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A diagnostic pitfall in anterior maxillary radiolucency: A case report

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ABSTRACT

Well-defined radiolucencies in the anterior region of the upper jaw, are often considered as anatomic structures or pathologic lesions. The most common anatomic structure in this area is the shadow of incisive foramen and the most common lesion is nonodontogenic cyst known as incisive canal cyst. However, other entities especially uncommon cysts and tumors should be considered as well. In this article, we present a case of odontogenic cyst known as glandular odontogenic cyst in the anterior maxilla with histopathologic findings reminiscent of a nasopalatine duct cyst. The diagnostic sequence and criteria for differential diagnosis are discussed. Also, the significance of thorough clinical and radiographic examinations are emphasized. Actually, we are going to focus on histopathological criteria known as Rushton body which is one of the important features for differentiate between nonodontogenic cyst like nasopalatine duct cyst and an odontogenic cyst, glandular odontogenic cyst.

Key words: Odontogenic cycts, Jaw cysts, Nonodontogenic cyst, Maxilla.

Introduction

Anterior maxillary radiolucencies consist of different groups of entities. Several lesions may occur in this region. Also, the incisive canal and foramen may normally vary greatly in size. Consequently, the clinician may have some difficulty in distinguishing between a large incisive foramen and a small asymptomatic incisive canal cyst on the basis of radiographic evidence alone. Some clinicians follow the rule of thumb that radiolucencies of the incisive canal measuring less than 0.6 cm in diameter should not be considered cystic in the absence of other symptoms [1].

In routine radiographic examination, a lucent shadow between central teeth area is usually considered as an anatomic structure. If a swelling with or without pain and a significant radiolucency develops, several lesions including odontogenic and nonodontogenic cysts and tumors such as radicular cyst, odontogenic keratocyst, traumatic bone cyst, central giant cell granuloma, buccal bifurcation cyst, nasopalatine duct cyst and mucoepidermoid carcinoma should be considered in the differential diagnosis [1,2]. Yih and Krump reported an unusual case of Odontogenic keratocyst (OKC) that occurred in the nasopalatine duct with cartilage formation in the cyst wall [3]. Frequently, a diagnostic problem arises when a cystlike radiolucency is projected over apices of maxillary central incisors. In this situation, the clinician must distinguish whether this is an incisive canal cyst or a radicular cyst. Aparna et al reported a case of radiolucent lesion in relation to the roots of maxillary central incisors which was provisionally diagnosed as radicular cyst and endodontically treated accordingly. They highlighted the relevant aspects in the diagnosis of Nasopalatine duct cyst (NPDC) when it is mistaken for a radicular cyst [4]. Hilfer et al reported a case of a patent Nasopalatine duct cyst which was originally diagnosed as a sinus tract with subsequent endodontic nonsurgical retreatment and eventual extraction before endodontic consultation [5].

Usually the final diagnosis is made by histopathological examination. The epithelial lining of nasopalatine duct cyst ranges from stratified squamous to pseudostratified columnar. Cuboidal and simple columnar epithelium may be seen as well. Cilia and goblet cells may be found in association with columnar linings. In many instances a mixture of two or more types of lining cells is seen. The connective tissue wall contains small arteries and nerves, representing the nasopalatine neurovascular bundle [6]. These features, although diagnostic, are not specific to nasopalatine duct cyst. Respiratory epithelium may be seen in many odontogenic and nonodontogenic cysts especially in maxillary lesions. Therefore, the role of thorough clinical and radiographic examination in addition to careful histopathological evaluation is crucial for final diagnosis. So, focusing on histopathological criteria known as Rushton body which is one of the important features for differentiate between nonodontogenic cyst like nasopalatine duct cyst and an odontogenic cyst, glandular odontogenic cyst is the main goal of this report.

Case Presentation

A 36 years old man was referred to our dental school on October 30, 2013 with a chief complaint of swelling in the maxillary anterior region of six months duration. The patient's past dental and medical histories were not significant. Orthopantomogram (OPG) showed unilocular periapical radiolucency at the midline of maxilla measuring 2.5x 2cm which extended unilaterally to the left side. The lesion was relatively well-defined. Root resorption of adjacent teeth and periosteal reaction were not present and the lamina dura was intact (Fig-1). Oral examination showed a soft and fluctuant swelling in the labial and palatal alveolar mucosa without any pain and tenderness. Preoperative aspiration showed an amber-color fluid suggestive of a cystic lesion. Accordingly and considering the location of the lesion, a clinical diagnosis of nasopalatine duct cyst was made. Odontogenic cysts (inflammatory and developmental and possibly cystic tumors were also included in the differential diagnosis. An excisional biopsy was performed and revealed a cystic lesion which easily removed from the surrounding bone. The cyst was totally enucleated with curettage of adjacent bony tissue and sent to the oral pathology lab for histopathologic evaluation. It should be noted that surgeon did not stated that enucleated the cyst from nasopalatine duct.

