ISSN: 2573-8224

Case Report

Journal of Oral & Dental Health

# Bilateral Dentigerous Cysts in A Nonsyndromic Patient: A Diagnostic Dilemma Mimicking Odontogenic Keratocyst: A Case Report and Literature Review

**Dr. Prachi Bheke[1*], Dr. Shiva Kumar H. R.[1], Dr. Amarnath Upasi[1], Dr. Vinayak G. Naik[1], Dr. Chatura K. R.[2] and Dr. Kirthi Kumar Rai[1]**

[1]*Department of Oral and Maxillofacial Surgery, Bapuji Dental College and Hospital, Davangere, Karnataka, India*

[2]*Bhagavan Pathology Laboratory, Mysore, Karnataka, India*

**[*]Corresponding Author**
Prachi Bheke, Department of Oral and Maxillofacial Surgery, Bapuji Dental College and Hospital, Davangere, Karnataka, India.

**Submitted:** 2025, Aug 15; **Accepted:** 2025, Sep 11; **Published:** 2025, Sep 18

**Citation:** Bheke, P., Kumar, S. H. R., Upasi, A., Naik, V. G. and Chatura, K. R., et al. (2025). Bilateral Dentigerous Cysts in A Nonsyndromic Patient: A Diagnostic Dilemma Mimicking Odontogenic Keratocyst: A Case Report and Literature Review. *J Oral Dent Health, 9*(2), 01-06.

## Abstract

***Background:*** *Dentigerous cysts are common developmental odontogenic cysts, constituting approximately 24% of all jaw cysts. They typically present as solitary, unilocular radiolucencies associated with the crowns of unerupted teeth, most often involving mandibular third molars, maxillary canines, and premolars. While usually asymptomatic and unilateral, bilateral occurrences are exceedingly rare and often linked to syndromes such as cleidocranial dysplasia or mucopolysaccharidosis. The presence of bilateral dentigerous cysts in nonsyndromic individuals is exceptionally uncommon and may be misdiagnosed due to radiographic similarities with other cystic lesions, particularly odontogenic keratocysts (OKCs), thereby complicating the diagnostic and treatment approach.*

***Case Report:*** *We report a rare case of a nonsyndromic patient with incidentally detected bilateral dentigerous cysts associated with impacted mandibular third molars. The patient was asymptomatic, and panoramic radiographs revealed well-defined, unilocular radiolucent lesions surrounding the crowns of both impacted teeth. Due to the diagnostic ambiguity, particularly the overlap with OKC radiographic features, surgical enucleation was performed. Histopathological examination confirmed the diagnosis of dentigerous cysts, characterized by a non-keratinized stratified squamous epithelial lining and fibrous capsule with chronic inflammatory infiltrate. No clinical or systemic signs of an underlying syndrome were noted.*

***Conclusion:*** *This case emphasizes the need to include bilateral dentigerous cysts in the differential diagnosis of multiple jaw radiolucencies, even in nonsyndromic individuals. The diagnostic overlap with OKCs highlights the necessity of correlating radiographic findings with histopathological evaluation to ensure accurate diagnosis. Early identification and appropriate surgical intervention are vital to preventing complications such as infection, bone destruction, or recurrence. Given the rarity of such presentations, continued reporting and literature review are essential to enhance diagnostic clarity and guide effective clinical management.*

## 1. Introduction

Dentigerous cysts are the second most common type of odontogenic cysts, representing approximately 24% of all true jaw cysts, second only to radicular cysts [1]. These cysts develop around the crown of an unerupted or impacted tooth, most commonly the mandibular third molars, maxillary canines, and premolars [2]. Typically, solitary and unilateral, dentigerous cysts arise due to the accumulation of fluid between the reduced enamel epithelium and the crown of an unerupted tooth [3]. While often asymptomatic and incidentally discovered during routine radiographic examinations, they can present with symptoms like swelling, pain, and displacement of adjacent teeth in advanced stages [4].

