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Letter to Editor

Adolescent Cherubism

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Dear Sir,

I came across an interesting case, a young 23-year-old man was referred to me for the assessment and removal of wisdom teeth and associated Orthopantomograph (OPG) findings. He reported that he was medically fit and well, a university student. The wisdom teeth were asymptomatic.

On examination, teeth 18, 28, 38 and 48 were erupted though difficult to clean. He had marked buccal expansion bilaterally in the region of the premolars extending posteriorly and to the lower border of mandible. His occlusion was normal and he reported normal sensation in the mental nerve bilaterally.

OPG showed multiple radiolucent lesions bilaterally in the ramus and body of the mandible (Figure 1). On CT, there were extensive bilateral expansive lesions extending from second molar tooth to the angle of mandible on each side. He had orthodontic treatment in the past. His cone beam CT shows a multilocular radiolucent lesion in the buccal aspect of the tooth 35 to 38 and similarly from tooth 46 to 48 (Figure 2).

My initial clinical impression was Fibro-osseous lesion most likely cherubism, but he was not very forthcoming in regard to his past history. It was decided to contact his parents with the patient's consent to facilitate the development of a comprehensive a past medical history.

The patient's mother advised that at the age of eight he was diagnosed as cherubism, based on mild bilateral swelling of lower face and multilocular lesions in the mandible on OPG. She was not able to provide any photographs and/or x-rays. The specialist who diagnosed him with cherubism confirmed that he had multilocular lesions in his mandible, but the condition was not full blown, and he was kept on observations. As the patient had been lost to follow-up, he had no further information. His Orthodontist provided OPG from 2016 which showed the lesions are regressing compared to recent OPG in 2019 (Figure 3). Patient was advised for regular follow-up and repeat OPG and/or CBCT.

Cherubism is a rare, non-neoplastic, self-limiting fibro-osseous disease, characterised by painless expansion of the mandible or maxilla, or both. Children are normal at birth and the expanding jaw is noticed within the first years of life, becoming progressively larger until the beginning of adolescence. Its usual course results in regression of the lesion during puberty, then stabilisation and finally regression in early adulthood. Unfortunately, documented reports with long-term follow-up are rare [1].

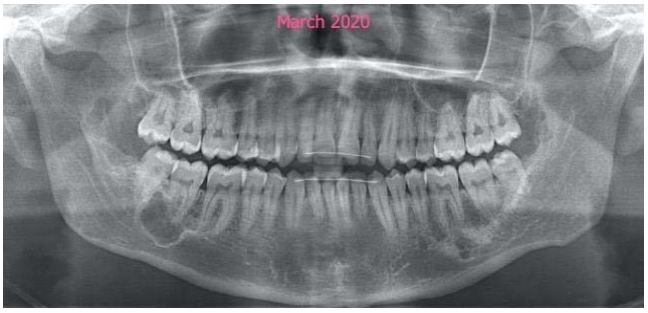

Figure 1: Orthopantomogram showing bilateral multiple radiolucencies involving body and ramus of the mandible.

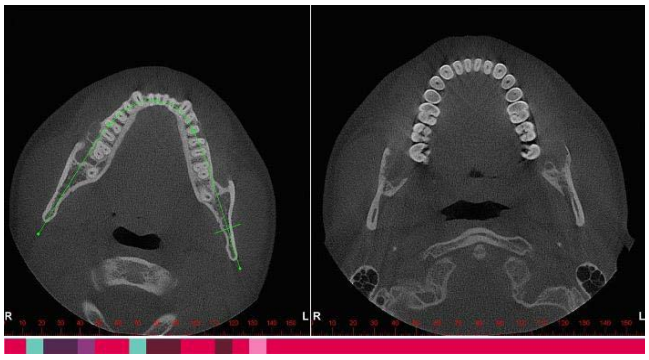

Figure 2: CBCT axial cut showing bilateral radiolucent lesions.

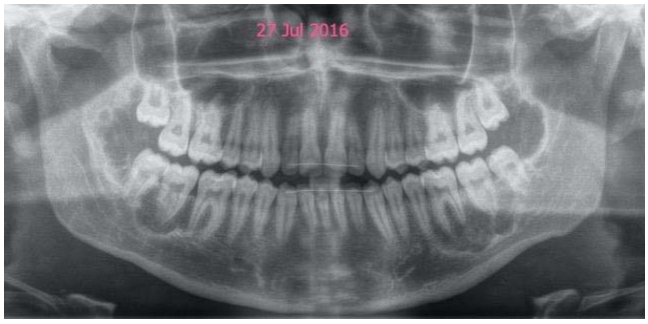

Figure 3: Orthopantomogram done in 2017 showing bilateral radiolucencies involving mandible.

Cherubism was first described in 1933 by Jones as familial multilocular cystic disease of the jaws, but the term cherubism was given to describe the rounded facial appearance resulting from jaw hypertrophy. The condition was initially characterised as familial, but both hereditary and sporadic cases have since been described. A mutation of the SH3BP2 gene from chromosome 4p16.3 has been found in75% of the reported cases.

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Bilateral mandibular involvement is a distinguishing feature, the lesions characteristically continue a pattern of variable enlargement until puberty, and then they partially or fully regress and show sclerotic involution in adulthood. Cherubism is similar to fibrous dysplasia radiographically, especially when the latter is confined to the jaw. The similarity in appearance has prompted the suggestion that cherubism may a be familial form of fibrous dysplasia localized to the jaw bones [2,3].

Radiographic differential diagnosis for cherubism includes craniofacial fibrous dysplasia, brown tumor of hyperparathyroidism, Jaffe-Campanacci syndrome and familial gigantiform cemeto-osseous dysplasia. Features more specific to cherubism include bilateral mandibular involvement, limited to maxilla and involution at the time of puberty. In contrast, patients with fibrous dysplasia typically do not present with cherubic appearance, swollen cheeks, upwards turning of eyes, or dental derangement. Histologically, patients with cherubism typically have a prominent number of multinucleated giant cells, which are rarely seen in fibrous dysplasia. Brown tumor and Jaffe-Campanacci syndrome are distinguished on clinical grounds and are easily eliminated from the differential diagnosis. Gigantiform cementoma lesions are located primarily in maxilla and are enlarged in a focal rather than diffuse manner. Histologically, cementomas contain cemental like tissue and lack multinucleated giant cells and vascularity [1-3].

The case reported here looks normal clinically and his radiological lesions are getting regressing. The patient was referred for wisdom teeth based on radiographic findings due to lack of proper history given by patient during the examination. Comparing OPG done in 2016 and 2020 showed the radiolucencies were getting involuted.

References

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