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Mandibular Central Odontogenic Fibroma in a 32-Year-Old Man: A Case Report

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ABSTRACT

Central odontogenic fibroma (COF) has been described as a rare benign tumor of the jaw, usually an asymptomatic slow-growing neoplasm. The aim of this study is an explanation of a young man's mandibular central odontogenic fibroma, a lesion that is expected to be seen mostly within the upper jaw. We reported a case of a COF, that went undiagnosed for about 16-years and was located within the right mandibular bone and accompanied by a moderate swelling crossed the mid-line. His chief complaint was the replacement of an implant into the mandibular right canine edentulous area. Anyway, the lesion was revealed by a plain radiographic image (an OPG) accidentally. The patient was treated surgically by an excisional biopsy without any noticeable post-operative complications finally. Actually, an acceptable enucleation and curettage can be the only treatment method for a COF. As a result, early diagnosis is key to detecting new pathological cases, especially ones that are not prevalent. Occasionally, both routine radiographic imaging and histopathological evaluation are useful to discover COF. Odontogenic lesions usually do not follow their common patterns so sufficient carefulness and a complete assessment of any patient are definitely important.

KEYWORDS

Central odontogenic fibroma; Benign; Tumor; Enucleation

INTRODUCTION

Odontogenic fibroma is considered as a benign neoplasm with odontogenic origin like dental follicle, periodontal ligament or dental papillae [1-3]. This lesion is uncommon, including 0.1 percent of all odontogenic tumors [4,5]. Until 2016, approximately 100 cases have been reported in the world [6]. Odontogenic fibroma can be central or peripheral in the upper or lower jaw, but usually located in the posterior of the mandible and the anterior of the maxilla [1,5]. The incidence of this lesion is more in females than males (1.8/1) with wide age range about 4 years to 80 years [6]. Central Odontogenic Fibroma usually appear radiolucent or mixed, multilocular or unilocular and 12% with radiopaque flecks in radiographic images; Additionally, this lesion can cause

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root resorption or root divergence of the associated teeth [6]. It can be detected in two separate histopathological types: The WHO (epithelium-rich) or the simple (epithelium-poor) type [6,7]. The aim of this report is an explanation about a young man's mandibular central odontogenic fibroma in the canine-premolar region.

CASE REPORT

A 32-years-old patient with the chief complaint of the replacement an implant into the mandibular right canine edentulous area went to a dental office about two years ago. The dental surgeon properly suspected a prominence below the toothless ridge, requested a panoramic view. Then, she noticed a lesion and also the impacted canine displacing to the molar region radiographically and in order to further diagnostic workup, referred him to the oral medicine department of Zanjan Dentistry Faculty. His medical and dental history was normal. According to him, the patient had the little solid painless swollen lesion from his adolescence until he noticed the absence of the right mandibular canine. On extra-oral examination, no facial asymmetry or swollen lymph node in submandibular or submental area was seen. The sensation of the lower lip, the color of overlaying mucosa and the occlusion pattern were normal, and no displacement of any erupted teeth was seen. However, there was a slight swelling in canine-premolar area on intraoral examination. Radiographically, a single radiolucency with a well-defined border and internal septa showing the mixed intra-osseous lesion extended from mesial side of right first molar to mesial side of the left lateral incisor (crossing the mid-line) and buccolingually, from alveolar crest to inferior border of the mandible was considerable. The vitality tests of all teeth near the lesion were positive and tapping with the end of a dental mirror didn't cause any pain. The first differential diagnosis according to his X-ray panoramic, was SBC (simple bone cyst), then Odontogenic Myxoma (Figure 1 and Figure 2).

