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Clinicopathological Evaluation of Lesions Mimicking Dentigerous Cysts: A Case Series

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Abstract

Dentigerous cyst is an odontogenic cyst typically associated with the crown of an unerupted or developing tooth. However, several odontogenic cysts and tumors may present with overlapping clinical and radiographic features, thereby mimicking a dentigerous cyst. In some instances, gross examination of an excisional biopsy may reveal a cystic lining attached to the cervical region of an impacted tooth, supporting a provisional diagnosis of dentigerous cyst. Nevertheless, such lesions warrant careful evaluation, as they differ in etiology, biological behavior, histopathology, treatment modalities, and malignant potential.

The most common mimickers include Adenomatoid Odontogenic Tumor (AOT), Orthokeratinized Odontogenic Cyst (OOC), Odontogenic Keratocyst (OKC), and Unicystic Ameloblastoma, among others. This case series presents three lesions— AOT, OKC, and OOC—that were initially diagnosed as dentigerous cysts due to their association with impacted teeth. Subsequent clinicopathological and radiographic correlation, followed by histopathological confirmation from excisional biopsy, established the final diagnoses.

Given the diagnostic overlap, odontogenic cysts and tumors associated with impacted teeth must be thoroughly differentiated from dentigerous cysts. Accurate distinction is essential for determining appropriate treatment and predicting prognosis. Importantly, misdiagnosing an aggressive lesion such as OKC or unicystic ameloblastoma as a dentigerous cyst may lead to inadequate management and increased risk of recurrence. In scenarios of clinico-radiographic ambiguity, histopathological examination remains the gold standard for definitive diagnosis.

Keywords: Odontogenic Keratocyst (OKC), Adenomatoid Odontogenic Tumor (AOT), Orthokeratinized Odontogenic cyst.

Introduction

Dentigerous cysts also referred as follicular cyst, are the second most common type of cyst in the jaws and are among the most common developmental cysts of the gnathic bones, accounting for 17–25% of jaw cysts1. These cysts develop due to the accumulation of fluid between the reduced enamel epithelium and the dental follicle of an unerupted tooth2. By definition, a dentigerous cyst is associated with the crown of an unerupted or impacted tooth. They are associated with impacted tooth most commonly mandibular third molars followed by maxillary canines and less commonly mandibular premolars. They mostly affects mandibular posterior region mostly involving impacted third molar3. Radiographically, dentigerous cysts classically present as unilocular radiolucent lesions. The lesion is generally well defined and demonstrates a sclerotic border. Radiographically it has 3 variants. Central, lateral and circumferential4.

It is important that this definition be applied strictly as the diagnosis of dentigerous cyst should not be made solemly on radiographic evidence , otherwise keratocysts (OKCs) of the envelopmental variety, follicular OKCs, follicular variant of Adenomatoid

odontogenic tumor and unilocular ameloblastomas involving unerupted teeth, are at risk of being misdiagnosed as dentigerous cysts5.

Stafne in 1948, considered it as a distinct entity and the term "Adenomatoid Odontogenic Tumor" was later proposed in 1969 by Philipsen and Birn.6 Adenomatoid odontogenic tumor (AOT) is an uncommon benign odontogenic tumor of epithelial origin presenting in a various histopathological patterns. While these lesions are solid , they may occasionally be cystic. Most central AOT's occur in a pericoronal relationship with an associated tooth making it difficult to differentiate from dentigerous cyst7. Odontogenic keratocyst (OKC) is 'a benign uni- or multicystic intraosseous lesion of odontogenic origin, lined by parakeratinized stratified squamous epithelium and is known for aggressive, infiltrative behavior8. Approximately 30-40 % of OKC are associated with impacted tooth, mimicking the follicular variant of dentigerous cyst. Similarly, Orthokeratinized odontogenic cyst (OOC) first reported as a subtype of OKC by Wright in 19819. They are associated with impacted 2nd and 3rd mandibular molars. These cyst are unilocular and may radiographically resembles dentigerous cyst.

This case series presents 3 cases of diverse clinical, radiographic, gross examination and histopatholgy presentation. All three cases that mimics of dentigerous cyst in atleast one aspect of this presentation. This series highlights the importance of thorough evaluation of these lesions to differentiate them from there mimickers. This series emphasis on the role of excisional biopsy for final diagnosis.

