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Bilateral Mandibular Orthokeratinised Odontogenic Cysts in a 14-year-old Patient

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Abstract

Orthokeratinised odontogenic cyst (OOC) is a developmental odontogenic cyst. It was previously classified as an orthokeratinised variant of the odontogenic keratocyst (OKC). However, due to the difference in histopathological features and clinical behaviour, it is now considered a distinct entity. OOCs are uncommon findings and the presence of bilateral lesions are even rarer. This article presents an unusual case of a 14-year-old male patient who was treated for bilateral OOCs in the posterior mandible, both of which were incidental findings.

Keywords: Orthokeratinised • Odontogenic cysts • keratocyst • Oral Health

Introduction

Odontogenic cysts are pathological cavities arising in tooth-bearing regions of the maxilla and mandible. They are frequently lined with epithelium and can be of inflammatory or developmental origin. If untreated they have potential to cause bony destruction, resorption or displacement of adjacent teeth [1]. The orthokeratinised odontogenic cyst (OOC) is an uncommon type of developmental odontogenic cyst, arising within the cell rests of the dental lamina [2]. OOCs were previously described as an orthokeratinised variant of the odontogenic keratocyst (OKC). They were first defined as a separate clinicopathological entity from OKCs by Wright JM due to the lower rates of recurrence and limited growth potential [3]. This is consistent with the most recent Classification of Head and Neck Tumours published by World Health Organisation in 2017, which recognises OOCs as a distinct odontogenic cyst from OKCs [4]. OOCs constitute to approximately 1% of all odontogenic cysts [4].

OOCs usually occur in the 3rd to 4th decade and have a predilection to males, with a ratio of 2–2.5:1 [3]. Clinically, OOCs present as a slow growing swelling of the jaw. In many cases, these cysts are asymptomatic and may be incidental findings. However, they can cause cortical expansion resulting in swelling and occasionally pain. The most commonly affected area is the molar and ramus region of the mandible. OOCs have been associated with impacted teeth in 46.5 - 75% of cases. They appear radiographically as a well-defined, unilocular or multilocular radiolucency [2,3]. On histological comparison, OOCs show orthokeratinisation and a prominent granular cell layer, whereas OKCs exhibit parakeratinisation. In addition, OKCs often display corrugation in the surface keratin layers [4]. 5% of OKCs present with multiple cystic lesions and are associated with naevoid basal cell carcinoma syndrome (NBCCS) [4]. However, multiple OOCs are a rare phenomenon and no association has been reported with NBCCS [5]. In this paper, we present a rare case of bilateral OOCs incidentally found in a 14-year-old male patient.

Case Presentation

A 14-year-old patient was referred by his general dental practitioner to Ashford and St Peter's Hospital originally for an orthodontic assessment of his lingually impacted LR7. The patient had no symptoms and his medical history was unremarkable. On examination, the patient had a Class I incisor relationship on a Class I skeletal base with average vertical proportions. Both arches were well aligned and both mandibular second molars were partially erupted. There was no evidence of any buccal-lingual cortical expansion. A dental panoramic radiograph (Figure 1) revealed an incidental finding of a well-defined unilocular radiolucency associated with the unerupted LL8 extending to the distal aspect of the LL7. The LL7 was positive to vitality testing and there was no associated paraesthesia.

A cone beam computed tomography (CBCT) scan was subsequently taken of the LL8 region and is shown in Figure 2. The scan confirmed a well-defined and corticated radiolucency extending from the mesial cemento-enamel junction of the LL8 to the distal surface of the LL7, measuring approximately 16mm in size. There were thinning of both lingual and buccal cortices observed but no evidence of resorption to the LL7. The inferior aspect of the radiolucency was in close proximity to inferior dentoalveolar (ID) canal, with no bony separation. Based on the initial clinical findings and radiographic investigations, the differential diagnoses of the lesion were either a dentigerous or paradental cyst. An odontogenic keratocyst or unicystic ameloblastoma were also considered as other possible differentials, however these were

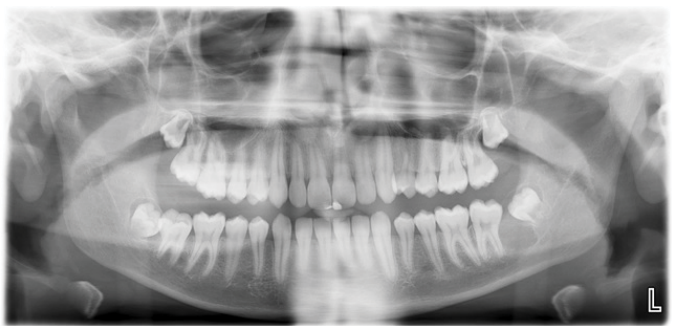

Figure 1. Panoramic radiograph revealing an impacted LR7 and a well-defined, unilocular radiolucency associated with the LL8.

