

Ameloblastoma Arising From Dentigerous Cyst: A Case Report

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Abstract

Case Report

Dentigerous cyst (DC) is a developmental odontogenic cyst that encloses the crown of an unerupted tooth by expansion of its follicle with accumulation of fluid between the reduced enamel epithelium and the tooth crown and is attached to the neck of the tooth. The lining of DCs shows a potential for neoplastic transformation to ameloblastoma, squamous cell carcinoma, and mucoepidermoid carcinoma. Here, we report a rare case of an acanthomatous ameloblastoma arising in the wall of a DC.

Keywords: Ameloblastoma, dentigerous cyst, neoplastic transformation.

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INTRODUCTION

Embryologically, the jaws originate from ectomesenchyme. As a result of this Odontogenic cysts and tumors are a commonly occurring lesions in the jaws [1]. Odontogenic cysts, most commonly affecting the jaw, are known as osseous-destructive [2]. Based on diagnosis, jaw cysts distribution is as follows: radicular cysts 56%, dentigerous cysts 17%, nasopalatine duct cysts 13%, odontogenic keratocysts 11%, globulomaxillary cysts 2.3%, traumatic bone cysts 1.0%, and eruption cysts 0.7% [3]. Dentigerous cysts are the most common of odontogenic cysts and can occur at any tooth location, but most often occur in third molars and maxillary canines, locations most often involved in tooth impaction [4, 5]. Dentigerous cysts are nonkeratinizing cysts that develop in association with the crown of an unerupted or impacted tooth, either primary or permanent. These cysts form most likely from residual remnants of reduced enamel epithelium present after odontogenesis. These remnants of epithelium are attached at the cemento–enamel junction of the tooth, hence the crown of the tooth is contained within the cyst [6]. Rarely lesions, such as ameloblastoma, squamous carcinoma, or mucoepidermoid carcinoma can arise or be associated with a dentigerous cyst [7-9].

CASE HISTORY

A 45 year old woman reported to the department with the chief complaint of diffuse swelling on the right side of lower jaw since 1 year. There was a gradual increase in size of swelling to the present

size. Swelling was followed by pain. Extraoral examination revealed facial asymmetry with a diffuse swelling on the left side of face centered chiefly over lower border of mandible measuring approximately 3cm x4cm. On palpation, all the inspection findings were confirmed, and the swelling was tender and firm to hard in consistency.



Fig-1



Fig-2

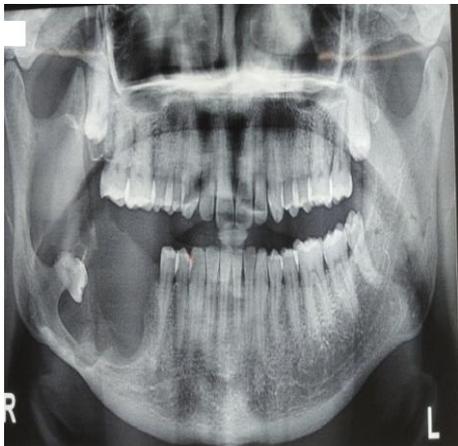


Fig-3

Intraoral examination revealed a swelling with smooth surface, obliterating buccal vestibule, and extending anteroposteriorly from 45 to alveolar ridge of 47. Buccal and lingual cortical expansion was also noticed. On palpation, the swelling was soft at 45 and firm in consistency at retromolar region. Missing teeth present in relation 46 47. Orthopantomograph revealed a unilateral multilocular radiolucency on right side extending from 45 to ascending ramus just below sigmoid notch with impacted 48.



Fig 4b



Fig-4c

Ultrasonography of the swelling showed a hypoechoic area measuring 4.54x2.8cm with semisolid ultrasound pattern, smooth boundary echo, heterogenous internal echo and intermediate posterior wall echoes. Color Doppler of the swelling showed no vascularity. Based on the clinical and radiological features, a provisional diagnosis of Dentigerous cyst was made. An incisional biopsy was done and specimen sent for histopathological examination. Contrary to the provisional diagnosis HPE showed the features of acanthomatous ameloblastoma.