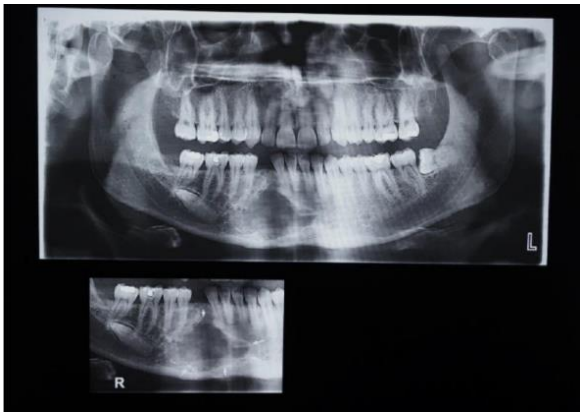

Figure 1: Radiographic features of central odontogenic fibroma. The X-ray panoramic shows a multi-locular radiolucency extended from mesial side of right first molar to mesial side of the left lateral incisor (crossing the mid-line) and buccolingually, from alveolar crest to inferior border of the mandible (white arrows).

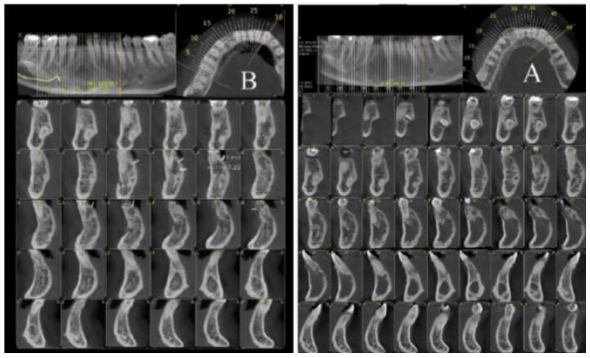

Figure 2: The CBCT (cone beam computed tomography) imaging, A) Before treatment and B) After a one-year follow up.

The CBCT (cone beam computed tomography) showed the lesion between the roots of the right lateral incisor and the first premolar, and also the displacement of the impacted right canine to apexes of the right molars. Actually, the canine moved distally. The extension of the lesion was more than the expansion of that. Unlike some previous studies, any root resorption or lamina dura destruction was not found. The inferior alveolar canal was intact. The jaw border only became thin without any perforation. The canine follicle was normal too. Due to all descriptions, at first Odontogenic Myxoma, secondly, Odontogenic Keratocyst (Keratocystic Odontogenic Tumor) and then desmoplastic fibroma can be the differential diagnosis. Next, to achieve a subtle diagnosis, an excisional biopsy was performed. The sample within a 10% formalin solution transferred to the laboratory.

The gross of the lesion is considerable in figure 3. Specimens consisted of three pieces of white-cream soft tissue with elastic consistency, totally measuring 2 cm × 2 cm × 1 cm.

Figure 3: The excisional biopsy. The gross of the lesion is considerable in these pictures, totally measuring 2 cm × 2 cm × 1 cm.

The histopathological specifications are shown in figure 4. The sections revealed a proliferation of stellate fibroblasts arranged in a whorled pattern with fine collagen fibrils. Small foci of odontogenic epithelial rests are seen in some parts, so it's actually the WHO type (epithelium-rich) of COF. Calcifications composed of cementum like material are also present. No increased mitosis or necrosis is seen. As a result, the pathological diagnosis is consistent with central odontogenic fibroma.

The treatment plan was the extraction of the whole lesion under local anesthesia. The only adverse effect of the surgery after a 6-months follow-up was a little paresthesia existed in first weeks. One-year follow-up, by CBCT, showed the small radiolucency in the lesion site, maybe meaning the recurrence probably due to the inadequate surgery. But it was well-defined with no invasion to the cortex, or the dental structures and its diameter was 7.2 mm × 9.1 mm maximum. Compared to the previous CBCT, these points are remarkable: 1) The size of the lesion become lesser, and the greatest parts of the area replaced by the new bone. 2) The former CBCT showed the considerations above too, so we don't have any recurrency truly. 3) We can see the fenestration of the buccal table (the surgical area). A reformatory surgery to elimination of remained parts of lesion is suggested. It should be removed by surgery again. Ultimately, the 5-years follow-up is necessary, too.

