

CASE REPORT

Central odontogenic fibroma, hyperplastic dental follicle, or dentigerous cyst? A diagnostic dilemma: A case report

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Abstract

Central odontogenic fibroma (COF) is an extremely rare benign odontogenic tumor, which is associated with an unerupted tooth in one-third of the cases. Hyperplastic dental follicle (HDF) is an odontogenic hamartomatous lesion associated with delayed or tooth eruption failure in young patients. Dentigerous cyst (DC) is an epithelial-lined developmental cyst surrounding the crown of an unerupted or impacted tooth. Here, we present a case of 13-year-old boy with pericoronal radiolucency around impacted right maxillary canine tooth, which detected during routine dental examination on panoramic radiograph. The size of radiolucency was varied between 5 and 10 mm and was asymmetrical. Lesion was underwent biopsy with suspicion to a dentigerous cyst. On microscopic examination, the lesion was composed of a cellular fibrous connective tissue with odontogenic epithelium in the form of strands or nests throughout the lesion and calcifications in the form of basophilic cementum-like material and dentinoid. Due to its nonspecific histological features, we encountered a diagnostic dilemma. In this case report, we discuss the clinical and radiologic features of COF, DC, and HDF. We preferred COF for this lesion. By combining surgical and orthodontic treatments, the impacted tooth was directed to its correct position in the maxilla. This case report highlights the importance of clinicopathological correlation in the diagnosis of pericoronal radiolucencies.

KEYWORDS

dental follicle, dentigerous cyst, fibroma, odontogenic cysts, odontogenic tumors

1 | INTRODUCTION

Central odontogenic fibroma (COF), dentigerous cyst (DC), and hyperplastic dental follicle (HDF) are frequently confused with each other in a pericoronal position.¹

Central odontogenic fibroma is a rare benign neoplasm of the jaw bones, accounting for less than 0.1%

of all odontogenic tumors. COF composed of varying amounts of inactive odontogenic epithelium embedded in a neoplastic mature and fibrous stroma.¹⁻³ The lesion may evolve from dental follicle.^{4,5} COFs occur in different ages ranged from 4 to 80. Both jaws are affected equally; most maxillary lesions are located anterior to the first molar. One-third of these tumors are associated with an

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unerupted tooth. Smaller lesions are usually asymptomatic, but larger ones may be associated with localized bony expansion or loosening of teeth.^{6,7} Radiographically, smaller tumors are well-defined unilocular radiolucent lesions. Approximately 12% of COFs will show radiopaque flecks within the lesion.² Formerly, solid fibrous masses that were associated with the crown of an unerupted tooth classified as COF. Most of such lesions today consider representing only HDF, and these should not be considered to be neoplasms.¹

Hyperplastic dental follicle is an odontogenic hamartomatous lesion associated with delayed or tooth eruption failure in young patients. The occurrence of this pericoronal dental lesion seems to be more frequent than the literature has reported.³ It involves mostly permanent first and second molars. The radiographic feature of HDF is characterized by well-circumscribed radiolucency surrounding the crown of an unerupted tooth, frequently mimicking dentigerous cyst. Microscopically, HDF consists of fibrous connective tissue containing odontogenic epithelium, multinucleated giant cells, and calcification foci.^{4,8}

Dentigerous cyst is an epithelial-lined developmental cyst formed by accumulation of fluid between the reduced enamel epithelium (REE) and crown of an unerupted tooth. They most often involve mandibular third molars. They are discovered frequently in patients between 10 and 30 years of age. Small DCs are usually asymptomatic. Radiographically, a DC shows a unilocular radiolucency that is associated with the crown of an unerupted tooth.⁹ An epithelial lining enclose the lumen of the cyst. Small islands or cords of inactive odontogenic epithelium may be present in the fibrous wall. Because a thin layer of REE normally lines the DF surrounding the crown of an unerupted tooth, it can be difficult to distinguish a small DC from simply a normal DF or HDF based on microscopic features alone.¹⁰

2 | CASE REPORT

A 13-year-old boy and his parents came to our dental clinic for routine dental examinations. During oral examinations, we found that his maxillary primary canine tooth has been retained and its succeeding permanent tooth has not been erupted. A panoramic radiograph was taken which showed impaction of right permanent maxillary canine tooth; a unilocular radiolucent lesion had been surrounded the crown of the tooth (Figure 1). The size of radiolucent space was varied in different areas from 5 mm in distal portion of impacted tooth to 10 mm in mesial portion of the tooth. A corticated border was also observed around the lucent space.



