An Inflammatory Dentigerous Cyst Shows Rim Uptake on Bone Scan: A Case Report

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Abstract Dentigerous cysts are developmental odontogenic jaw cysts, commonly manifesting in the 2nd and 3rd decades of life. This article presents an extremely rare case of dentigerous cyst showing increased uptake in the peripheral rim on bone scan. Herein, we discuss the clinical, radiographic, and histologic features of dentigerous cysts as well as the pathological mechanism underlying their activities on the bone scan. Bone scan was a sensitive tool for detecting the biologic activity of dentigerous cyst in our case.

Keywords Dentigerous cyst · Bone scan · Inflammatory cyst · Expansile cyst · Osteogenesis

Introduction

Dentigerous cysts are the second most common type of odontogenic cysts, following radicular cyst [1]. The frequency of dentigerous cyst formation is 1.44 of every 100 unerupted teeth, representing nearly 17.1 % of all true jaw cysts [1].

According to the World Health Organization (WHO) classification of jaw cysts, dentigerous cysts are defined as epithelial developmental odontogenic cysts [2]. There are two types of dentigerous cysts by origin: developmental or inflammatory [3]. Our case is an extremely rare presentation of an inflammatory dentigerous cyst showing increased uptake in the peripheral rim on bone scan. We focused on the

two pathological mechanisms, osteogenesis and hyperemia during the inflammatory process, to explain the increase of uptake in the peripheral rim on a bone scan.

Case Report

A 31-year-old man presented at the Department of Dental Health of our hospital, with a complaint of progressive swelling in the right cheek for 3 months. He did not complain of any pain. Extraoral examination showed a soft swelling on the right side of the face at the angle of the mandible, which did not show any tenderness on palpation. There were no caries or wounds.

The orthopantomogram revealed a unilocular radiolucent cystic lesion containing the unerupted third molar tooth in the right mandible (Fig. 1). The cyst was invading adjacent bone, at the right angle of the mandible. Cortical thinning was observed. Computed tomography (CT) was performed for further evaluation of the content and the extent of the lesion. There was a 4.5-cm×3.4-cm sized well-defined expansile cystic lesion involving the right mandibular angle near the unerupted right third molar tooth. Moreover, contour bulging and cortical thinning were observed (Fig. 2). A ^{99m}Tc -3,3-diphosphono-1, 2-propanodicarboxylic acid (^{99m}Tc -DPD) bone scan was performed for evaluating the degree of biological activity. The bone scan showed an expansile bone lesion showing increased uptake in peripheral rim, accompanied with a central photon defect (Fig. 3).

Provisional diagnosis of dentigerous cyst was made, correlating the clinical and radiographic findings. Decompression of the cyst was performed. The entire specimen along with the tooth bud of the right mandibular molar tooth was sent to the pathologist for histopathologic evaluation. Sections were taken after processing and were stained with papanicolaou stain for histologic evaluation. Microscopic examination of the sections showed a thin fibrous wall covered with stratified

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Fig. 1 A unilocular radiolucent bone cyst (arrowheads) containing a third molar tooth (yellow arrow) is noted in right mandibular angle on orthopantomogram

squamous cells. The lining epithelium was focally eroded and the wall was mildly infiltrated with chronic inflammatory cells (Fig. 4). Based on these features, the final diagnosis of inflammatory dentigerous cyst was given. The patient remained in follow-up and no complication was observed.

Discussion

Dentigerous cysts are defined as odontogenic cysts forming surrounding the crown of an unerupted tooth [4]. Specifically,

the accumulating fluid between the reduced enamel epithelium (REE) and the tooth enamel gives pressure on the fluid which, in turn, causes separation between the follicle and the crown [4]. The pressure on an impacted follicle by an erupting tooth obstructs venous outflow and induces the swift transudation of serum through the capillary walls [5].

According to Benn and Altini [3], there are two types of dentigerous cyst. The first type is of developmental origin occurring in mature teeth, which usually happen around the late 2nd and 3rd decades [3]. These cysts involve mandibular third molars and are caused by impaction and enamel hypoplasia [3]. Histologically, they are often characterized by a thin nonkeratinizing stratified squamous epithelium resembling REE, and are typically not inflamed, unless there is a secondary infection [3]. Dentigerous cysts of inflammatory origin, the second type, occur in immature teeth and usually happen around the first or early 2nd decade of life. They typically involve mandibular premolars [3]. The inflammation from a nonvital deciduous tooth or other source is intense and often spreads to the tooth follicle. Histologically, they are characterized by hyperplastic nonkeratinizing stratified squamous epithelium with frequent anastomosing rete ridges [3].

Three possible mechanisms regarding the histogenesis of inflammatory dentigerous cysts were identified by Benn and Altini [3]. (1) The spreading of periapical inflammation from nonvital deciduous predecessors causes intrafollicular

Fig. 2 On coronal (a) and sagittal (b) CT images, cortical thinning of the expansile cystic lesion (arrowheads) with eccentrically located third molar tooth (yellow arrow) is revealed. The size is measured 4.5 cm×3.4 cm

Fig. 3 Bone scan images (a right lateral image, b anterior image, c posterior image) show central photon defect with surrounding peripheral rim uptake of right mandibular cyst (arrows), suggestive of inflammatory nature of the cyst wall

Fig. 4 On microscopic findings (a papanicolaou, magnification $\times 10$; b papanicolaou, magnification $\times 70$), the cyst shows a thin fibrous wall covered with stratified squamous cells (*asterisk*). The cyst wall also shows focal erosion and mild infiltration of chronic inflammatory cells

developmental dentigerous cysts formed around the crowns of permanent tooth. (2) Radicular cysts located at apices of deciduous teeth fuse with the follicles of impacted permanent successors. Then, protrusion of the successor teeth into the cyst result in the creation of an extrafollicular dentigerous cyst. (3) Inflammation at periapical site from nonvital deciduous teeth spreads to the follicles of unerupted permanent successors. As a result of the inflammatory exudate, the reduced enamel epithelium separates from the enamel and leads to the development of cyst formation.

To explain the increased uptake in the peripheral rim on the bone scan, we present two hypotheses, osteogenesis and hyperemia during an inflammatory process. Dentigerous cysts often grow progressively. Our patient had a history of a slowly enlarging painless swelling. The orthopantomogram and CT showed an expansile lesion evading the adjacent bone. As the cyst grew, the bony walls overlying the cyst thinned out to transmit pressure to the walls of the adjacent bone. Bone remodeling and reparative osteogenesis occurred on the peripheral borderline of the expansile cyst. This indicates high osteoblastic activity of the peripheral rim as detected on the bone scan. In addition, microscopic examination of the sections showed the lining epithelial wall was mildly infiltrated with chronic inflammatory cells. As a result, hyperemia during the inflammatory process is thought to be responsible for the increased uptake in the peripheral rim on the bone scan.

Our case, an expansile, inflammatory, dentigerous cyst showing increased uptake in the peripheral rim on the bone scan, points to the possible mechanism of uptake: reparative osteogenesis and hyperemia during the inflammatory process. Hence, scintigraphic evaluation of a dentigerous cyst would be of immense diagnostic significance, increasing a sensitivity of detecting the biologic activity of inflammatory dentigerous cyst.

Conflict of Interest The authors declare that they have no conflict of interest.

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