Hindawi Publishing Corporation
Case Reports in Dentistry
Volume 2016, Article ID 4190827, 3 pages
http://dx.doi.org/10.1155/2016/4190827

Case Report

A Rare Pathological Entity of Multiple Calcified Hyperplastic Dental Follicles

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Received 9 November 2016; Accepted 12 December 2016

Academic Editor: Luis M. J. Gutierrez

Multiple calcified hyperplastic dental follicles (MCHDF) are an extremely rare condition which has been categorized as a separate pathological entity very recently. It was initially described by Sandler et al. Gardner and Radden proposed this as a separate pathological entity. This disease is characterized by multiple unerupted teeth with abundant calcifications and rests of odontogenic epithelium in enlarged dental follicles.

1. Introduction

Multiple calcified hyperplastic dental follicles are an extremely rare condition which is found to be associated with enlarged dental follicles. MCHDF has been described as odontogenic hamartomatous lesion that occurs in pericoronal tissues of unerupted tooth [1, 2]. Most cases found in literature are found to affect young individuals [3] involving first and second molars [3–7].

Differentiating MCHDF from other odontogenic tumors is important as they have different pathogenesis and recurrence rates. There is always a clinical significance evaluation of enlarged dental follicles with unerupted teeth as they can turn into cysts or tumors easily. As this case is a very rare case, its rare presentation and the management have not been decided yet.

Up to now, patients are being managed multidisciplinarily.

2. Case Report

This is an effort to report a case of an 8-year-old male patient who presented with a complaint of absence of multiple permanent teeth in the year 2012. The patient was in his early mixed dentition. The dentition was 55, 54, 53, 11, erupting 22, erupting 22, 63, 64, 65, 75, 74, 73, 31, 41, 82, 83, 84, and 85. The patient was diagnosed to have amelogenesis imperfecta clinically. Later, considering the clinical, histopathological, and radiological findings together, amelogenesis imperfect was excluded from the differential diagnosis. A Dentopanaromic Tomography (Figure 1) and Intraoral Periapical radiographs of 12, 11, 21, 22, 32, 31, 41, and 42 were ordered. In the DPT, there were permanent tooth buds of 17, 16, 15, 14, 13, 23, 24, 25, 26, 27, 37, 36, 35, 34, 33, 43, 44, 45, 46, and 47. In the IOPA radiographs, permanent tooth buds of 12, 22, 32, and 42 were present.

After radiological analysis, it was decided to review the patient after three months. However, the patient revisited after two years in the year 2014 with same complaint. After clinical examination of the patient, it was decided to give a bite raising appliance to facilitate the eruption of the permanents 16, 26, 36, and 46. And later, surgical exposure of 16 and 46 was done. In subsequent visits, the patient has undergone a root canal treatment of 21 (because the tooth was nonvital due to caries). Study models were taken in order to plan the treatment as there was not enough Occlusal Vertical Dimension (OVD) for the eruption of permanent teeth. It was decided to place stainless steel crowns (SS) on 55, 54, 64, 65, 75, 74, 84, and 85, thus creating an anterior open bite of two to three millimeters. However, SS crowns of 54, 64, 74, and 84 showed poor prognosis. SS crowns were placed on 55, 65, 75, and 85 and were changed time to time in order to maintain

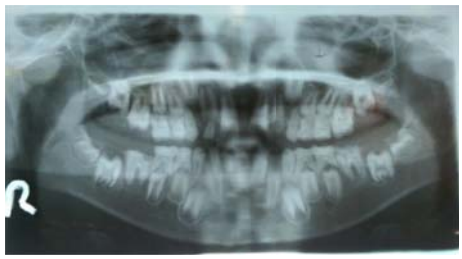

Figure 1: DPT taken in December 2012.

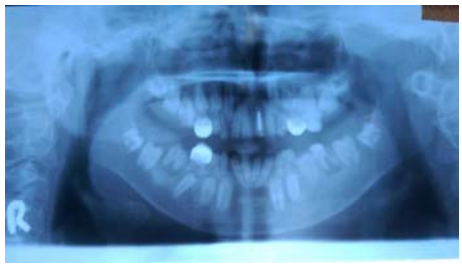

Figure 2: DPT taken in September 2015.

the occlusion intact. During the review visits, SS crowns of 64 and 74 and 55, 65, and 75 were replaced to further increase OVD in order to facilitate permanent eruption.

Once again, the patient presented in the year 2015 complaining of pain from lower jaw tooth. On examination, teeth present were 16, 55, 54, 53, 12, 11, 21, 22, 63, 64, 65, 26, 36, 74, 73, 32, 31, 41, 42, 83, 84, 85, and 46. 16, 26, and 36 were found to be septic roots. A DPT (Figure 2) was taken again and extractions of septic roots were done. In the DPT, there were permanent tooth buds of 18, 17, 15, 14, 13, 23, 24, 25, 27, 28, 38, 37, 35, 34, 44, 45, 47, and 48 with radiolucent dental follicles covering the crowns of them.

