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Single Unilocular Radiolucent Cyst in the Angle of Mandible of a Young Man: A Case Report

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Abstract

Odontogenic keratocyst (OKC) is a benign cyst arising from the remnants of the odontogenic epithelium. OKC tends to be aggressive and recurrent. It has an array of features mimicking a variety of cysts that make the correlation of clinical, radiologic, and histopathological analysis necessary for definitive diagnosis and treatment planning. This case discusses a 25-year-old male patient presenting with extraoral swelling in the right mandible and discharge from a pericoronal flap in relation to an impacted right lower third molar tooth. The radiograph indicated the presence of a unilocular radiolucency making it indistinguishable from other radiolucent cysts of the mandible. A diagnosis of OKC was made by analyzing the aspirate. The cyst was enucleated, and Carnoy's solution was applied. The histopathologic evaluation confirmed the diagnosis of OKC, and the patient is on regular follow-up with no signs of recurrence to date.

Keywords: Odontogenic Keratocyst; Diagnosis; Pericoronal Flap; Histopathologic Evaluation; Infiltrating Quality; Odontogenic Tumor; Swelling; Orthopantomogram; Radiolucent Lesion; Carcinoma Syndrome

Introduction

In 2005, the World Health Organization renamed odontogenic keratocyst, a cyst arising from the remnants of the odontogenic epithelium "keratocystic odontogenic tumor" due to its histologic appearance and the presence of tutor markers. Recently, however, it was again renamed the original term, OKC. The cyst shows aggressive clinical behavior and high recurrence owing to its infiltrating quality. It shows male predilection, and the peak age of presentation is in the second decade [1]. The most commonly presenting region is the angle and ascending ramus [1,2] of the mandible. Histologically, the cyst may be para keratinized or orthokeratinized-the former is found to have more recurrent potential. We present a case report of a classical presentation of OKC in a young man.

Case Presentation

A 25-year-old male patient presented with concerns of pain and swelling in the lower right jaw for the past five days. The patient developed a dull, aching, continuous pain localized to the right side of the mandible. He had no systemic illness and was non-syndromic. The swelling extended from the right infraorbital margin superiorly to the lower border of the mandible on the right side inferiorly and from the right ala of the nose medially to the right angle of the mandible laterally. The swelling was spherical and smooth, with a color and temperature similar to the surrounding skin. It was tender, soft, non-fluctuant having no pulsations or thrills, non-reducible, and the skin over the swelling was freely mobile and pinchable. On the right side, a single submandibular lymph node was enlarged and tender on palpation, mobile, and firm. The patient's mouth opening was reduced to 22mm. On intraoral inspection, we noted inflammation in the pericoronal flap with pus discharge distal to the right second mandibular molar, which was also tender. The buccal vestibule was obliterated, and the lingual cortex had expanded which was bony and hard with mild crepitus on palpation.

Orthopantomogram revealed a well-defined unilocular radiolucent lesion adjoining the lower right third molar tooth extending from the upper border of mandible superiorly to 3 mm above the inferior border of the angle of the mandible inferiorly and up to 7 mm short of the posterior border of the ramus posteriorly.

A computed tomography (CT) scan revealed a well-defined, radiolucent lesion with mild scalloping in the posterior aspect of

the body and the anterior aspect of the ramus of the mandible on the right side abutting the root of the impacted lower right third molar tooth measuring 2.1 cm x 1.7 cm x 2.7 cm. The mandibular cortex was thinned severely lingually and moderately buccally. There was erosion of the anterior and medial bony walls of adjacent portions of the right mandibular canal. Pus culture revealed the presence of a few pus cells and short chains of few gram-positive cocci. On aspiration, a straw-colored cheesy fluid was obtained. A diagnosis of OKC was made.

With the patient under general anesthesia, we made an envelope incision extending from the right second premolar region posteriorly to the cyst margin. We enucleated the cyst completely along with the extraction of an associated impacted right third molar tooth, then sent the samples for histopathological examination. Soft tissue and hard tissue curettage were done followed by application of freshly prepared Carnoy's solution for three minutes after application of protective petroleum jelly coating to the exposed inferior alveolar nerve. After a wash of betadine and saline, the surgical site was packed with Bactigras dressing.

Discussion

OKC is one of the more aggressive odontogenic cysts owing to its high recurring and invading behavior. The cyst shows the presence of tumor markers such as proliferating cell nuclear antigen; Ki67; the BCE2 sequence of the enzyme dihydrolipoyl acetyltransferase; matrix metalloproteinase 2,9; and p53 [3]. The inherent growth potential of OKC is greater than any other cyst [4].

Our case represents a classic presentation of OKC. Our patient was a 25-year-old man, which correlates with the peak incidence of presentation of OKC (i.e., during the second decade of life) and aligns with the male predilection established for keratocysts and dentigerous cysts [1,5]. Also, keratocyst and ameloblastoma both present predominantly in the posterior aspect of mandible, especially in the angle region. The site of presentation is suggestive of keratocyst or ameloblastoma [1]. Our patient's clinical presentation of pain, trismus, and discharge from the site of presentation are suggestive of either classic presentation of OKC or an infected impacted third molar [1,5]. However, the radiologic and CT images showing unilocular radiolucency and scalloped margins mimic unilocular ameloblastoma and OKC [1,6]. Bone resorption in adjacent sites suggests ameloblastoma, adenomatoid odontogenic tumor or OKC. Adenomatoid odontogenic tumor was ruled out because of the site predilection of OKC and ameloblastoma [6]. The aspirate of cheesy yellow fluid is suggestive of OKC. Finally, the histopathologic report shows orthokeratinized stratified squamous lining with palisading basal cells without any dysplasia confirming the diagnosis of OKC of the orthokeratinized variety [5]. The lack of recurrence following the surgical procedure also correlates with existing data on orthokeratinized OKC's lack of recurrence [7].

OKC has a high degree of association with syndromes such as Gorlin Goltz Syndrome [7,8]. Mutation in narrow-host-range groups C gene is present in OKC patients and nevoid basal cell carcinoma syndrome patients. Therefore, a combined clinicopathologic evaluation is necessary to establish the exact variety of the cyst and tailor the management appropriately.

Conclusion

This case presents with the clinical features of a classic OKC case but also mimics the possibility of being a wide array of different cysts and tumors in certain aspects. The details of the case amplify the importance of carefully correlating the clinical and histopathological feature to arrive at a diagnosis.

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