Case 1 –

35 year old female patient reported to the department of oral pathology with chief complaints of pain and swelling on left side of the face since 2 month. She also reported a gradual reduction in mouth opening since the onset of swelling. The Patient's medical history was not contributory. On extraoral examination a diffuse extraoral bony hard swelling was noted on right side of the face which was tender on palpation. Swelling seen extended anteroposterior extension from angle of the ramus to the corner of the mouth and superoinferiorly from midcheek region to lower border of mandible.(Fig.1A) Mouth opening was reduced bilaterally. Intra-oral examination revealed a bony hard swelling on right side of the mandible with obliteration of buccal vestibule(fig.1B). Patient was further adviced to undergo for radiographic evaluatation. Orthopantamograph showed multilocular radiolucency with well corticated borders associated with impacted mandibular third molar(fig.1C). Cone Beam Computed tomography demonstrated buccolingual expansion of jaw(fig.1D). Based on Clinico-radiographical findings, a provisional diagnosis of OKC and Ameloblastoma was considered. Fine Needle Aspiration Cytology yielded blood tinged fluid containing Pus cells and abundant RBCs(fig.1E). An incisional biopsy was performed which were consistant with Odontogenic Keratocyst. Excisional specimen appeared as cystic lesion with its lining attached at the neck of an impacted tooth deceiving as Dentigerous cyst.

(fig.1F)