Clinically, there was thick mucosal growth on the posterior ridge area. When considering all these factors, there was no eruption potential for the impacted teeth. It was decided to review the patient after three months. On the review appointment, it was found that thick mucosal growth is improving and teeth were not still erupting in relation to lower molar region. An incisional biopsy from 47 region including both mucosa and alveolar bone was planned (Figures 3 and 4).

Histologically, mucosa covered by acanthotic parakeratinized stratified squamous epithelium was evident. The corium is composed of loosely arranged collagen fibers with focal myxoid changes. Numerous calcifications with a few odontogenic epithelial rests are also noted. Those histopathological features were consistent with the diagnosis of multiple calcified hyperplastic dental follicles (Figure 5).

3. Discussion

Multiple calcified hyperplastic dental follicles are an extremely rare presentation in the international literature. Up to 2014, only 13 cases have been reported. They seem to mostly affect young males [8]. Mostly, mandible is more affected than maxilla [8]. This case is also inconsistent with

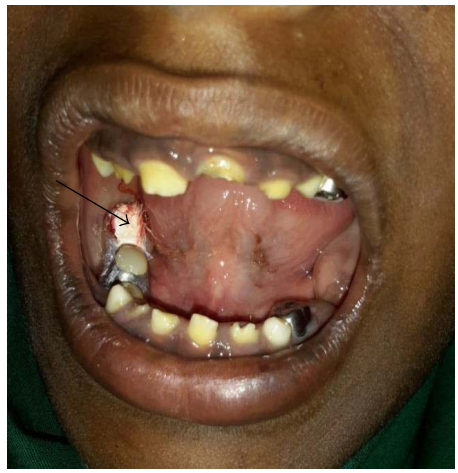

Figure 3: The incisional biopsy was taken in 47 area.

Figure 4: Firm tissue part measuring 0.6 × 0.1 × 0.4 cm consisting of mucosa and alveolar bone.

the literature. Microscopic findings in hyperplastic dental follicle include the presence of fibrous connective tissue, wavy collagen fibers, strands and islands of odontogenic epithelium, multinucleated giant cells, and varying sizes of basophilic mineralized areas present in cementum like calcifications [9, 10].]. The present case has also shown the histopathological features reported in the literature.

Histopathologically, MCHDF resembles WHO type of central odontogenic fibroma. Both lesions have connective tissue, odontogenic epithelium, and calcifications. The connective tissue of odontogenic fibroma is very cellular and is often interwoven with less cellular areas, while it might be quite vascular [9]. However, this case also showed more fibrous tissue and only focal myxoid changes. Moreover, WHO type of central odontogenic fibroma shows osseous development and a family trait, whereas no such features are found in MCHDF [11, 12].

Radiographically, calcifying cystic odontogenic tumor, adenomatoid odontogenic tumor (AOT), and ameloblastic fibroodontoma and calcifying epithelial odontogenic tumor

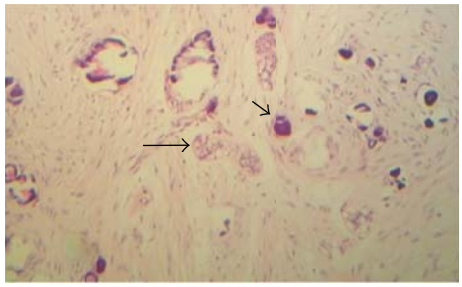

Figure 5: (H&E, 40x) calcified areas (short arrow); odontogenic epithelial islands (long arrow).

(CEOT) may show radiopaque foci, and they may be associated with unerupted teeth [13–16]. MCHDF also shows radio opaque foci associated with unerupted teeth [13]. Therefore, radiographical differentiation from same kind of lesions is also important. The aetiology and the biological mechanisms of this disease are not yet clearly understood. There is no apparent relationship between this disease and cleidocranial dysplasia or Gardner's syndrome, two other disorders in which there are multiple unerupted teeth [9]. Some medical conditions also can cause tooth impaction such as hypothyroidism. Therefore, excluding them is also important. In this case, the patient did not have any known systemic diseases. Making a definitive diagnosis with all clinical, radiological, and histopathological features is important because the management of other similar diseases is different from this particular disease.

4. Conclusion

This is a case report of a young male patient who presented with unerupted multiple permanent teeth and diagnosed as multiple calcified hyperplastic dental follicles as indicated by clinical, radiological, and histological features.

Competing Interests

The authors declare that there are no competing interests regarding the publication of this paper.

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