Odontogenic Myxofibroma: A Case Report

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Abstract

Odontogenic myxofibroma (OF), slow-growing, extremely rare tumor, originated from odontogenic ectomesenchyme. OF is a subtype of odontogenic myxoma (OM) which composed of lots of collagen fiber in the extracellular matrix. The characteristics of OF are similar to OM including treatment options. Clinically, OF presents as painless swelling. Radiographically, OF shows the multilocular radiolucency honey comb appearance which are similar with other odontogenic tumor likes ameloblastoma, odontogenic myxoma, central giant cell granuloma and odontogenic fibroma.

Histopathological examination should perform to confirm the diagnosis. In Thailand, Odontogenic myxoma including odontogenic myxofibroma in jaw bone was reported only 3.1% of odontogenic tumors1. Since OF was rarely reported in Thailand and limited study of OF around the world, the present study report case of OF found in Thailand including diagnosis and treatment approach.

The objective of the present case report were to report an odontogenic myxofibroma case, describe its clinical manifestations, histological features and treatment protocol, and emphasize clinical awareness of this uncommon odontogenic tumor among dental general practitioner.

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Introduction

Odontogenic myxofibroma (OF), slow-growing, extremely rare tumor, originated from odontogenic ectomesenchyme. OF is a subtype of odontogenic myxoma (OM) which composed of lots of collagen fiber in the extracellular matrix. Most of the patients are painless swelling. The characteristics of OF are similar to OM including treatment options. OM including OF exhibits radiographic characteristics as a unilocular or multilocular radiolucency with internal septa separating the lesion (honeycomb or tennis racket appearance)[2], tooth resorption or displacement was common seen[3,4]. In Thailand, Odontogenic myxoma including odontogenic myxofibroma in jaw bone was reported only 3.1% of odontogenic tumors[1]. Mean age was 28.78 ± 12.56 years[1]. Also, A slight predilection of female than in male (1.3 :1) [1]. This result showed similar to the prevalence of odontogenic myxofibroma from previous study[5]. Since OF was rarely reported in Thailand and limited study of OF around the world, the present study report case of OF found in Thailand including diagnosis and treatment approach.

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Case Report

A 50-year-old Thai male was referred to dental department, Thammasat university hospital because of a radiolucency was found accidentally from panoramic film for 3 days. He had no known disease and drug allergy. Not any symptoms, e.g., pain, swelling, hypoesthesia was found. No history of trauma of jaws.

He shows normal general appearance. Extraoral findings show normal skin color and temperature, no swelling, normal sensation, normal mouth opening and no lymphadenopathy. Intraoral findings show normal gingiva, no gingival swelling, no shallow vestibule, no sign of inflammation, no dental caries, lower anterior teeth are normal to vitality test, normal to percussion, no periodontal pocket.

From panoramic film shows corticated, well-defined margin, multilocular radiolucency with internal septate at the apical area of permanent mandibular left canine to permanent mandibular right central incisor (22-25) size 2x1 cm^2, heterogenous density, displace root of 22,23, intact lamina dura, no root resorption (Figure 1).

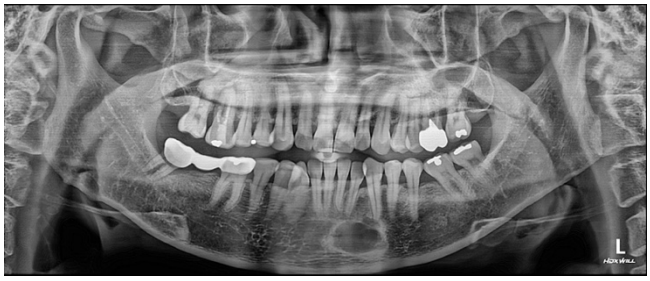

Figure 1. Panoramic film shows corticated, well-defined margin, multilocular radiolucency with internal septate at the apical area of 22-25 size 2x1 cm^2, heterogenous density, displace root of 32,33, intact lamina dura, no root resorption.

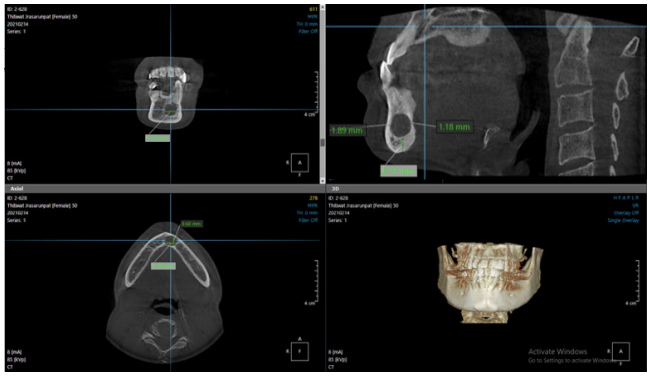

Figure 2. CBCT shows Cone beam CT was performed. From CBCT shows multilocular radiolucency with internal septate at mandible involved area from distal 22-mesial of 25 size MD12 x BL10 x H 10 mm^3, heterogenous density, displaced root of 22,23, no root resorption, no bony expansion and perforation. The thinnest part of buccal plate 2 mm and lingual plate 1 mm. The lesion distance to inferior border of mandible 6 mm.