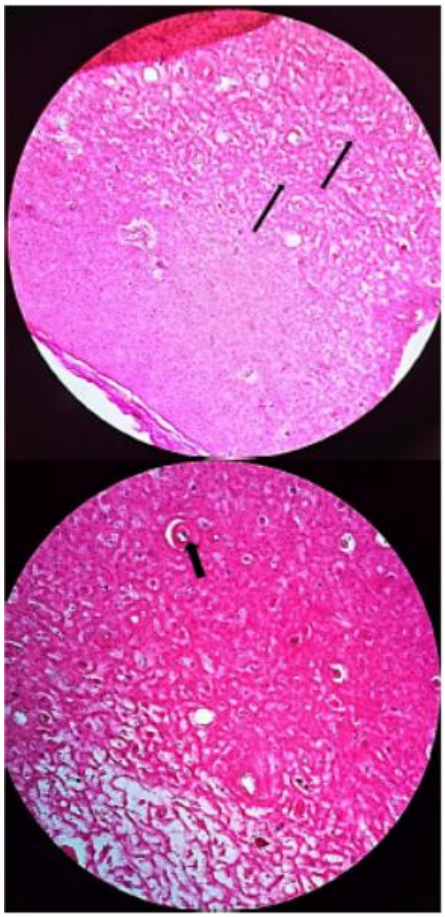

Figure 4: The microscopic details are shown here. The sections revealed a proliferation of stellate fibroblasts (narrow arrows in the upper image) arranged in a whorled pattern with fine collagen fibrils. Calcifications composed of cementum like material (wide arrow in the lower image) are also present. No increased mitosis or necrosis is seen.

DISCUSSION

Thoroughly, the occurrence of Central Odontogenic Fibroma in women is about two times more than men. Anyway, the mentioned patient was not a woman. The lesion formation caused a slow expansion of the cortical bone in anterior right part of the mandible, though central odontogenic fibroma usually occurs in the posterior region of the lower jaw. It. The lesion went undiagnosed for some years similarly Bandura et al. reported a young adult woman whose COF lesion within the body and the ramus of the mandible was not discovered despite recurrent radiographic assessments [5]. The source of this lesion generally is the ectomesenchyme cells with or without the odontogenic epithelium. The etiology of COF is still unknown and no special environmental factor about that have been found. The WHO (World Health Organization) presented some different classifications of the lesion from 1971 until now. In 2005, the existence of the epithelium layer defined new COF subclassifications called: the simple (epithelium-poor) type and the complex (epithelium-rich) type, but the last classification of head and neck tumors taken by the WHO, eliminated the simple form in 2017 [2].

Bandura et al showed that COF is probably a local etiologic factor to delay tooth eruption. Also, the case that they reported is the first issue introducing a unilateral delayed tooth eruption [5]. So, clinicians should notice carefully to any clear delayed tooth eruption, specially, if the contralateral tissue is normal. Finally, the article suggested more orthopantomography assessment to find any radiologic abnormality. In A. Santoro et al. research, the localized radi-olucent lesion at the superior margin of the mandibular right angle was related to the impacted second molar [3].

About the size of the lesion, there are many researches reporting variable measures. The lesion estimated 1 cm × 2 cm in a large amount of them, similarly in our report measured 1 cm × 2 cm × 2 cm. Therefore, Shanab reported a case of COF measured 2 cm × 3 cm or Santora submitted a case was about 3 cm × 4 cm × 3 cm with both buccal and lingual swelling in canine-premolar region [3,8]. In Pippi‘s research, a lesion with multiple fragments was not exceeded 2.5 cm in diameter [2].