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less likely. Following the discussion of treatment options with the patient and parents, arrangements were made for the patient to have the surgical removal of the unerupted LL8, enucleation of the associated cyst in the left mandible and removal of the lingually inclined LR7 under general anaesthesia.

A three-sided buccal mucoperiosteal flap was raised in the LL78 and LR7 region. The LL8 crown was sectioned and roots were elevated with intact apices. The cyst in the left mandible was enucleated with complete curettage and no damage to the ID nerve was observed. The LR7 crown was sectioned and roots elevated. On examination of the socket, a similar but smaller cystic lesion was incidentally observed on the lingual aspect of the LR8 and this was also enucleated. The unerupted LR8 was left *in situ*. The clinical appearance of both cystic lesions were indicative of odontogenic keratocysts. The specimens were placed in formalin and separately sent for histopathological evaluation. The flap was repositioned with 3-0 vicryl rapide sutures. There were no complications reported in the immediate post-operative healing period. The patient is to be reviewed in 6 months' time to ensure bony infill at the surgical sites and assess for any signs of recurrence.

The histopathological analysis (Figure 3) confirmed that both cystic lesions had similar appearances, containing yellow cheesy materials when sliced. The cyst in the left mandible measured 16 × 12 mm and that in the right mandible was 6 × 6 mm. Both cysts were lined with a fairly uniform layer of orthokeratinising squamous epithelium of around 7-12 cells thickness. There was a prominent granular layer throughout with surface orthokeratin. A thin layer of corrugated parakeratin was not seen. The basal layer of the epithelium was not particularly prominent or palisaded. The cyst contained orthokeratin. The wall of the cyst was fibrotic and occasionally myxoid, showing only very focal mild lymphocytic inflammation. These histopathological features were consistent with a diagnosis of bilateral orthokeratinised odontogenic cysts.

Discussion

A systematic review carried out by MacDonald-Jankowski DS reported that 41% of OOC cases were discovered as incidental findings and 68% were associated with un-erupted teeth. These results are suggestive that many OOCs develop during adolescence at the time of third molar formation. However, the lesions may be noticed later due to the delayed presentation of symptoms [6]. With both cystic lesions incidental findings in this case, it allowed for the early presentation of these cysts for this patient in his 2nd decade. The appropriate management for the patient was carried out based on the findings at those specific times along the treatment pathway. Early presentation of cystic lesions allows for less complex surgery and a better long-term prognostic outcome. It allows for the prevention of any other potential complications that may have arisen if the cysts were not observed, such as bony destruction, swelling, purulent discharge and pain.

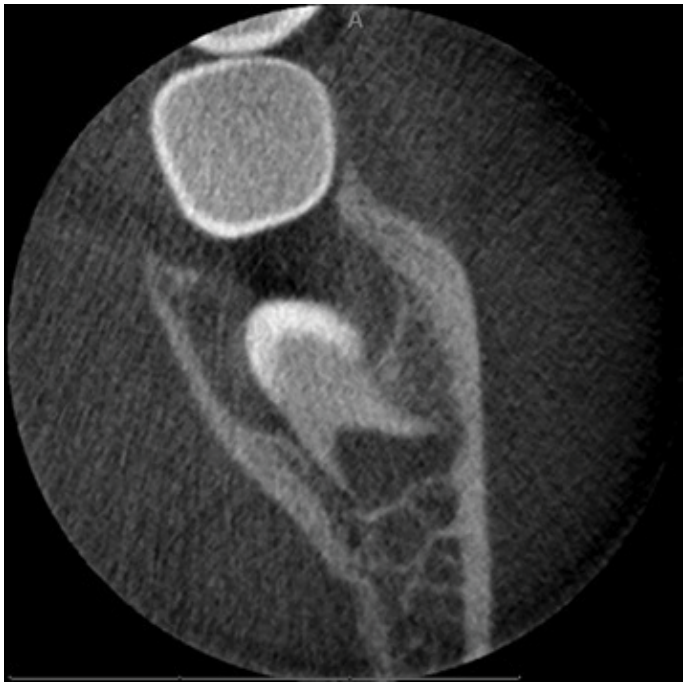

Figure 2. Cone beam computed tomography scan axial view showing a well-defined, corticated radiolucency extending from the cemento-enamel junction of the LL8.

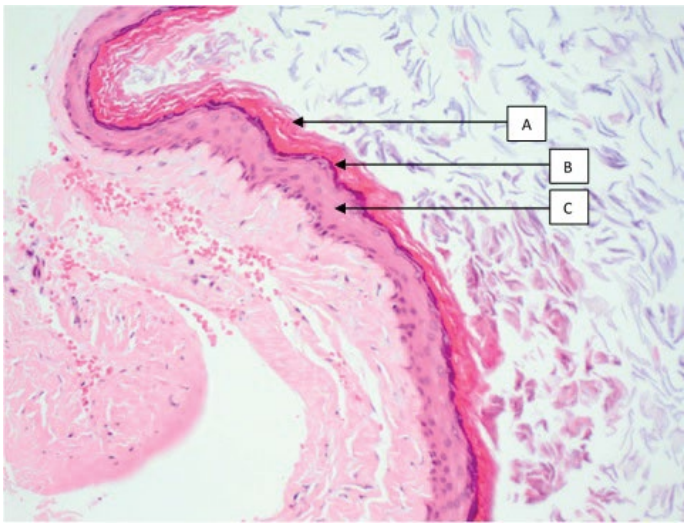

Figure 3. Histological appearance of the cyst at X20 magnification, with annotation showing the orthokeratinised squamous epithelium (A), granular layer with surface orthokeratin (B) and basal layer (C).

