



CASE REPORT

Bilateral dentigerous cyst in suprarenal tumor child: a case report

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Abstract

Bilateral dentigerous cyst in patients with no syndrome diagnosis is rare. Only 20 cases are published, to our knowledge. Here, a 6 years old child with a previous adrenal tumor diagnosis at 2yo and surgically treated at 4yo presented at our outpatient clinic in a quaternary children's hospital with a painless mandibular bone expansion on both sides. Physical and tomographic assessments suggested odontogenic cysts. Due to continuous hormone therapy with hydrocortisone and fludrocortisone, excisional biopsies were performed. Teeth associated with the cysts were removed to avoid post-operative infections. The patient had an uneventful follow-up and no recurrence signs. This case report reveals a potent focus of oral infections in an immunosuppressed patient. In these cases, the multidisciplinary approach is necessary.

Keywords: dentigerous cyst; adrenal adenoma; osteolytic lesion; bone cyst.

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Introduction

Dentigerous cysts (DC) are the second most common odontogenic cysts, with an estimated incidence of 1.44 cysts for every 100 un-erupted teeth. DC results from fluid accumulation in the inner space of the reduced enamel epithelium of un-erupted teeth. The clinical presentation is a painless tooth eruption failure and rarely bone expansion. Infected lesions may have pain, fever, cervical lymph node enlargement, and suppurative drainage. The radiographic presentation includes unilocular, radiolucent lesion characterized by well-defined sclerotic margins and increased follicular space (>4mm) surrounding the crown of an un-erupted tooth¹.

Bilateral and multiple cysts are usually associated with Basal Cell Nevoid Carcinoma Syndrome (Gorlin Syndrome), cleidocranial dysplasia, Maroteaux-Lamy syndrome, and in mucopolysaccharidosis². Bilateral dentigerous cyst in the absence of a syndrome is rare³. Non-syndromic bilateral odontogenic cysts have only rarely been reported in the literature, with less than twenty cases reported to date⁴.

This paper reports a rare case of bilateral dentigerous cyst in the mandibular ramus of a non-syndromic child 6 years old patient previously diagnosed with a bilateral supra-renal tumor, treated by surgical enucleation.

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ETHICAL MATTERS: This case report was approved by patient's parents through signed consent form. This case was conducted as part of a research the previously approved by Research Ethics Committee (CEP/ CONEP number 1.034.483/2015).



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Case-report

A 6 years old girl presented at the outpatient dental clinic of quaternary hospital accompanied by her mother with a chief complaint of bilateral mandible expansion. Medical history included the diagnosis of a suprarenal tumor at 2 years old, treated by surgical excision of both supra-renal glandules. The patient received hydrocortisone and fludrocortisone orally, 100mg/day and 20mg/day, respectively.

A physical examination of the patient's good general health, afebrile, eupneic, hydrated, stature, and weight at 75th percentile, no extra-oral asymmetries, and normal cognitive status. The reactional cervical lymph node was smaller than 1cm. An intra-oral examination of the oral mucosae had normal color and aspect, teeth and gums without any significant changes, no carious lesions, normal gustative papillae, and normal tongue coating cumulus. Primary teeth exfoliation was compatible with the patient's age and both inferior first molars were present.

At panoramic radiographs, there was bilateral unilocular radiolucency with sclerotic borders associated with the crowns of the inferior second molars. The right-sided lesion measured approximately 2.0×1.5 cm, which extending anteromedially to the distal surface of mandibular first molars. The left-sided lesion was slightly smaller, measuring approximately 1.5×1.5 cm and had similarly extension. The inferior second molars showed two-thirds of tooth root formation, mesiodistal impaction, and Nolla staging #6. No signals were suggesting the root resorption of adjacent teeth. Computed tomography scans showed mandible with bilateral expansive osteolytic lesions, cortical perforation posteriorly to inferior first molars, and cortical thinning at mandible basal bone (Figure 1).