Fig-4a

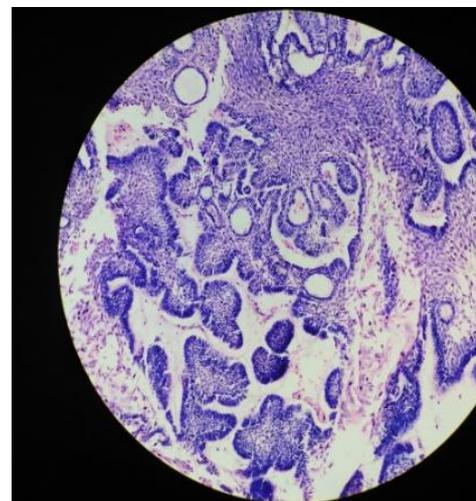


Fig-5a: HPE at 10X

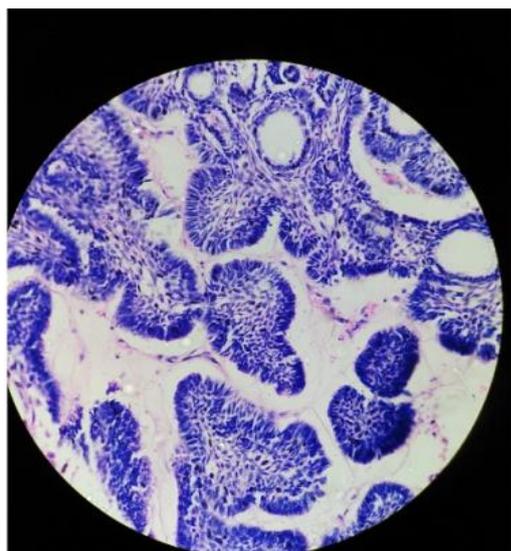


Fig-5b: HPE at 100X

DISCUSSION

Dentigerous cyst is an odontogenic lesion that represents the second most common odontogenic cyst after radicular cyst, accounting for ~24% of all true cysts in the jaw [10]. A typical dentigerous cyst presents clinically as an asymptomatic unilocular radiolucency enclosing the crown of an unerupted or impacted tooth [11]. However, dentigerous cyst can cause local destruction, bony expansion, root resorption, or displacement of teeth, which occurs more commonly with long-standing lesions [12]. In most of the cases, the diagnosis of a DC is straight forward; but even radiographically, a 'typical' DC can be diagnosed as something else, such as a dental follicle, a hyperplastic dental follicle, an odontogenic keratocyst [a keratocystic odontogenic tumour (KCOT)] or a unicystic ameloblastoma on histological analysis [11]. The histological diagnoses of these lesions are therefore critical [13]. Most of the dentigerous cysts manifest in the second and third decades of life, with peak incidences in teenagers, often developing around the crowns of mandibular third molars [14] as it was seen in our case

Ameloblastoma represents the second most common odontogenic tumor. It is slowly growing, locally invasive and has a high rate of recurrence if not treated adequately [15]. It accounts for ~1% of all tumors and cysts of the jaws [16]. There are three clinicoradiographic variants of this tumor, namely the solid or multi-cystic variant (86%), the unicystic variant (13%), and the peripheral variant (1%) [17]. The lining of a dentigerous cyst develops from reduced enamel epithelium that envelops the crown prior to eruption [18], whereas the tissues from which ameloblastoma may arise involve dental lamina rests, the developing enamel organ, the epithelial lining of an odontogenic cyst, or the basal cells of the oral mucosa [17].

Unicystic ameloblastomas are variants of ameloblastomas, which were first described by Robinson and Martinez, which refer to those cystic lesions that show clinical and radiological characteristics of odontogenic cysts, but which on histological examination, show typical ameloblastomatous epithelium which lines part of the cyst cavity, with or without a luminal or mural tumour proliferation [19].

Fifteen percent to 20% of all unicystic ameloblastomas form in the wall of dentigerous cysts [20]. The immuno-histochemical data on Ki-67 expression in ameloblastomas which arise from dentigerous cysts confirm the hypothesis that ameloblastomas which arise from dentigerous cysts have a biological behaviour which is similar to that of unicystic ameloblastomas [21].

In our case ultrasonography showed the contents of lesions to be semisolid i.e both cystic & solid. Intraosseous ameloblastomas arising in the jaws are classified as unicystic, desmoplastic, and mixed cystic & solid [15]. The mixed cystic and solid form demonstrates more aggressive behavior and is more likely to recur than unicystic and desmoplastic ameloblastomas [22]. Mixed cystic and solid ameloblastomas occur in the mandible and maxilla, with the posterior mandible the most common site of involvement [1, 23, 24, 15].

HPE showed the follicles and trabeculae of odontogenic epithelium with squamous metaplasia in the centre of islands. At microscopic analysis, discrete islands of odontogenic epithelium are seen in the follicular type; In the follicular type, cyst formation is caused by degeneration of cellular islands [15].

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