There is no reported association of OOCs with root resorption, however displacement of neighbouring teeth has been observed [2]. The cyst location in the right mandible is a likely factor in the lingual displacement of the LR7, which was the reason for referral from the patient's general dental practitioner in the first instance. In this case report, the differential, provisional and definitive diagnoses were all different. This underlines the difficulties in diagnosing cases with multiple OOCs, with the clinical presentation being similar to other odontogenic lesions such as OKCs and dentigerous cysts. Furthermore, it emphasises the importance of histopathological analysis to confirm the type of cyst and in turn, determine prognostic factors including the risk of recurrence.

OOCs and OKCs are similar in the age of occurrence, sex and site of predilection. Though, they do differ in biological activity. OKCs are highly aggressive and have higher rates of recurrence compared to other odontogenic cysts. The recurrence rate in OKCs is reported as 42.6% compared with 2.2% for OOCs [7]. OKCs more commonly occur at multiple sites and can be associated with NBCCS. However, there is no evidence suggesting an association of OOCs with NBCCS [7]. Macdonald-Jankowski DS concluded that although there is a low risk of recurrence in OOCs, there is a lack of continuing follow-up in cases showing long-term prognostic outcomes. In addition, there is insufficient clinical and radiographic information of OOCs on initial presentation [6]. This stresses the importance of long-term follow up for patients to assess for recurrence.

In an extensive review of current literature, there have only been a few cases with reported bilateral OOCs, as summarised in Table 1 [5,8-13]. These cases further highlight links to male predominance and association with impacted teeth, in particular mandibular third molars. All cases were treated conservatively with either marsupialisation, enucleation or curettage and had no reported recurrences. The age of presentation of these cases ranged from 17-35 years, with the younger cohort of patients largely presenting with no or mild symptoms. To the best of our knowledge from the review of literature relating to OOCs, this case report describes the youngest patient to be treated with bilateral lesions.

Table 1. A summary of cases with reported bilateral orthokeratinised odontogenic cysts.

Authors	Age	Gender	Symptoms	Number of Cysts	Location	Treatment	Associated with Un-erupted Teeth	Recurrence
Pereira PAC , et al. (2012) [9]	23	Female	Asymptomatic	2	Bilateral posterior mandible	Enucleation of both cysts	Yes (impacted lower third molars)	No (27 months follow-up)
Premalatha , et al. (2012) [11]	35	Male	Swelling, dull intermittent pain & restricted mouth opening	2	Bilateral posterior mandible	Enucleation of 1 cyst and marsupialisation of 1 cyst	No	Not known
Pimpalkar , et al. (2014) [8]	23	Male	Intra-oral intermittent watery discharge & sour taste	2	Bilateral posterior mandible	Enucleation of both cysts	Yes (impacted lower third molars)	Not known
Cheng , et al. (2015) [10]	23	Male	Swelling & discomfort	4	Bilateral posterior maxilla and mandible	Curettage of 3 cysts and marsupialisation, followed by curettage of 1 cyst	Yes (all four impacted third molars)	No (14 months follow-up)
Crane , et al. (2019) [5]	23	Male	Asymptomatic	3	Bilateral posterior mandible and left posterior maxilla	Curettage of all 3 cysts	2 out of 3 (impacted upper left third molar and lower left second and third molars)	No (48 months follow-up)
Crane , et al. (2019) [5]	20	Male	Asymptomatic	2	Bilateral posterior maxilla	Enucleation of both cysts	Yes (impacted upper third molars)	No (24 months follow-up)
Alhumaidan , et al. (2019) [12]	19	Male	Asymptomatic	2	Bilateral posterior mandible	Enucleation of both cysts	Yes (impacted lower third molars)	No (4 months follow-up)
Lucamba , et al. (2020) [13]	17	Male	Asymptomatic	2	Bilateral posterior mandible	Enucleation of 1 cyst and marsupialisation of 1 cyst	No	No (4 months follow-up)

Conclusion

In summary, the presentation of bilateral OOCs is a rare occurrence. Clinically and radiographically they may appear to be other lesions such as OKCs or dentigerous cysts. Histopathological investigation is essential to distinguish between other differential diagnoses and in turn understand the risk of recurrence. Early presentation allows for less complex surgery and better long-term prognosis.

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