FIGURE 1 Unilocular radiolucency around the crown of impacted maxillary right permanent canine

Lesion was underwent biopsy with suspicious to a dentigerous cyst. On microscopic examination, the lesion was composed of a fairly cellular fibrous connective tissue with collagen fibers arranged in interwoven bundles in some areas. Odontogenic epithelium in the form of strands or nests was present throughout the lesion and was a striking component (Figure 2). Calcifications consisted of basophilic cementum-like material and dentinoid were present in some areas (Figure 3). Since these microscopic findings were common between COF (epithelium rich type or WHO type), DC, and HDF, we encountered a diagnostic challenge. After consulting with an oral and maxillofacial radiologist and considering the clinical, radiologic, and histopathologic features of the lesion, especially the size of the lesion, a central odontogenic fibroma (epithelium rich type) diagnosis was preferred for the lesion. The patient was referred to an orthodontist for continuation of his treatment. By combining surgical and orthodontic treatments, the impacted tooth was directed to its correct position in the maxilla.

3 | DISCUSSION

Classification of odontogenic lesions and tumors has always been debated. For example, odontogenic keratocyst (OKC) which had been previously classified as an odontogenic tumor has been restored into the cystic lesions according to the 4th edition of WHO Classification. Epidemiologic data related to odontogenic lesions have been associated with a variety of results due to the change in the classification of lesions over the years.¹¹ In the same way, at first, the term “COF” was given to every enlarged DF until Gardner separated between those different types although difficulties persisted in distinguishing between HDF and COF.⁶

Clinically, DCs, HDF, and pericoronal COF can be in the differential diagnosis of each other.¹⁰ Due to its non-specific histological features, COF may be confused with other odontogenic lesions such as HDF that highlights the

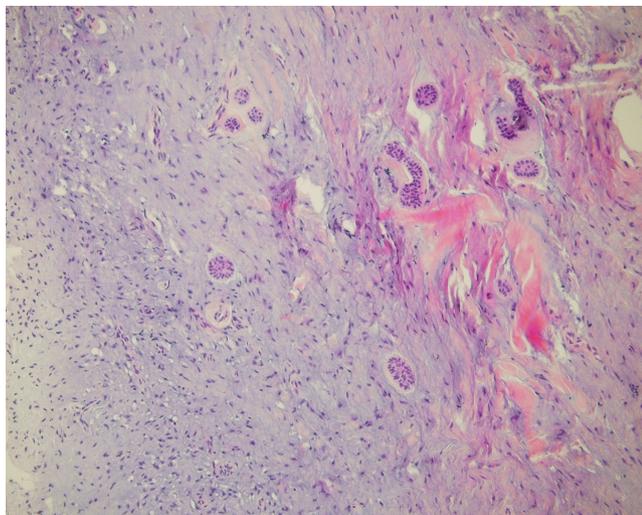


FIGURE 2 Fibrous connective tissue containing strands and nests of odontogenic epithelium throughout the lesion (100× magnification)

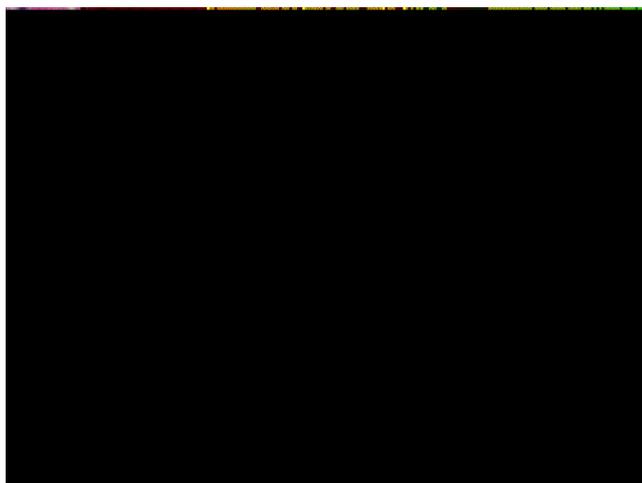


FIGURE 3 Odontogenic epithelial nests with cementum-like calcification (400× magnification)

importance of clinicopathological correlation in the diagnosis of COF.⁵

Radiographic distinction between a small DC and an HDF around the crown of an unerupted tooth is difficult. For a lesion to be considered as a DC, some authors believe that the radiolucent space surrounding the crown of tooth should be at least 3–4 mm in diameter, but this diameter is just a suggestion.³ Because the radiolucent space in our case was > 4 mm, it was more likely to be a DC rather than HDF. In histopathologic examination of a DC, there is an epithelial lining consists of 2–4 layers of flattened non-keratinizing cells.¹⁰ In our case, no epithelial lining was seen. Therefore, the diagnosis of a DC was excluded from our differential diagnosis.