Bilateral or multiple dentigerous cysts are rare and most frequently associated with syndromes such as cleidocranial dysplasia, Maroteaux-Lamy syndrome, or mucopolysaccharidosis [5]. The occurrence of bilateral dentigerous cysts in the absence of systemic disease or syndromic features is exceptionally rare, with fewer than 50 cases reported in the English literature to date [6]. Such rarity often leads to diagnostic challenges and under-reporting.

This case report highlights the occurrence of bilateral dentigerous cysts in a nonsyndromic patient, emphasizing the importance of comprehensive radiographic evaluation even in the absence of clinical symptoms. The discussion includes a detailed review of similar rare cases from existing literature to underscore the infrequency of such presentations and the importance of recognizing this pathology. Given their potential for significant bone destruction, early diagnosis and appropriate management are critical to preventing complications [7].

## 2. Case Report

A 16-year-old male patient presented to the Department of Oral and Maxillofacial Surgery at Bapuji Dental College & Hospital, Davangere, Karnataka, India with a primary complaint of pain and swelling in the lower left back teeth region since past 2 months. Extra-oral examination showed slight facial asymmetry with mild diffuse swelling over left lower one third of face region. Bilateral submandibular lymph nodes were palpable, tender and enlarged. No obvious abnormality was observed on systemic examination. Intra-oral examination showed absence of mandibular right second molar and mandibular and maxillary 3rd molar teeth on both (right and left) side of the jaw. Buccal vestibular obliteration with buccocortical expansion was seen in the left posterior teeth region (Figure 1).

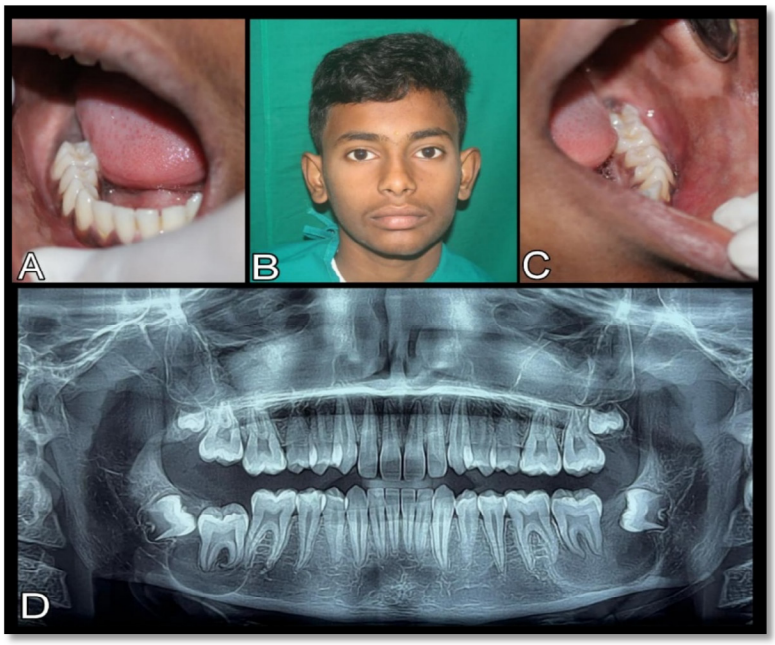


**Figure 1:** Preoperative profile, A.Intraoral right side, B.Extraoral frontal profile, C.Intaoral left side, D.Preoperative opg

Bilateral cyst enucleation with extraction of the impacted second and third molars and excisional biopsy were performed under general anesthesia. Bilateral surgery was performed with intraoral crest incisions distal to the first molars extending upto the anterior border of ramus. The incisions extended medially to the mesiobuccal area of the first molar, where oblique incisions extended to the vestibule. Triangular mucoperiosteal flap was reflected and bone windows were created to expose the cysts. The impacted teeth were carefully removed with the surrounding cystic lesion, ensuring the preservation of the inferior alveolar nerve (Figure 2).