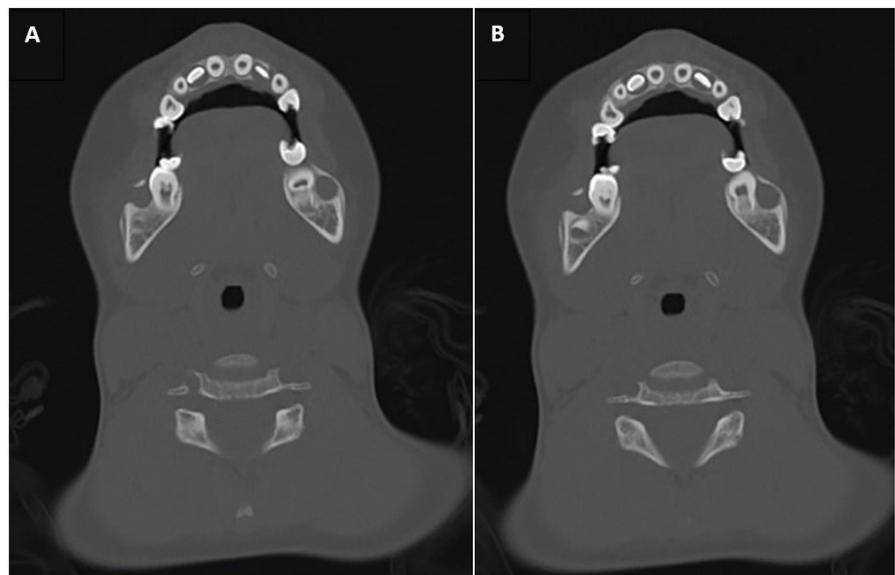


Figure 1. Two different slices of an axial view of mandible CT-scan showing bilateral cysts. Image **A** reveals a more superiorly sliced image than image **B** - Both images reveal the vestibular cortical bone perforation on the left cyst.

The diagnostic hypothesis included dentigerous cyst, unilocular keratocyst, and solid ameloblastoma. The patient was prepared for surgical enucleation under general anesthesia by hydrocortisone and fludrocortisone dose increase administered endovenous 2h before surgery and orally two days after surgery. Pre-operative endovenous cefazoline 600mg and ondansetron 2mg were administered.

Surgical cyst enucleation was performed under general anesthesia with nasotracheal ventilation. Intra-oral mucosae incisions were performed at the retromolar dentoalveolar crest with full bone exposition followed by osteotomies with surgical burs under irrigation. Second molars attached to cystic capsules were removed. The procedure followed with thorough curettage of bone defects, visual inspection of surgical sites, and wounds closures by sutures to primary healing.

Anatomopathological exams of surgical specimens revealed 2 pieces of mucosa aspect soft tissues measuring approximately 2cm attached to the dental crown. Histological findings suggestive of the dentigerous cyst were thin fibrous cystic wall lined by two-to-three cell layers of thick, nonkeratinized, stratified squamous epithelium. The connective tissue showed an inflamed infiltrate.

Post-operatively, the oral diet was restricted to soft cold foods and beverages for 72h and oral hygiene by gaze embedded in 0,12% chlorhexidine. General cares included torsal elevation, extra-oral ice packs for 48h, and lips moisture by topical burn creams (Bepanthol, Cream). EV hydrocortisone (DOSES), fludrocortisone (DOSES), antibiotics (cefazolin DOSE), and antiemetics in hospitalization. The evolution followed smoothly, and the patient was discharged after 72h post-operative with oral antibiotics prescription and recommendations of intensive physical activity and diet restriction. Normal daily corticosteroids therapy (DOSES) was resumed after patient discharge.

The patient was reevaluated one-week post-operative at the hospital outpatient clinic, showing good general evolution, no pain, no signs of infection at the surgical site, no wounds dehiscence. Three months post-operatively TC-scan showed satisfactory new-formed bone trabeculae without suggestive images of bone resorption nor cystic recidive in the retromolar region of the mandible (Figure 2). There were no signs suggestive of periodontal pockets formed in the distal aspect to the lower first molars, and parents were advised to annual follow-up.

Discussion

Dentigerous cyst is benign that affects patients three to 57 years old but is most commonly seen in the second and third decades of life. Dentigerous cysts usually occur as a solitary cyst in mandibular third molars, maxillary canines, and mandibular premolars, and they rarely involve deciduous teeth. Bilateral dentigerous cysts in the absence of a syndrome is a rare condition.