In some articles one or two missing or delayed erupted teeth observed. In Shanab‘s report the first premolar and in Bandura’s report both first and second premolars were missed [2,8]. Contrastingly, no tooth absence was seen in ours, sometimes it can be irrelevant to COF lesions consequently. But the interesting point to say is the displacement of the impacted canine with a normal follicle to the molar region. At first look it might lead us to misdiagnosis. Obviously, the canine impaction cannot be completely disjoined to COF because it seems that the lesion pushed the impacted canine distally. This solid fibrous mass almost always is associated with the crown of an unerupted tooth [6]. Bandura et al. observed positive pulp vitality test result of all teeth closing to an asymptomatic COF lesion located in mandibular ramus and body the with no sensitivity of the inferior alveolar nerve [5]. The intact inferior alveolar nerve canal, vital teeth and not any sign of root resorption or lamina dura destruction were also seen in this study. Although the vitality test of the impacted canine was impossible to perform, mandibular incisors and premolars had normally positive result. The teeth located near the lesion were vital too in another article. So, it can be useful to consider the vitality test of the teeth near a radiolucency; however, the relationship between a large radiolucency and a tooth is mostly improbable. Regardless, The Neville says that about one-third of the odontogenic fibromas including central odontogenic fibroma, associated with an unerupted tooth. COF commonly occurs in the mandible rather than maxilla confirmed by some articles. According to the Neville Handbook of Pathology published in 2016, the most distribution of this lesion is posterior to the first mandibular molar and anterior to the first molar of the maxilla, both jaws equally affected. COF can be appeared

at the age of four to 80, significantly a wide range and even there is a case of this lesion in a 3-years old girl's mandible, which resected and reconstructed by bone graft without any recurrence [6]. Radiographically, the distinguished sclerotic border of COF was seen in nearly all of the reports. KOT (Keratocyst Odontogenic Tumor) is a differential diagnosis of COF because the extension of both is more than the expansion of them. The existence of collagen fibrils, odontogenic epithelial islands and fibrous connective tissue is the most prominent highlight in histopathological features. Some cementum-like materials were also noticeable confirmed by Parkash et al. research [2]. No sign of any increased mitosis or observable necrosis in the lesion rules out malignancies basically (Figure 5).

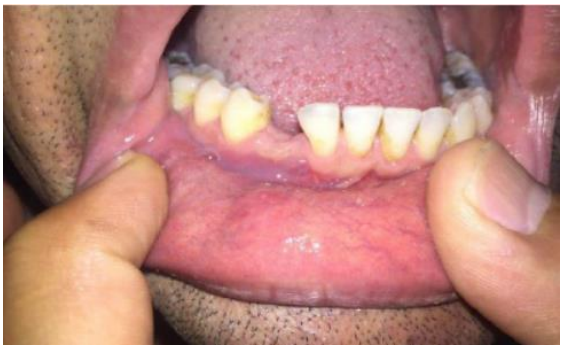

Figure 5: The buccal view of the mandibular teeth after a one-year follow-up.

COF treatment is a conservative surgery to enucleation of the lesion [2,7]. Relapse is mostly uncommon, but it can happen after an incomplete surgery. Thus, a 5-years follow up is advisable [2]. In the recent report, remaining some little parts of the previous lesion after a one-year period, indicated an additional surgery and a long-term follow-up continues.

Finally, the importance of the initial diagnosis then the surgical methods is declared clearly by Yasuyuki Shimada et al.; so that a nevoid basal cell carcinoma in a 14-year-old boy, firstly presented a central odontogenic fibroma in his mandible instead [9]. Interestingly, a curios case report of COF with unusual histologic features of the entrapped neural elements and the mast cells, revealing a detailed pathogenicity of this lesion by mast cells secretion as stem cell factors and the over-expression of C-kits, published in 2018. In summary, surgical treatment approach can be substituted by the prospect of using C-kit inhibitor drugs, specially, when they are contraindicated [10].

CONCLUSION

The accurate diagnosis of the oral-cavity lesions roughly needs a proper clinical, radiographic and pathological assessment simultaneously. Odontogenic lesions usually do not follow their common patterns so a sufficient carefulness and a complete assessment of any patient is definitely important.

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CONFLICT OF INTERESTS

There is no conflict of interest between co-authors.

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