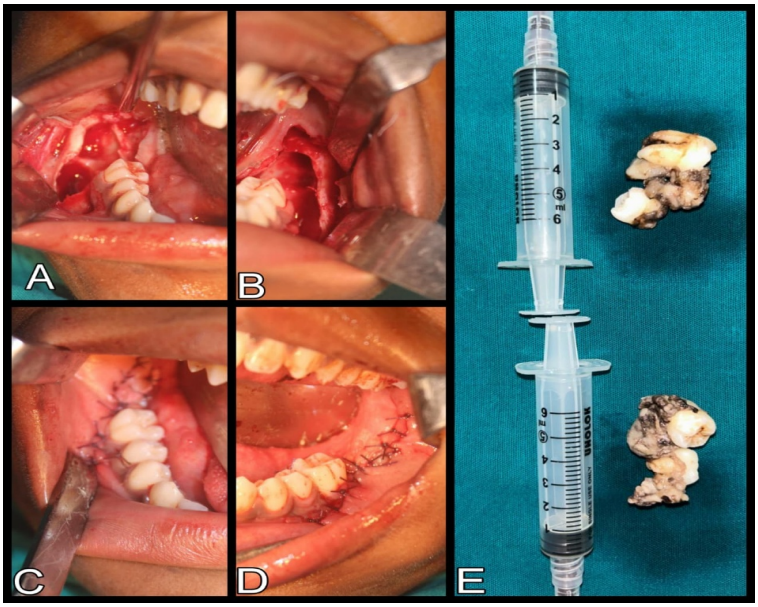


**Figure 2:** Intraoperative Profile, A.Enucleation of cystic lesion over right side, B.Enucleation of cystic lesion over left side, C.Closure over right side, D.Closure over left side, E.Cystic lesion specimens

Histological examination of the left-sided cyst revealed predominantly denuded or ulcerated lining epithelium with dense lymphoplasmacytic infiltration, proliferating capillaries, hemosiderin deposits, and a collagenized cyst wall. Focally preserved stratified squamous epithelium of variable thickness with elongated rete ridges was noted, along with small odontogenic epithelial rests, some showing continuity with the overlying epithelium. Uninflamed areas displayed thin non-keratinized epithelium, and focal Rushton bodies were identified. Fragments of bone were seen beneath the stroma, and no mucin-secreting cells were present. The right-sided cyst showed a partially preserved non-keratinizing squamous epithelial lining without parakeratosis, palisading basal cells, or surface corrugation. Focal areas exhibited elongated rete ridges and moderate lymphoplasmacytic infiltration, while other areas showed only two to three epithelial layers with luminal cuboidal to columnar cells. The stroma contained scattered odontogenic epithelial rests without mucous cells (Figure 3). The case was diagnosed as Bilateral Dentigerous Cysts.

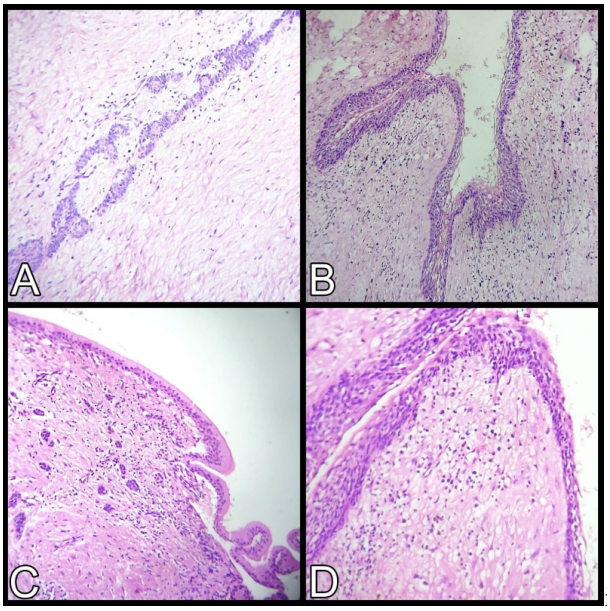


**Figure 3:** Histopathology, A.Odontogenic epithelial rests in left cyst, B.Non-keratinizing squamous epithelial lining on left side, C.Scattered odontogenic epithelial rests with luminal cuboidal to columnar cells in right cyst, D.Non-keratinizing squamous epithelial lining on right side

Patient was kept on five days of parenteral antibiotics, analgesics and anti-inflammatory drug. Healing was satisfactory with no complications at six months follow up (Figure 4). Patient has been advised for periodic regular follow up visit further.