This case reports a child with previous supra-renal resection at 2yo due to an adrenal tumor. No chemotherapy was administered and there were no signs of tumor metastasis. Until now, there no previous correlation between dentigerous cysts and supra-renal tumors in children.

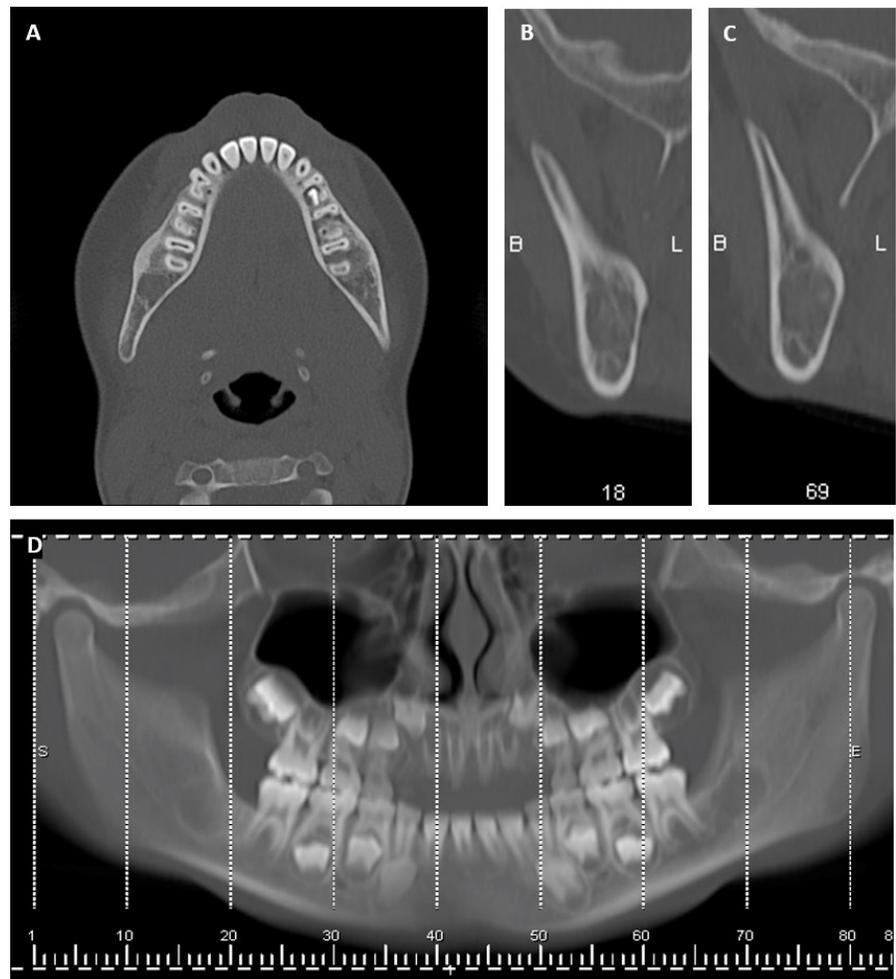


Figure 2. CT-scan images showing a 3-month post-operative. Image **A** - shows an axial slice with bilateral new-formed bone trabeculae. Images **B** and **C** - show right and left-side slices in coronal view, revealing expected outcomes. Image **D** - shows both jaws in a panoramic view.

Our patient main complaint was a bilateral cortical expansion of mandible with no signs of infection or clear causes. Panoramic radiographs and CT scans showed osteolytic lesions with well-defined sclerotic margins and increased follicular space associated with permanent second molars in mandible. Dentigerous cysts may cause slightly cortical expansion, but in the face of the patient's history of malignancies and increased risk of infections due to continuous corticosteroid therapies, a more aggressive approach was chosen. Conservative treatments include radiographic follow-up and surgical marsupialization⁵.

In these cases, the multidisciplinary approach included the participation of endocrinology, oncology, anesthesiology, pediatric surgery, head & neck surgery and the oral & maxillofacial surgery. Also, the patient's future rehabilitation may be done by an dental clinician and implant dentist. Also, we highlight the importance of multiple and long-term cares of childhood cancer survivors to improve the quality of life.

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