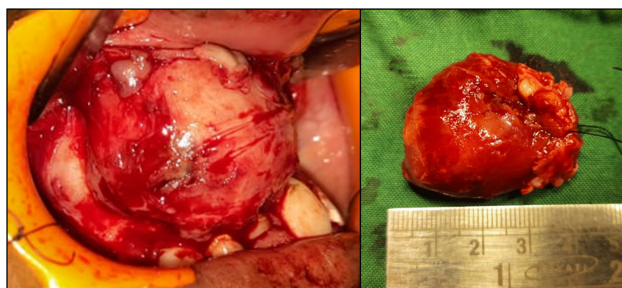


Figure 5: Gross specimen after excisional biopsy

PART B

Diagnosis: Adenomatoid Odontogenic Tumour arising from Dentigerous Cyst, associated with impacted 11

In 1948, Staphne was the first to identify and describe AOT as a distinct entity. [1] AOT is defined by the World Health Organization in its second edition of “Histological Typing of Odontogenic Tumours” as “a tumour of the odontogenic epithelium with duct-like structures and varying degrees of inductive change in the connective tissue”. [2]

The sclerotic margin was seen attached at the middle third of the root in our current case, but histological examination revealed features of both the dentigerous cyst and AOT. This encouraged us to review the scientific literature regarding the reported lesions of such associated pathology. Table 1 summarises the reported cases of the AOT associated with dentigerous cysts in English literature.

Typically, AOTs are solid, but they can be cystic, and to date, 22 cases have been documented to arise from the cystic wall of a dentigerous cyst. The majority of these cases have been reported in Asian countries in general, with 2/3rds of the cases coming from India. The maxillary canine was found to be the most frequently involved tooth (12 cases) followed by the mandibular canine (6 cases), one mandibular lateral incisor, maxillary premolars (2 cases), maxillary molars (2 cases), and a maxillary lateral incisor (two cases).

There is debate over the origin of the tumour from the cystic lining and the true nature of such lesions. Marx and Stern [3] classify AOT as a cyst rather than a tumour, implying that Adenomatoid Odontogenic Cyst (AOC) does not originate in the follicle of the tooth crown but rather from Hertwig’s Epithelial Root Sheath (HERS), which would account for the tooth being completely contained within the lumen rather than the tooth root being contained within a bony crypt. [4] In the present case, AOT might have developed from the dental lamina remnants induced by the trauma, along with a dentigerous cyst at the time of cyst expansion. The high proliferative activity of the dentigerous cyst that develops into AOT may be a result of inflammation caused by trauma (as in our case, trauma to retained deciduous teeth at the

age of 12) or infection. [5]

There is still debate over whether the tumour develops from the cystic lining, whether two pathologies co-exist concurrently, or whether numerous pathologies co-exist in a single lesion. As a result, a comprehensive assessment of anatomical changes, sectioning of gross specimens, and meticulous histopathologic investigation are required in order to unravel the enigma of tumour origin. [6]

Additionally, there is disagreement over the naming of cystic AOT, and as a result, the term “AOT arising from dentigerous cyst” can be used when a cystic tumour is discovered enclosing the crown of an impacted tooth and attached to the cemento-enamel junction by a thin non-keratinized stratified squamous epithelium, resulting in nodules of AOT in the cyst capsule. [7] Although studies indicate the occurrence of cystic AOT developing from dentigerous cysts, little differentiation was noted between the two except for size, and both require the same surgical excision. [8] As a result, subclassifying AOT into cystic and classic lesions serves no additional value.

Both dentigerous cyst and AOT can have origins in the dental lamina structures, and in this case, the patient has a history of trauma, as well as continuity of the tumour with the cyst lining, the development of AOT may be due to trauma from an existing dentigerous cyst lining. It does not warrant further dissection in nomenclature. However, the reporting of such cases enhances the understanding of their pathophysiology.

Declaration of patient/parent consent

The authors certify that they have obtained all appropriate patient consent forms. There, parents of the patient gave their consent that images and other clinical information can be used for scientific publication purposes. The parents understand that the names and initials will not be published, and due efforts will be made to conceal the patient’s identity, but also acknowledge that anonymity cannot be guaranteed. Ethical approval was waived off due to a retrospective case report. **R**

Prior publication: Nil

Source(s) of support: Nil

Conflicts of Interest: Nil



Figure 1: A - Extraoral Frontal view showing the asymmetry in the right upper lip region; B - Intraoral view of the lesion with diffused margins in the right upper anterior teeth region with no secondary surface changes and showing vestibular obliteration.