Macroscopically, the specimen consisted of three pieces of irregular green-brownish soft tissue. The larger one measured 2.5*1.4*0.7 cm. Cross section showed cyst wall with 0.2 cm maximum thickness. The smaller pieces measured 0.8*0.6*0.4 cm totally. Histopathologic examination revealed a cystic lesion predominantly lined by stratified squamous epithelium with varying thickness. In other parts, the epithelial cells were flat to cuboidal and arranged in 1 to 3 cells in thickness. Also, ciliated columnar cells resembling respiratory epithelium of the sinus mucosa were observed in some areas. The epithelium was supported by a dense fibrous connective tissue with mild diffuse chronic inflammatory infiltration. According to these features and considering the radiographic findings, a diagnosis of nasopalatine duct cyst was made. Final review of microscopic slides was performed to confirm the diagnosis. Focal tiny eosinophilic structures were found in some sections; therefore, multiple sectioning of all blocks was performed and all slides were inspected carefully. Several eosinophilic linear calcifications reminiscent of Rushton bodies were found and the odontogenic origin of the lesion suspected. Superficial cuboidal and columnar cells sometimes with palisading pattern along with some goblet cells and evidences of mucus pooling were in favor of glandular odontogenic cyst (Fig 2,3). PAS positive staining especially in epithelial pools confirmed the diagnosis. Finally should noted that we were recieved patient's consent to report this case.

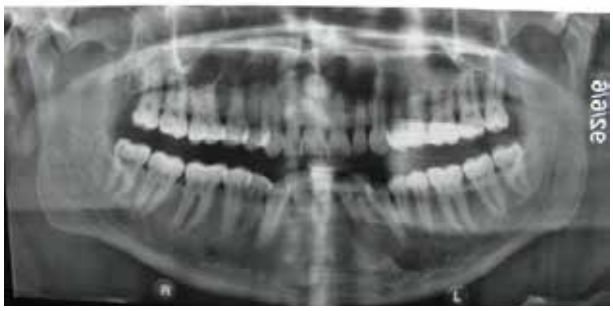

Fig 1. Panoramic radiography shows a well defined lucency with cortical border in anterior region of maxilla.

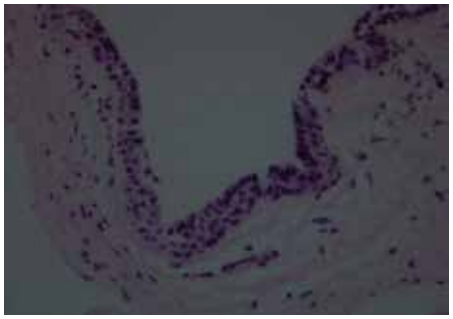

Fig 2. Histopathological slide shows cystic structure lined by stratified squamous epithelial with cuboidal superficial cell (x100 magnification).

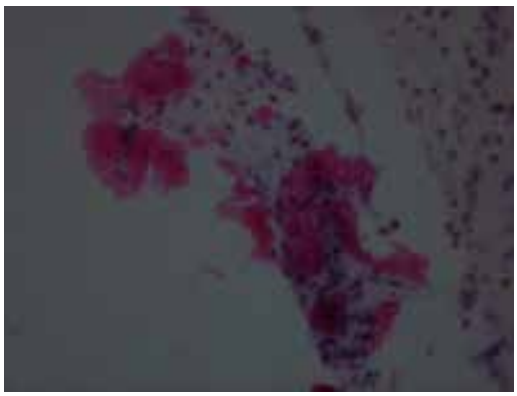

Fig 3. Histopathologic slide shows rushton bodies is placed within epithelial lining of the cyst.

Discussion

Unusual mucus producing odontogenic cyst was first described by Gardner in 1984 [7] followed by two cases reported by Padayachee and Van Wyk in 1987 [7] and eight cases in 1988 by Gardner, et al. The first reports used the term "sialodontogenic cyst" based on histopathologic similarities to salivary gland tissue. In 1992, WHO replaced the initial term with glandular odontogenic cyst (GOC) because no evidence of salivary gland differentiation was confirmed [8]. Some authors believed to develop from reduced enamel epithelium, indicating an odontogenic rather than sialogenic origin [8-10]. The most common site of involvement is the anterior region of mandible, which usually crosses the midline, but it can occur in any site of the jaws [9, 11]. There is a slight male predilection (M:F ratio1.3:1) and the mean age of patients is 45.7 year [8,9,11].

Based on radiographic and histopathological features of glandular odontogenic cyst, several lesions should be considered in the differential diagnosis. Usually, it is difficult to distinguish unilocular or multilocular glandular odontogenic cyst from other odontogenic and nonodontogenic cysts and neoplasms such as keratocyst odontogenic tumor (KCOT), ameloblastoma and myxoma. Histopathologically, intraosseous mucoepidermoid carcinoma and lateral periodontal cyst may show similar features with glandular odontogenic cyst [3,9,12]. Prabhat et al reported a case of maxillary lesion with overlapping features of glandular odontogenic cyst and central mucoepidermoid carcinoma (MEC) [9].

