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Orthokeratinized Odontogenic Cyst of the Posterior Mandible with Lingual Cortical Expansion: A CBCT-Based Case Report

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Abstract

An orthokeratinized odontogenic cyst (OOC) is an uncommon developmental odontogenic cyst that predominantly affects the posterior mandible. It is considered a distinct clinicopathological entity because of its characteristic histopathological features, less aggressive biological behaviour, and low potential for recurrence. Clinically, OOC is often asymptomatic and may remain undetected until discovered during routine radiographic examination. Radiographically, it commonly appears as a well-defined unilocular radiolucency, frequently associated with impacted teeth, thereby mimicking other odontogenic cysts and tumours. Advanced imaging modalities such as cone beam computed tomography (CBCT) are valuable for assessing the exact extent of the lesion, cortical expansion, cortical plate thinning, and the lesion's relationship with adjacent anatomical structures.

The present case describes a 34-year-old male patient with a slowly progressive painless swelling involving the right posterior mandible. Radiographic and CBCT evaluation revealed a well-defined unilocular radiolucency associated with an impacted mandibular third molar, along with significant thinning and expansion of the lingual cortical plate. Histopathological

examination following surgical enucleation confirmed the diagnosis of OOC.

This case emphasises the importance of correlating clinical, radiographic, CBCT, and histopathological findings for accurate diagnosis of OOC. CBCT played a significant role in demonstrating the extent of cortical involvement and guiding appropriate surgical management, contributing to a favourable postoperative outcome and prognosis.

Keywords: CBCT, Mandible, Odontogenic keratocyst, OPG, Orthokeratinized odontogenic cyst.

Introduction

Orthokeratinized odontogenic cyst (OOC) is an uncommon developmental odontogenic cyst characterised by a distinct orthokeratinized epithelial lining and relatively less aggressive biological behaviour when compared with odontogenic keratocyst (OKC) [1,2]. OOC predominantly occurs in the second to fifth decades of life and is more frequently observed in the posterior mandible, with a male predilection[3,4]. Histogenetically, it is believed to arise from the remnants of the dental lamina or from the basal cell layer of the oral epithelium [2]. Clinically, OOC usually presents as a slow-growing, asymptomatic lesion, although larger lesions may cause cortical expansion, swelling, and occasional pain [3].

Radiographically, OOC usually presents as a well-defined unilocular radiolucency and is commonly associated with impacted mandibular third molars, often resembling dentigerous cysts or odontogenic keratocysts [4,5]. Because of these overlapping radiographic features, diagnosis based solely on conventional imaging can be challenging. Cone beam computed tomography (CBCT) plays an important role in evaluating the exact extent of the lesion, degree of cortical expansion and thinning, and its proximity to adjacent anatomical structures, thereby aiding in accurate diagnosis and treatment planning. Histopathological examination remains essential for definitive diagnosis, as OOC characteristically exhibits a uniformly orthokeratinized stratified squamous epithelial lining with a prominent granular cell layer and a relatively flat epithelial-connective tissue interface [2,6].

The present report describes a case of OOC involving the right posterior mandible in association with an impacted mandibular third molar. The case is presented to highlight the clinical findings, radiographic and CBCT characteristics, histopathological features, and conservative surgical management of this uncommon odontogenic cyst.

Case Presentation

A 34-year-old male patient presented to the Department of Oral Medicine and Radiology with a mild swelling in the lower right posterior region of the jaw, persisting for approximately two years. The swelling exhibited a gradual onset and progressive enlargement. There was no associated pain, paresthesia, discharge, or additional symptoms. The patient denied any history of trauma to the affected area. Medical, dental, familial, and personal histories were non-contributory.

During the extraoral examination, a mild swelling was observed over the right posterior region of the mandible (Figures 1 and 2). The swelling extended anteroposteriorly from the angle of the mandible to about 2 cm in front of it, and superoinferiorly from the lower border of the mandible to roughly 2 cm below, measuring approximately 2 × 2 cm. The skin over the swelling appeared normal in colour and texture, with no signs of redness, ulceration, sinus tract, or discharge. On palpation, these findings were confirmed: the swelling was hard, nontender, and noncompressible, nonreducible, and nonfluctuant. There was no increase in local temperature. The margins were indistinct, and the surface felt smooth. The swelling was firmly attached to the

underlying bone, but not to the overlying skin, suggesting it originated within the bone itself.

During the intraoral examination, tooth 48 was found to be clinically missing. The mucosa covering the area looked healthy, with no signs of ulceration, discolouration, or sinus tract formation. On palpation, there was a localised, mild, and non-tender expansion of the lingual cortical plate in the region of tooth 48 (Figure 3).