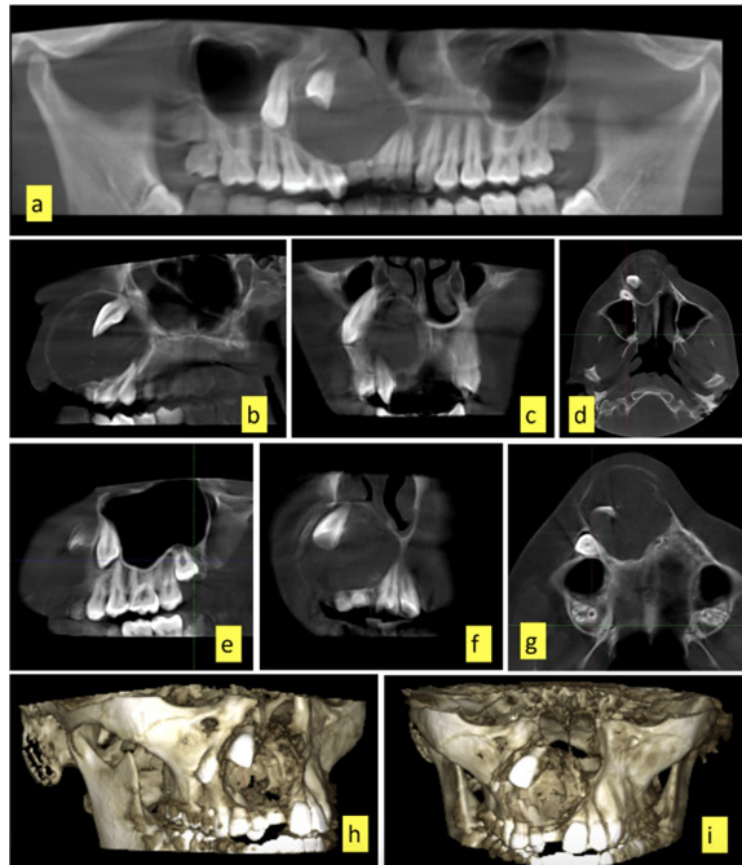


Figure 2: a - Orthopantomogram showing the well-defined radiolucency with an impacted tooth.; b to g - CBCT images in axial, coronal and sagittal sections showing the extent of the lesion and the position of the impacted tooth with the lesion; h and i - 3D reconstruction images showing the extent of bone resorption due to the lesion.

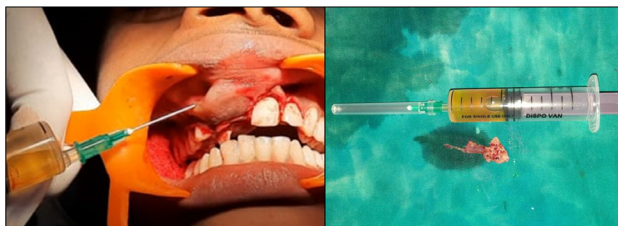


Figure 3: Fine needle aspiration and incisional biopsy revealing the straw-coloured fluid suggestive of a cystic lesion.

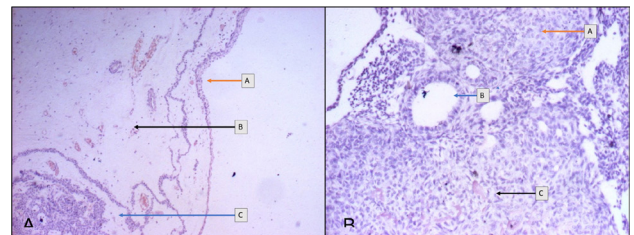


Figure 4: A - Stained section shows cystic cavity lined by 2-4 layers of cuboidal cells (A), underlying primitive connective tissue stroma (B) and tumour mass growing from the cystic lining (C). B - The stained section shows odontogenic epithelial cells in the form of sheets (A), ducts (B) and rosette form with eosinophilic secretions (C).

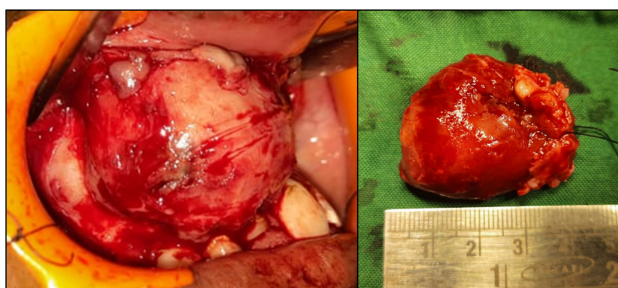


Figure 5: Gross specimen after excisional biopsy with cystic lining and also with a tumour gross appearance.



KEY WORDS

Odontogenic cyst, Adenomatoid odontogenic tumor, Clinical Pathology, Cone beam computed tomography, Aspiration

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CITATION

Sanjay Chikkarasinakere Jogigowda, Sathish Radhakrishna, Karthikeya Patil. A case of swelling in the upper front tooth region, p. 66-70. *Hell J Radiol* 2024; 9(3): 66-70.