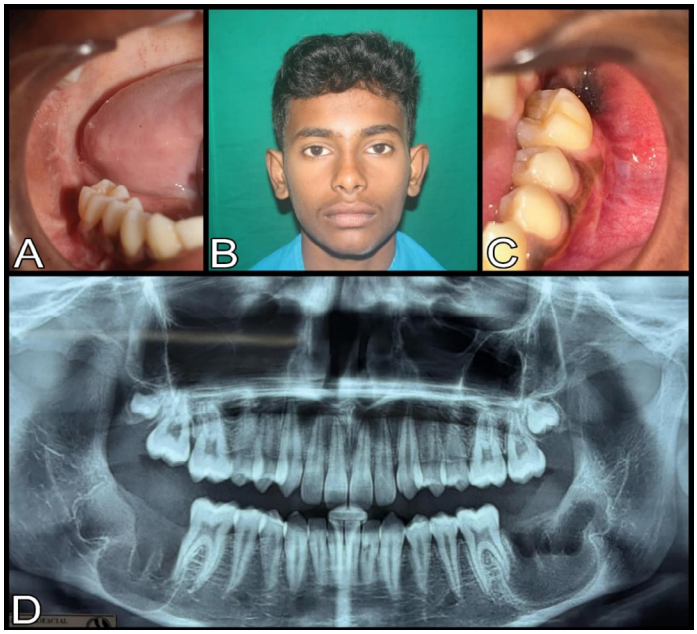


**Figure 4:** One-Month Follow-Up, A.Healing over right side, B.Extraoral frontal profile, C.Healing over left side, D.One-month postoperative opg

## 3. Discussion

Odontogenic cystic lesions often pose diagnostic challenges due to overlapping clinical, radiographic, and histopathological features. Among these, dentigerous cysts and odontogenic keratocysts (OKCs) are particularly notable for their similarity in presentation, which can lead to misdiagnosis and suboptimal treatment outcomes.

Dentigerous cysts are developmental in origin and typically present as well-defined unilocular radiolucencies associated with the crowns of unerupted or impacted teeth. Most commonly found around mandibular third molars, maxillary canines, and premolars, they arise from the accumulation of fluid between the reduced enamel epithelium and the tooth crown. In contrast, OKCs, although also radiolucent and often pericoronal, are more aggressive, have a higher recurrence rate, and a potential for neoplastic transformation.

Radiographically, both lesions can appear strikingly similar. Aldelaimi et al. reported a case where an OKC was initially misdiagnosed as a dentigerous cyst based solely on imaging, leading to an inadequate treatment plan that was later revised following histopathological evaluation [8]. This case underlines the diagnostic ambiguity between these two lesions and the potential clinical consequences of relying solely on radiographic features.

Advanced imaging modalities have been proposed to help differentiate these entities. According to Wang et al., computed tomography (CT) and magnetic resonance imaging (MRI) can identify subtle features—such as buccolingual expansion and scalloped margins more commonly seen in OKCs—that may not be visible in standard radiographs [9]. Pauwels et al. also stressed the value of cone-beam CT (CBCT), which provides enhanced views of cortical bone perforation and internal architecture [10]. However, they cautioned that even CBCT cannot offer definitive diagnoses, reinforcing the necessity for histopathological confirmation.

Distinguishing a dentigerous cyst in place of odontogenic keratocyst (OKC) can be diagnostically challenging, particularly in inflamed specimens where classical epithelial features may be obscured. However, certain histopathological hallmarks aid in their differentiation. In the present case, both cysts exhibited a non-keratinized stratified squamous epithelial lining without basal cell palisading or surface corrugation—features that are typically characteristic of OKCs. Crucially, the right-sided cyst showed areas lined by reduced enamel epithelium, a defining feature of dentigerous cysts, which originate from the dental follicle. Additionally, numerous odontogenic epithelial rests were seen scattered within the fibrous cyst wall of both lesions—another common finding in dentigerous cysts but generally absent or less prominent in OKCs [11].