In our case, because of the site of occurrence and radiographic view, the first clinical impression was nasopalatine duct cyst. Other lesions included odontogenic keratocyst, central giant cell granuloma and radicular cyst. Histopathologic evaluation showed various epithelial linings, ciliated columnar cells resembling respiratory epithelium and also nerve bundles and arteries which were consistent with nasopalatine duct cyst. Careful inspection revealed superficial cuboidal and columnar cells sometimes with palisading, localized epithelial thickening and some goblet cells with evidence of mucus pooling. Most importantly, multiple sectioning showed tiny eosinophilic structures on the epithelial surface (Rushton's bodies) which confirmed the odontogenic nature of the lesion (In fact exclusively occurred in odontogenic cyst) [13].

The glandular odontogenic cyst has two clinically important attributes: 1) A high recurrence rate especially in cases treated with a conservative approach and 2) An aggressive growth potential with high resemblance to mucoepidermoid carcinoma [2]. Therefore, a correct diagnosis is of critical importance [8,12]. Sittitavornwong et al reviewed 64 reported cases of glandular odontogenic cyst and clearly demonstrated neither specific nor pathognomonic clinical and radiographic presentation for this lesion [12]. It seems that histopathological examination is the only approach recommended for final diagnosis. Several histopathologic features have been reported in the literature which are classified into major and minor categories. The major criteria include:

1- Non-keratinizing squamous epithelial lining, flat interface between cuboidal basal cells (without basal palisading) and the connective tissue wall.

2- Varying thickness of epithelium with or without epithelial focal luminal proliferation (Epithelial swirls).

3- Cuboidal eosinophilic cells or hob-nail cells.

4- Mucous (goblet) cells with intraepithelial mucous pools containing mucicarmine or Periodic Acid Schiff (PAS) positive/diastase resistant material.

5- Intraepithelial microcystic or duct-like structures.

The minor criteria include:

-Papillary proliferation of the lining epithelium or irregular surface.

-Ciliated cells.

-Multicystic architecture.

-Clear cells in basal or spinous layers [10, 12].

Each individual lesion usually presents some but not

all of different features described, often only focally within the cyst lining. Although focal presence of major criteria is necessary for diagnosis but the minor ones are not mandatory but can support it. In addition some of these individual features may be found in other lesions such as botryoid cyst, radicular or dentigerous cysts with mucous metaplasia, surgical ciliated cysts and low-grade mucoepidermoid carcinomas. There is no specific stain that differentiates glandular odontogenic cyst from these lesions [8,9,12]. However, to some extents, immunohistochemistry may help to separate GOC from central mucoepidermoid carcinoma. It has been shown that assessment of cytokeratins (CKs) 18 and 19 could be useful in the differential diagnosis [9,12].

The differential diagnosis between odontogenic and non-odontogenic cysts in anterior maxilla is somehow difficult especially when the lesion is not associated with an impacted tooth. In the present case, our main challenge was diagnosing glandular odontogenic cyst from nasopalatine duct cyst. Nasopalatine duct cyst is the most common non-odontogenic cyst of the oral cavity. It most frequently occur in 2 to 5 decades and presents as a well-defined radiolucency in the midline of maxilla [8,14]. Histopathologically, the nasopalatine duct cyst is characterized with different epithelial lining as well as nerve bundles and small muscular arteries and veins in the cyst wall [3]. These features were also present in our case. Apparently, non-odontogenic cysts should not include tooth-related structures. In our case, the key to final diagnosis was the presence of Rushton's bodies which are considered enamel-related structures and appeared after multiple sectioning.

Generally, glandular odontogenic cysts show high recurrence rates and conservative treatment is an important factor which contributes to this fact. The treatment of choice for glandular odontogenic cyst ranged from conservative approach (enucleation, marsupialization, curettage with or without peripheral ostectomy) to marginal resection and segmental resection. Gardner and Morency suggested that curettage or enucleation might be the treatment of choice for glandular odontogenic cyst, provided that the clinician closely monitors the patient. Long-term follow-up should be carried out. A period of 3 years is recommended, although some authors suggest that patients should be followed for at least 5 years after treatment [11]. Our patient was treated by complete excision and recommended for regular follow up.

Conclusion

Glandular odontogenic cyst is a rare but important odontogenic cyst which should be differentiated from several other lesions. Radiolucent lesions in anterior maxillary region should be carefully inspected to diagnose glandular odontogenic cyst from the more common nasopalatine duct cyst. Careful clinical and radiographic interpretation as well as a thorough histopathological examination including multiple sectioning are recommended for correct diagnosis.

Conflict of Interest

There is no conflict of interest to declare.

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