It is possible to demonstrate an epithelial lining in its inner aspect (the REE) in microscopic view of most of DFs, a finding not seen in COF. In our case, no epithelial lining was seen, a finding that complicated the differentiation of our case from HDF microscopically. Variable amounts of odontogenic epithelial remnants are found in up to 79% of DFs, sometimes exhibiting foci of squamous metaplasia, which differ morphologically from the epithelial islands/cords found in COF; in our case, no squamous metaplasia was found, a finding that further complicated the differentiation of our case from HDF microscopically.⁷

DF may also have similar features like COF such as odontogenic rests and some calcifications but differ in absence of fibroblastic connective tissue arranged in interwoven strands, which is a characteristic of COF.^{6,7} In histopathologic view of our case, interwoven collagen strands that are the characteristics of COF were seen. It has been reported that examination of the thickness of collagen fibers can serve as a method to differentiate between the normal and abnormal collagen. In Hirshberg et al.'s study, they found different polarized colors of collagen fibers in COF and HDF lesions by using picrosirius red staining followed by polarizing microscopy, which can selectively demonstrate collagen. Polarization colors of the thick collagen fibers of COF showed small percentage of orange and yellowish-orange colors, while they were found in high percentage in HDF.⁶

Radiographically, HDF is usually symmetrical unlike COF.^{6,7} The pericoronal radiolucency around the impacted canine tooth in our case was asymmetric. The radiolucent space in mesial portion of the impacted tooth was very wider than distal portion; thus, according to this, the lesion was a COF rather than a HDF.

Shanab and Mosqueda-Taylor suggested that HDF cannot be more than 4 mm in diameter^{6,7} although this cutoff point is just a suggestion. Since the lesion's size of our case was very > 4 mm, it cannot be a HDF according to this suggestion and it is more compatible with COF (the size of radiolucent space was varied from 5 mm in the distal portion of the crown of canine tooth to 10 mm in the mesial portion of the tooth).

Radiolucent areas > 3 mm may be indicative of alteration in the dental follicle. Some authors consider a follicle as normal with a maximum thickness of up to 2.5 mm; follicles with a radiolucent area larger than 2.5 mm in size should be evaluated to rule out possible cysts or tumors.⁸ This criterion emphasizes that our case was a COF.

Our patient's age (13 years old) was very lower than the reported mean age of COFs (40 years old). Our patient's gender (male) did not match the overall gender predilection (strong female predilection). These features added to the rarity of our case. The location of the tumor in our patient (anterior to the first molar) was corresponding to

the dominant site of maxillary COFs. Like many reported lesions, our patient's tumor had a unilocular radiolucency appearance with cortical border that caused the root divergence of adjacent teeth.

In contrast to a DF, COF is a destructive lesion with persistent growth.⁶ COF is considered to be a neoplasm, and surgical therapy is usually more extensive than that required for the removal of unerupted teeth and their associated DFs. COFs are usually treated by enucleation and severe curettage. A few recurrences have been documented, but the prognosis is very good.¹

4 | CONCLUSION

Sometimes, it is difficult to differentiate central odontogenic fibroma from hyperplastic dental follicle. A correct diagnosis should be based on clinical, radiographic, and histological findings.

AUTHOR CONTRIBUTIONS

Mahdiah Rajabi-moghaddam, Ghazaleh Mozafari, and Hamid Abbaszadeh have made substantial contributions to conception and design of the study. Mahdiah Rajabi-moghaddam and Hamid Abbaszadeh have been involved in data collection and analysis. Mahdiah Rajabi-moghaddam, Ghazaleh Mozafari, and Hamid Abbaszadeh have been involved in data interpretation and drafting the manuscript. Mahdiah Rajabi-moghaddam, Ghazaleh Mozafari, and Hamid Abbaszadeh have critically revised the manuscript. All authors have given final approval of the version to be published.

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CONFLICT OF INTEREST

There is no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author, upon reasonable request.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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