Volume 8, Issue 6; November-December 2025; Page No 263-270

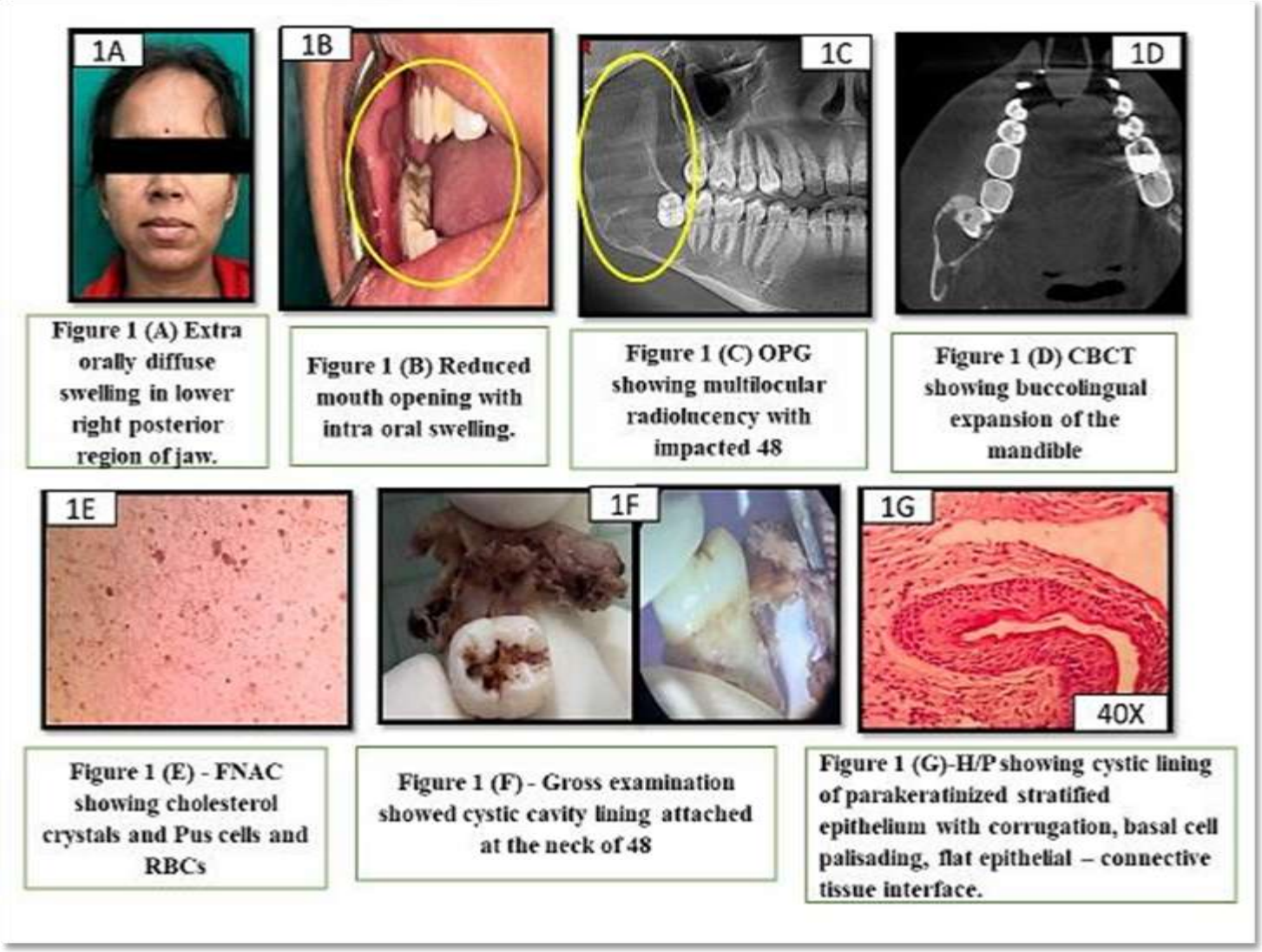

Figure 1 (A) Extra orally diffuse swelling in lower right posterior region of jaw.

Figure 1 (B) Reduced mouth opening with intra oral swelling.

Figure 1 (C) OPG showing multilocular radiolucency with impacted 48

Figure 1 (D) CBCT showing buccolingual expansion of the mandible

Figure 1 (E) - FNAC showing cholesterol crystals and Pus cells and RBCs

Figure 1 (F) - Gross examination showed cystic cavity lining attached at the neck of 48

Figure 1 (G)-H/P showing cystic lining of parakeratinized stratified epithelium with corrugation, basal cell palisading, flat epithelial – connective tissue interface.

Histopathological examination of specimen h & e stained section revealed a of cystic lining showing parakeratinized stratified epithelium of 6-8 layer cell layer in thickness, exhibiting corrugated appearance. Basal cell wear tall columnar cells with palisading or tombstone appearance. The epithelial connective tissue interface was flat and lifting of epithelium seen at places(fig.1G). The connective tissue capsule was thin and fibrocellular in nature. Overall feature suggested final diagnosis as Odontogenic keratocyst.

Case 2 –

15 year old male reported with chief complaint of pain and swelling on left side of the face since 4 month. The Patient also reported Difficulty in chewing food. On extraoral examination, there was a diffuse , Bony hard , non-tender swelling crossing midline on the lower left side of the face involving the lower border of mandible(fig.2A). Intraoral examination revealed obliteration of buccal vestibule extending 42 to 35 region(fig.2B) . An over-retained 73 and a missing lower left mandibular canine were noted. Radiographic assessment revealed a well defined unilocular radiolucency involving impacted 33 with thining of lower border of the mandible(fig.2C). FNAC showed blood tinged fluid containing pus cells and RBCs. For further analysis , an incisional biopsy showed a thin cystic lining composed of non keratinized stratified squamous epithelium with 2-3 cell layers thick and fibrocellular connective tissue wall suggestive of dentigerous cyst. Subsequent , excisional biopsy revealed a cystic lesion with its

Volume 8, Issue 6; November-December 2025; Page No 263-270

lining attached at the neck of the impacted 33(fig.2D). The cystic lining was thick and showed mural growth. histopathological examination of H & E stain section showed the cystic cavity lined by 2- 3 layers of non-keratinized stratified squamous epithelium. The lining composed of Cuboidal to low columnar cells with hyperchromatic polarized nuclei arranged in duct like or rosettes patterns. Amyloid like material was present at periphery of ductal lumen. & thick fibrous capsule seen at periphery(fig.2E). Overall features were characterstics for Adenomatoid Odontogenic Tumor.