In contrast, OKCs are typically lined by a parakeratinized stratified squamous epithelium with a corrugated surface and a palisaded, hyperchromatic basal layer. They may also exhibit budding into the underlying stroma and contain satellite or daughter cysts, reflecting their more aggressive and recurrent nature. Inflammatory changes can cause OKCs to temporarily lose these features,

leading to diagnostic overlap [12]. However, in this case, the lack of keratinization, absence of palisading basal cells, and presence of reduced enamel epithelium and epithelial rests, along with the radiological context of pericoronal unilocular radiolucencies, collectively supported the diagnosis of bilateral dentigerous cysts.

Thus, while the histological similarities between these two entities can create diagnostic ambiguity—particularly in the setting of inflammation—a careful evaluation of epithelial morphology, cyst wall components, and correlation with clinical and radiographic findings remains essential for accurate diagnosis and appropriate treatment planning.

Misdiagnosis has critical implications for treatment. Dentigerous cysts are usually treated conservatively by enucleation, often with preservation of the associated tooth. In contrast, OKCs require more aggressive approaches such as peripheral ostectomy or resection to prevent recurrence. Kavitha et al. detailed a case where a lesion initially treated as a dentigerous cyst recurred multiple times over five years before being correctly diagnosed as an OKC [13]. This illustrates how an incorrect diagnosis can lead to prolonged morbidity and repeated interventions.

Adding to the diagnostic complexity is the occurrence of atypical presentations of dentigerous cysts. Narayana et al. reported a case involving melanin pigmentation in a dentigerous cyst, an uncommon feature that could mimic more ominous lesions like melanotic neoplasms [14]. Such findings highlight the necessity of complete histopathological evaluation, even in cases that appear straightforward radiographically.

Furthermore, bilateral or multiple dentigerous cysts, especially in nonsyndromic patients, are rare and can confound diagnosis. Sindi et al. described bilateral mandibular cysts in a 44-year-old nonsyndromic male, identified incidentally, with classic histology confirming their benign nature [6]. Bang et al. similarly reported bilateral cysts in a 55-year-old female, citing only 48 comparable cases reported over 75 years [2]. These authors emphasized that such occurrences are not always linked to syndromic conditions and highlighted the value of panoramic radiography in early detection.

Vasiapphan et al. presented a case involving bilateral mandibular cysts in a 27-year-old male with horizontally impacted molars, reinforcing the need for routine radiographic assessment in patients with multiple impacted teeth [4]. Pediatric presentations add further complexity. Tamgadge et al. described bilateral dentigerous cysts in a 10-year-old child, a demographic where such cases are exceedingly rare[1]. Similarly, Pant et al. and Sethi et al. reported inflammatory follicular cysts in children, mimicking dentigerous cysts but arising from periapical infections of overlying primary teeth [3,5]. They emphasized the importance of distinguishing inflammatory from developmental origins, as treatment and prognosis may differ significantly.

While developmental dentigerous cysts generally have excellent prognoses post-enucleation, inflammatory variants may require additional management of the source infection. OKCs, due to their potential for recurrence and aggressive growth, require close long-term follow-up. Shear and Speight recommend periodic imaging for at least 10 years following treatment of OKCs to monitor for recurrence [15].

In the present case, bilateral mandibular cysts were incidentally discovered in a nonsyndromic patient and managed successfully with enucleation. Radiographic appearance initially suggested dentigerous cysts, which was later confirmed through histological evaluation. The absence of syndromic signs did not preclude bilateral presentation, reinforcing the need for comprehensive evaluation.

The diagnostic overlap between dentigerous cysts and OKCs presents significant clinical challenges. Radiographic similarity can mislead diagnosis and alter treatment decisions. A multidisciplinary approach, incorporating clinical findings, advanced imaging, and definitive histopathology, is essential to ensure accurate diagnosis and optimal patient care. Clinicians must remain vigilant, particularly in atypical or bilateral cases, and adopt a conservative diagnostic approach with an appropriately aggressive treatment plan when necessary. Future research into molecular and genetic markers may offer more precise tools for differentiating these lesions and further improve outcomes.