Radiographic Investigation

The orthopantomogram (OPG) showed a well-circumscribed, unilocular radiolucent lesion with a thin, corticated border. The lesion extended from the distal root of tooth 46, extending up to the ascending ramus and surrounding the impacted tooth 48. There was no sign of root resorption in the adjacent teeth (see Figure 4).

Cone Beam Computed Tomography (CBCT) axial images (Figures 5–8) revealed significant thinning and expansion of the lingual cortical plate. The impacted tooth 48 shows lingual inclination.

Based on the clinical and radiographic findings, a preliminary diagnosis of odontogenic keratocyst was made.

Surgical Management

The lesion was treated conservatively with complete enucleation under local anaesthesia, along with the extraction of teeth 48 and 47 (Figures 9 and 10). The removed specimen was a single cystic soft-tissue mass measuring approximately 5 × 2 cm. It had an off-white to brown appearance, with mottled areas due to scattered bleeding. On palpation, the texture ranged from soft to firm, indicating variations within the tissue. The specimen was promptly preserved and submitted for histopathological examination (Figures 11 and 12).

Histopathological Examination

Microscopic examination showed a cystic lesion lined by a uniformly well-keratinised epithelium, supported by mature connective tissue. The cystic lining was 7–9 cells thick, with a well-developed granular layer and varying thickness of orthokeratinisation. The supporting stroma contained patches of dystrophic calcification, areas of haemorrhage and hemosiderin pigmentation, and a few regions with condensed cholesterol clefts and giant cells. Some areas of the cystic epithelium were hyperplastic. Based on these histopathological features, a final diagnosis of orthokeratinized odontogenic cyst was made.

Follow Up

The patient was followed up after surgery and again at six months post-operatively. Post-operative OPG and CBCT scans were taken (Figures 13–16).

Discussion

When evaluating well-defined, unilocular radiolucent lesions associated with impacted teeth, especially in the posterior mandibular region, an orthokeratinized odontogenic cyst (OOC) should be included in the differential diagnosis. OOC can closely resemble dentigerous cysts and odontogenic keratocysts (OKCs) both clinically and radiographically, making accurate diagnosis challenging. Careful correlation of clinical, radiographic, and histopathological findings is essential [8]. In the present case, the lesion was found to be associated with an impacted mandibular third molar and showed radiographic features similar to those of OKC, underscoring the need for histopathological confirmation. OOC was once thought to be just a variant of OKC, but research now shows it's actually a separate condition. It tends to be less aggressive and much less likely to recur after treatment [2,4]. Under the microscope, OOC has a very regular orthokeratinized squamous cell lining, a

distinct granular layer, and a relatively flat interface between the epithelium and connective tissue. In contrast, OKC usually has a wavy, parakeratinized lining and palisaded basal cells, and exhibits greater cellular growth [2,4]. These microscopic differences are important because they help explain why OOC behaves less aggressively than OKC.

Immunohistochemical studies have helped further clarify the biological differences between OOC and OKC. Da Silva and colleagues found that OKC tends to grow more aggressively and shows higher cell proliferation than OOC, which supports why OOC doesn't recur as often [7]. Likewise, Thosaporn et al. used the IPO-38 marker and found OOC has much lower cell proliferation than OKC and ameloblastoma [8]. Dong and colleagues, in a large study of 61 OOC cases, also found much lower levels of markers such as Ki-67 and p63, further confirming that OOC is a more benign lesion [9].

On X-rays, OOC usually appears as a unilocular radiolucency with clear, defined borders, and it's often found with impacted teeth [2,9]. MacDonald-Jankowski's review of the literature found that over 90% of OOC cases were unilocular, and about 70% were linked to unerupted teeth [2,9]. The current case matched these findings. However, regular X-rays might not always show just how much of the jawbone is affected or the details of the lesion's internal structure.

In the present case, CBCT imaging was especially helpful. While the orthopantomogram gave a general idea of the lesion's size, CBCT offered a detailed 3D view, revealing severe thinning and expansion of the lingual cortical of the jawbone as well as the tilted position of the impacted mandibular third molar. Knowing how much the bone was expanded is important because extreme thinning can increase the risk of the bone breaking and can affect how the surgery is planned. CBCT also helped clearly show the lesion's edges and its relationship to nearby structures, making treatment planning more accurate.