Fig 2(A) -Extraoral view showing bony hard swelling in lower left side of mandible

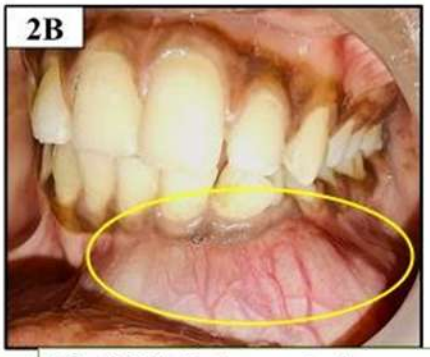

Fig 2(B) -Intraoral view showing bony hard swelling with obliteration of buccal vestibule in mandible

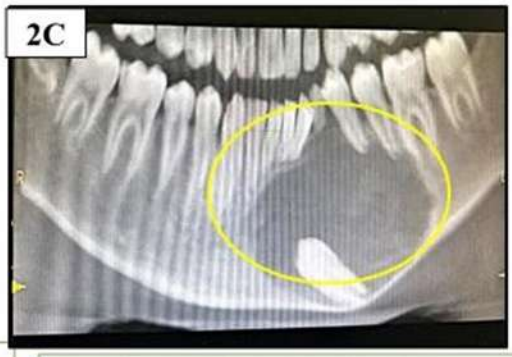

Fig 2(C) -OPG showing a unilocular radiolucency associated with impacted 33

Fig 2(D) -Encapsulated mass with cystic cavity lined at neck of impacted 33 demonstrating mural growth

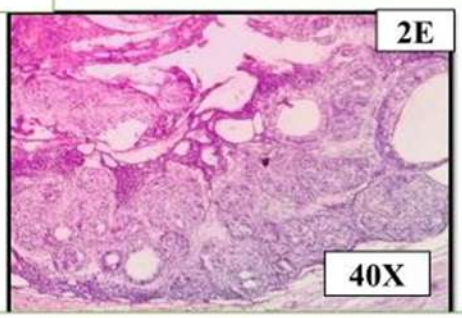

Fig 2(E) -Histopathology showing duct like areas, rosettes patterns, amyloid material

Case 3 –

A 36 year old male patient reported to the department of oral pathology with chief complaint of Pain and swelling on left side of the face for past 3 month. Extra oral examination revealed diffuse swelling that was tender on palpation(fig.3A). Intra orally swelling was seen in left retromolar region of mandible with impacted mandibular third molar(fig.3B)). Radiographic examination of orthopantamograph revealed a unilocular radiolucency with impacted 38(fig.3C). Aspiration cytology was inconclusive. Incisional biopsy showed features of an Infected dental cyst. Excisional biopsy showed gross specimen with cystic lining attached to the neck of the impacted tooth 38 at CEJ(fig.3D). Histopathological examination of H & E stained section demonstrated a cystic surface with thick , lamellated non corrugated orthokeratinized stratified squamous epithelium. A prominent granular cell layer was present , composed of flat cuboidal cells with absence of basal cell palisading(fig.3E) . Thus, based on this findings , the final diagnosis was given as Orthokeratinized odontogenic cyst.