## 4. Conclusion

Bilateral dentigerous cysts in nonsyndromic patients are exceedingly rare, with limited cases reported in the literature. This uncommon presentation increases the risk of underdiagnosis or misdiagnosis, especially in asymptomatic individuals. The radiographic resemblance of dentigerous cysts to other odontogenic lesions, particularly odontogenic keratocysts, further complicates diagnosis and underscores the importance of a multidisciplinary approach. Our case highlights the critical role of comprehensive radiographic evaluation and the necessity of histopathological confirmation to differentiate between similarly presenting cystic lesions. Early and accurate diagnosis enables appropriate treatment planning, prevents unnecessary interventions, and minimizes the risk of recurrence. Enucleation remains the treatment of choice with generally favorable outcomes. Continued reporting of such rare presentations will contribute to improved recognition, diagnostic clarity, and clinical management of odontogenic cysts.

## Declarations

**Ethics Approval:** Not Applicable
**Consent to Participation:** Obtained
**Consent for Publication:** Obtained
**Availability of Data and Material:** Not Applicable
**Competing Interests:** The Authors Do Not Have Conflicts of Interest.
**Funding:** No Financial Support Was Provided for This Work

## Authors' Contributions

**Acknowledgements:** Not Applicable
**Authors' Information (Optional):** Not Applicable

## References

1. Tamgadge, A., Tamgadge, S., Bhatt, D., Bhalerao, S., Pereira, T., & Padhye, M. (2011). Bilateral dentigerous cyst in a non-syndromic patient: Report of an unusual case with review of the literature. *Journal of Oral and Maxillofacial Pathology, 15*(1), 91-95.
2. Bang, K. O., Yadav, S. R., Shenoi, R., Budhraja, N., & Ingole, P. (2021). Bilateral non-syndromic dentigerous cyst–rare case report and literature review. *Advances in Oral and Maxillofacial Surgery, 4,* 100043.
3. Pant, B., Carvalho, K., Dhupar, A., & Spadigam, A. (2019). Bilateral nonsyndromic dentigerous cyst in a 10-year-old child: A case report and literature review. *International Journal of Applied and Basic Medical Research, 9*(1), 58-61.
4. Vasiapphan, H., Christopher, P. J., Kengasubbiah, S., Shenoy, V., Kumar, S., & Paranthaman, A. (2018). Bilateral dentigerous cyst in impacted mandibular third molars: a case report. *Cureus, 10*(12).
5. Sethi, A., Shetty, D. C., Tandon, A., & Afroz, N. (2020). Bilateral Nonsyndromic Cystic Lesions Involving Impacted Teeth: Nomenclature and Diagnostic Protocol. *International Journal of Clinical Pediatric Dentistry, 13*(4), 429.
6. Sindi, A. M. (2019). Bilateral mandibular dentigerous cysts presenting as an incidental finding: A case report. *The American Journal of Case Reports, 20,* 1148.
7. Holla, V. A., Chatra, L., Shenai, P., Rao, P. K., Veena, K. M., & Prabhu, R. V. (2012). Bilateral inflammatory cysts of the jaw: report of an unusual case. *Imaging science in dentistry, 42*(2), 105.
8. Aldelaimi, T.N. et al. (2023). Odontogenic Keratocyst Mimicking Dentigerous Cyst, FOMM Journal.
9. Wang, C. et al. (2018). Imaging Features of Odontogenic Keratocysts: A Pictorial Review, Insights Imaging.
10. Pauwels, R. et al. Radiologic Differentiation Between OKC and Dentigerous Cyst, Radiopaedia.
11. Austin, R. P., & Nelson, B. L. (2021). Sine qua non: dentigerous cyst. Head and neck pathology, 15(4), 1261-1264.
12. Chaudhary, S., Sinha, A., Barua, P., & Mallikarjuna, R. (2013). Keratocystic odontogenic tumour (KCOT) misdiagnosed as a dentigerous cyst. *Case Reports, 2013*, bcr2012008741.
13. Kavitha, B. et al. Recurrent OKC Misdiagnosed as Dentigerous Cyst: A Case Series.
14. Narayana, S. et al. (2022). Melanin Pigmentation in Dentigerous Cyst: A Rare Case, PMC.
15. Shear, M., & Speight, P. M. (2008). Cysts of the oral and maxillofacial regions. John Wiley & Sons.