The preferred treatment for OOC is conservative surgical removal, primarily because the cyst rarely recurs and the overall outlook is good [6]. In this case, complete enucleation was performed along with removal of the impacted tooth, and the procedure proceeded successfully, indicating a positive outcome. Follow-up visits showed that the area had healed well and that the bone had begun to regenerate, with no signs of the cyst returning. Catching OOC early, along with thorough imaging and confirming the diagnosis under the microscope, is key to making sure patients do well in the long run.

Conclusion

Orthokeratinized odontogenic cyst is a distinct clinical and histopathological entity that requires careful differentiation from other odontogenic cysts, given its relatively benign behaviour and lower recurrence potential compared with parakeratinized OKC. This case highlights the importance of correlating clinical findings with radiographic and histopathological features to arrive at an accurate diagnosis. Despite its deceptively aggressive radiographic appearance, OOC typically follows a favourable course post-enucleation when appropriately managed. Advancements in molecular diagnostics may further refine our understanding of its pathogenesis and support more individualised treatment protocols in the future.

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Legend Figures

Figure 1:

Figure 2:

Figure 3:

Figure 4: Unilocular radiolucency extending from the distal root of 46 to the ramus of the mandible with impacted 48.

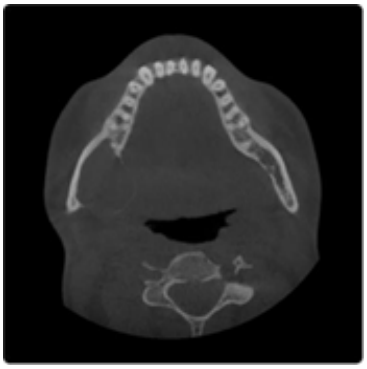

Figure 5:

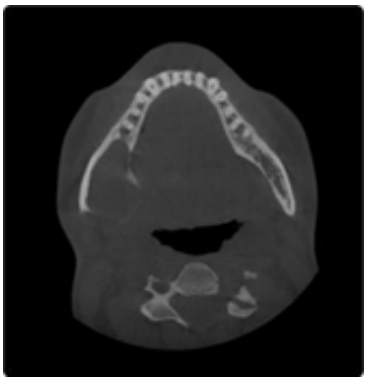

Figure 6:

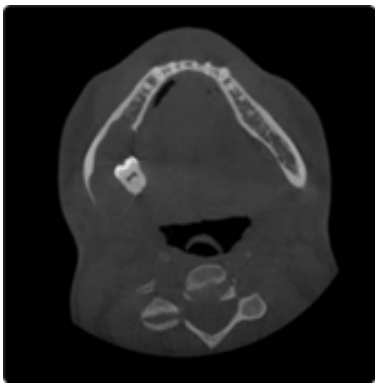

Figure 7:

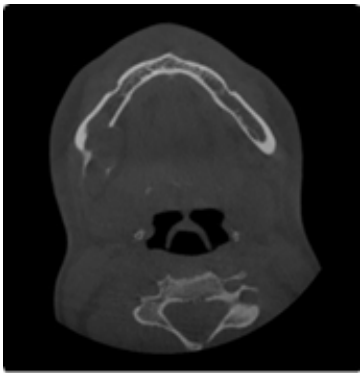

Figure 8:

Figure 9:

Figure 10:

Figure 11: Soft tissue specimen measures approx 4mm Supero-inferiorly.

Figure 12: Soft tissue specimen measures approx 3mm Antero-posteriorly

Figure 13: Postoperative OPG reveals extraction of 47 & 48, with preservation of the inferior border of the mandible.

Figure 14: Post-operative OPG (after 6 months) shows regeneration of bone noticed at the surgical site.

CBCT Images -Follow Up (6 Months)

Figure 15: (Sagittal Section) - Shows satisfactory osseous regeneration at the surgical site.

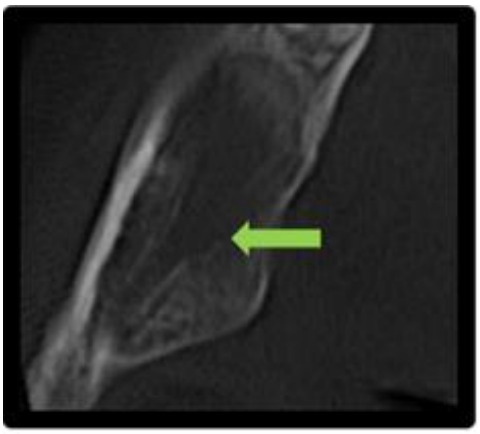

Figure 16: (Axial Section)- Thinned out lingual cortical bone noticed in the 47 & 48 region.