Volume 8, Issue 6; November-December 2025; Page No 263-270

Fig 3(A) –E/O -bony hard swelling in lower left side of mandible

Fig 3(B) -Intraoral swelling in retromandibular area of left side of mandible

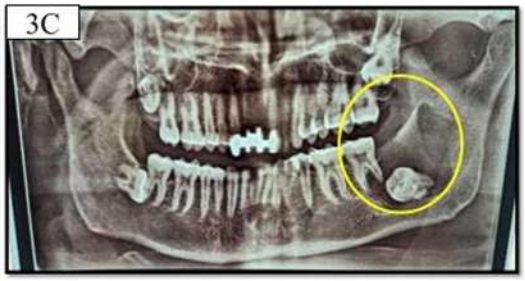

Fig 3(c) –OPG showing unilocular radiolucency with impacted tooth-38

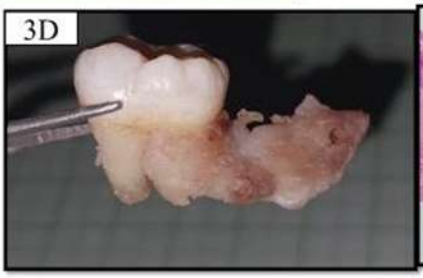

Fig 3(D) -cystic lining attached to the neck of tooth

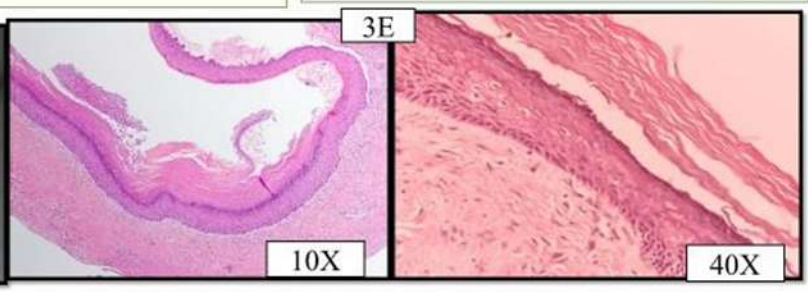

Fig 3(E) H & E stained section showing orthokeratinized epithelium with prominent granular cell layer

Discussion

Odontogenic cysts were first described by Gorlin and co-workers in 196210. Dentigerous cyst are the 2nd most common cyst of oral cavity. These Odontogenic cyst of epithelial origin account for approximately 15% of all true jaw cysts. Also known as follicular cyst, they occur more in the mandible (70 %) than in the maxilla11. The most commonly involved sites, in decreasing order are the mandibular third molars, maxillary canines and mandibular premolars followed by supernumerary teeth and central incisors. A dentigerous cyst develops when the follicle become separated from the crown of a developing tooth. As a result of this, fluid accumulates between the reduced enamel epithelium and tooth crown forming a cystic cavity that surrounds the crown of an unerupted tooth and is attached to the cementoenamel junction3. As a result, the cystic lining is characterstically attached at the neck of an impacted tooth. Radiographically these cystic lesion present as unilocular radiolucency around neck of an impacted tooth usually with corticated borders.

DC is the most common jaw lesion to be associated with an impacted tooth, while unicystic ameloblastoma, Adenomatoid odontogenic tumor ranks next to DC as the most frequently occurring pathologic pericoronal radiolucency12. Low prevalence of such an association with impacted teeth is reported for calcifying odontogenic cyst Orthokeratinized odontogenic cyst and odontogenic keratocyst12.

In the present case series, all the 3 cases were associated with impacted teeth. Two cases involved posterior mandibular region with impacted third molar. Radiographically, 2 cases exhibited unilocular radiolucency around the neck of an impacted tooth, resembling dentigerous cyst. One case showed multilocular radiolucency in posterior mandibular region where the differential diagnosis included OKC and Ameloblastoma. Aspiration cytology was

Volume 8, Issue 6; November-December 2025; Page No 263-270

performed for all the three cases. One case yielded straw coloured clear aspirate fluid containing abundant cholesterol crystal and pus cells. This aspiration fluid were charaterstic of Dentigerous cyst. The remaining two cases showed blood tinged aspirates suggesting an infective nature of the lesion. All the three cases underwent incisional biopsy. Histopathological examination confirmed in one case demonstrating diagnosis of Dentigerous cyst.

Surprisingly gross examination of excised lesion showed one characterstic feature in common for all the three cases i.e, cystic lining attached at the neck of an impacted tooth. This is the pecular feature of dentigerous cyst. Histopathological evaluation demonstrated that none of the cases were true dentigerous cyst.

The first case presented with multilocular radiolucency associated with an impacted tooth favouring a diagnosis of OKC, although it grossly mimicked dentigerous cyst due to its cystic lining attached at the neck of an impacted tooth. Although dentigerous cysts often appear as a simple radiolucent area surrounding the crown of an impacted tooth, large cysts may show a multilocular feature thereby mimicking lesions such as ameloblastoma and OKC13. Similar findings have been reported by Daware et al(2022)14 and Chaudhary et al (2013)15 who reported case of OKC presenting as dentigerous cyst. In present case , histopathological examination turn out to be odontogenic keratocyst for this case sloving the dilemma between multilocular radiolucency as OKC and not Dentigerous cyst.