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The differentiate diagnosis of multilocular radiolucency are varied. The odontogenic tumors like ameloblastoma which originated from odontogenic epithelium and odontogenic myxoma, central giant cell granuloma and odontogenic fibroma which originated from odontogenic ectomesenchyme was included[4,6]. Also, fibro-osseous lesion like early stage of Ossifying fibroma could be showed similar appearance[7].

Even if the incisional biopsy was recommended[4] but in this case consider its non-aggressive behavior, small size of lesion and no bony perforation, the prognosis seems favorable. The treatment plan was enucleation and curettage were performed instead of more aggressive like resection. Therefore, excisional biopsy was performed as definitive treatment. After local anesthesia, triangular flap was done. The full thickness flap was reflected. Since there is no bony perforation, reduction of bone was done. Then, the lesion was found and enucleation from bony wall. The lesion is round shaped, smooth surface, pale pink in color, tumor mass size 3x1x1 cm^3, firm consistency (Figure 3). The mass was sent for histopathological examination.

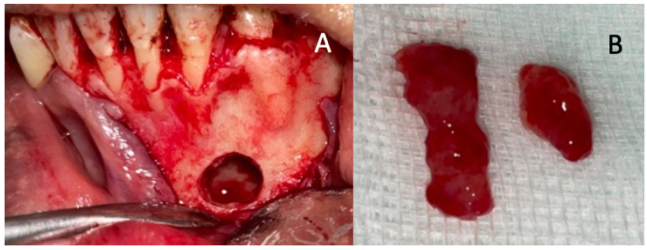

Figure 3. shows a bony cavity after tumor mass was removed (A). The lesion is round shaped, smooth surface, pale pink in color, tumor mass size 3x1x1 cm^3, firm consistency (B).

The result confirmed odontogenic myxofibroma. From Hematoxylin and Eosin staining showed loosed fibromyxomatous connective tissue containing haphazardly

arranged stellate, spindle shaped, and round cells. Numerous small blood vessels and moderate chronic inflammatory cell infiltrate consisting of lymphocytes and plasma cells are seen (Figure 4 A-D). Histopathological of OF may similar to other soft tissue myxofibroma such as myxoid nerve sheath tumor[8]. Therefore, S-100 s100 immunohistochemical staining was performed to differentiate nerve sheath tumor, which positive to s-100 immunohistochemical staining. In this case, the result showed negative to S-100 (Figure 4 E-F).

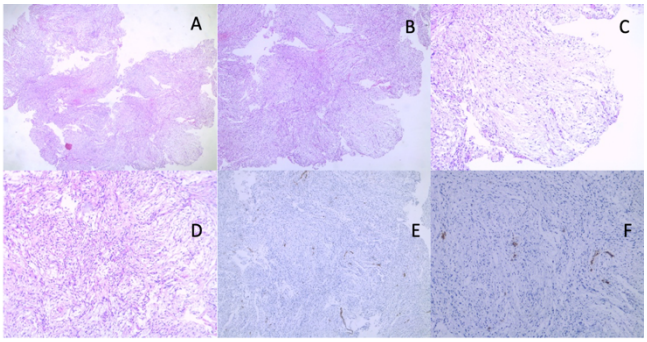

Figure 4. Shows loosed fibromyxomatous connective tissue making a feature of delicate fibrillar structure containing haphazardly arranged stellate, spindle shaped, and round cells. (H&E staining) (A-D) and Negative to S-100 (E-F).

In this case, there is no recurrence after 1 year treatment from panoramic film and CBCT. As the period check-up was an important part of treatment and the previous study showed the recurrence tend to occur in 2 years[5], at least 3 years follow-up period was preferred.

Discussion

In the present case, OF was originated in jaw bone like the previous study showed most of OF was a central type (91.7%)[5]. Slightly predilection in female (male to female ratio 1:1.4). Average age was 29.5 years. OF was slightly more common in mandible than maxilla and tend to occur in posterior areas[5,8]. However, in the present case OF was found in the anterior region of the mandible. Clinical symptoms of OF that commonly found was swelling (92.86%) some patients may experience various degrees of pain (35.71%) and root resorption or root displacement (28.57%)[5]. Myxofibromas grow slowly without causing any symptoms. Because they enlarge painlessly, they can reach a considerable size prior to being noticed. Lesions can expand the bone, but they perforate the cortex only if they reach a great size[8]. In the present case, there was no swelling or any symptoms reported. The lesion was accidentally found from panoramic film. Radiographic finding was multilocular radiolucency which was commonly found in OF. Histopathological findings showed loosed fibromyxomatous connective tissue containing stellate, spindle shaped, and round cells which may similar to myxoid nerve sheath tumor. Therefore, S-100 immunohistochemical staining was performed and showed negative to S-100.

Treatment options for OF were conservative treatment (enucleation and curettage) and radical surgery including En-bloc and partial resection. In 2017, Wright et al. advised that treatment choice is resection with free margins however small lesions can be treated with conservative surgery with the expectation of a low risk of recurrence[9]. Although most clinically evident recurrences occur within 2 years, in some patients, a recurrence was discovered several years after surgery; therefore, long-term follow-up is recommended[10].

Conclusions

Even OF was rarely found but the dentist should keep mind and could be one of differential diagnosis so that we can make a proper management. The early detection in this case importantly helped dentist to manage with lesions before its get expansion so that can be treated conservatively. These can reduce morbidity and made better patient's quality of life.

Declaration of Interest

The authors report no conflict of interest.

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