The 2nd case also closely resembled a dentigerous cyst clinically, radiographically and on incisional biopsy. Though involvement of impacted mandibular canine and Gross examination revealed impacted 33 and showed the mural growth favouring towards adenomatoid odontogenic tumor but dilemma between Dentigerous cyst and Adenomatoid odontogenic tumor were resolved only after histopathological examination of excised tissue which showed features of AOT. This can be attributed to the theory of origin of AOT. One hypothesis suggest the origin of AOT is from the odontogenic epithelium of the dentigerous cyst, while some believe that the tumor could be derived from epithelial remnants of the dental lamina complex system. The follicular variant of AOT is associated with the crown and often part of the root of an impacted (unerupted) tooth16. According to Takahashi et al (2001)17, 77% of follicular variants of AOT are clinically & Radiographically misdiagnosed as dentigerous cysts. AOT usually occurs in 2/3rd cases in maxilla, involving canine. Similar findings of AOT mimicking dentigerous cyst has been reported by Bravo et al (2005)18, Uppada et al (2015)19, Manjunatha et al(2015)20

Orthokeratinized odontogenic cyst (OOC) is a uncommon developmental odontogenic cyst that arises from the cell rests of the dental lamina21. It shows male predominance and involved mandibular posterior region22. OOC usually associated with unerupted teeth, it can be confused with dentigerous cyst clinically and radiographically. In present case series, third case demonstrated a unilocular radiolucency with cystic lining attached at neck of an impacted 3rd molar(38) were deciptive of Denigerous cyst which were only ruled out after an excisional biopsy. Though gross examination of excisional lesion favouring diagnosis of dentigerous cyst, histopathology played an important role to give final diagnosis as OOC and not Dentigerous cyst. Nandini et al (2022)23 and Gunasekaran et al (2018)24 reported the cases of orthokeratinized odontogenic cyst with lining attached to neck of impacted tooth mimicking dentigerous cyst.

The common features favouring dentigerous cyst and features differentiating from dentigerous cyst for individual lesion has been summerised in table no 1.

Volume 8, Issue 6; November-December 2025; Page No 263-270

Dentigerous cyst	**Case 1**	**Case 2**	**Case 3**
	All 3 cases Clinically , Radiographically , and histopathologically mimics dentoigerous cyst		
Key features Mimicking dentigerous cyst	impacted teeth , buccolingual expansion	Unerrupted tooth , well-defined unilocular radiolucency	Impacted tooth , unilocular radiolucency
Key feature differentiating from dentigerous cyst	Multilocular radiolucency parakeratinized stratified squamous epithelium with a corrugated surface and a palisaded basal layer	Encapsulated mass, Contains calcified flecks, Duct like epithelial structures, rosettes, and eosinophilic material (amyloid-like).("snowflake" calcifications) on radiographs.	orthokeratinized stratified squamous epithelium prominent granular cell layer

Overall, odontogenic cysts share common clinical, radiographic and histopathological features. frequently these lesions closely resemble one-other making the initial diagnosis challenging. Majority of such lesions tends to occur in the posterior region such as dentigerous cyst, OKC, Orthokeratinized odontogenic cyst ,ameloblastoma. Many times these lesions include impacted tooth, mimicking more commonly Dentigerous cyst. This diagnostic dilemma can be challenging for the accurate diagnosis. Each lesions have their unique behavior and rate of recurrence. Though lesion like OKC are benign in nature, they are aggressive, diagnosed in later stage and have high rate of recurrence. Misdiagnosing such lesion as dentigerous cyst can lead to failure of treatment and recurrence. On the other hand, lesion like AOT though considered as benign tumor, is an well defined encapsulated mass with well corticated border. They have no tendency of recurrence. Treatment like complete enucleation

with preservation of impacted tooth can be done for such lesions.

Conclusion

This case series reinforces the necessity of correlating clinical, radiographic, and histopathological findings to achieve an accurate diagnosis of odontogenic lesions.A sound understanding of their origin, etiopathogenesis, and biological behavior is crucial, as gross examination may at times be misleading. Establishing the correct diagnosis is fundamental for determining appropriate surgical management and preventing recurrence. Therefore, in cases of diagnostic uncertainty, histopathological evaluation of excisional specimens remains the most reliable and indispensable gold standard.

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Volume 8, Issue 6; November-December 2025; Page No 263-270

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Volume 8, Issue 6; November-December 2025; Page No 263-270