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Case Report

Gargantuan infected complex odontome of the maxilla: A case report and a brief review

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ABSTRACT

Odontomas are the most common benign tumours of odontogenic origin and are characterised by their slow growth and non aggressive behaviour. Odontomas are classified as complex, when the calcified dental tissues are simply present as an irregular mass bearing no morphologic similarity even to rudimentary teeth, or compound, if there is superficial anatomic similarity to even rudimentary teeth. A few odontomas have a mixture of compound and complex elements that consist of miniature well formed teeth associated with disorganised sheets of tooth structures. An event of huge odontoma intruding the maxillary sinus, emerging into the oral cavity because of expansive growth, illustrating both complex and compound elements is unusual. We describe such a treated case of an infected large complex odontoma, in which the lesion has breached the alveolar bone and oral mucosa in the area of the right maxillary molars in a 16-year-old male.

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1. Introduction

The World Health Organisation (WHO) classifies odontomas as a benign odontogenic tumour composed of odontogenic epithelium and odontogenic ectomesenchmye with dental hard tissue formation [1]. Broca first coined the term "odontome" in 1866 and defined it as a tumour formed by an overgrowth of complete dental tissue. Later, authors defined odontoma as a tumour that has developed and differentiated enough to produce enamel and dentin. But tooth-like material is laid down in an abnormal pattern because of failure of the progenitor odontogenic cells to reach a full state of morphodifferentiation [2]. Although the WHO continues to classify these lesions as tumours, most investigators consider them as hamartomas. The WHO defines a complex odontoma as a malformation in which all of the dental tissue are represented and individual tissues mainly are well formed but occur in a disorderly pattern [2]. The compound type is described as a malformation in which all of the dental tissues are represented in a more orderly

Odontomas, especially complex, may become large and produce expansion of bone and facial asymmetry [2]. Large odontomas, however, measuring up to 6 cm or more in size and causing jaw expansion have been reported in literature. The largest odontoma found in human weighed 0.3 kg [4]. Eruption of a giant odontoma in the oral cavity is rare [5,6]. This situation can cause pain, inflammation of the adjacent soft tissues, or infection associated with suppuration. This paper describes an unusual case of a huge infected complex odontoma with an impacted tooth which produced remarkable expansion of the cortical bone, obscuring the maxillary sinus and emerging into oral cavity because of its magnanimous growth. The lesion was totally removed through a conservative intraoral approach.

2. Case report

A 16-year-old male presented with a history of slowly enlarging, painful swelling in the right upper back teeth region since 2 years, with pus discharge. Pain was relieved on medication. The patient was otherwise healthy with no significant medical history.

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pattern than in the complex, so that the lesion consists of many tooth like structures [3]. Compound odontomas are more common than the complex. The most common location for compound odontomas is the anterior maxilla and for the complex, it is the posterior mandible or maxilla [2].

[☆] AsianAOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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Fig. 1. Pre-operative intraoral view showing yellowish calcified mass erupting through the oral mucosa. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

Extraoral examination revealed a diffuse swelling in the right mid face region, with the overlying skin being smooth, stretched and pinchable. On palpation, the swelling was tender, firm to hard in consistency, fixed to the underlying structures with localised rise in temperature. Mouth opening was adequate and with no palpable lymph nodes. Intraoral examination revealed a solitary mass, approximately 4 × 3 cm in dimension, medially extending up to the midline of the palate, involving the vestibular region laterally, up to the first premolar anteriorly, and extending up to maxillary tuberosity posteriorly. The mucosa distal to tooth 14 and up to the tuberosity area was breached by a large yellowish-brown hard mass, which resembled avascular bone, measuring 2×2.5 cm. The hard mass was seen in the region of the clinically missing maxillary second premolar and maxillary molars with the expanded buccal and palatal cortices (Fig. 1). The mucosa around mass was inflamed with evidence of pus discharge and bleeding. On palpation, the swelling was hard in consistency, tender near the vestibular regions, margins appeared well defined from the adjacent structures. Swelling was fixed to underlying structures. Orthopantamograph (OPG) revealed an extensive predominantly radiopaque mass with areas of radiolucency with well-defined borders, seen on the right maxilla measuring about 5×4 cm. The radiopaque mass was amorphous, circumscribed by a thin, irregular radiolucent halo (Fig. 2A and B). The OPG revealed a well circumscribed radio-opaque mass anteriorly involving premolar region and posteriorly up to maxillary tuberosity superiorly extending up to floor of the orbit and inferiorly up to the maxillary alveolus encroaching the right maxillary sinus with impacted 15 and missing right maxillary molars. The case was provisionally diagnosed as odontome with impacted 15. Based on clinical presentation, radiographic findings and clinical course of lesion following entities were considered in the list of differential diagnosis like focal chronic sclerosing osteomyelitis, odontogenic tumours like odontoameloblastoma, ameloblastic fibroodontome and cementoblastoma, fibro-osseous lesions like cemento-ossifying fibroma, cemento-osseous dysplasia, familial gigantiform cementoma, benign bone tumour like sequestrating osteoma. Decalcified sections of incisional biospy showed haphazardly arranged dental tissue mainly consisting of tubular and globular dentin enclosing pulp spaces, empty enamel spaces, some odontogenic epithelium and ectomesenchyme elements were noted. This was suggestive of odontogenic tumour probably an exuberantly grown, infected odontoma breaching the alveolar bone and oral mucosa. Digital volume tomography (DVT) was done to know the extension of the lesion. Limited volume tomography revealed a field of view of 5×3 cm, showing an amorphous mass of mixed densities. Axial view disclosed a radiolucent zone surrounding the amorphous mass. Curvilinear view demonstrated impacted 15 in relation to the mass (Fig. 2C). The mass was surgically removed by giving a crevicular incision extending from 21 to the right maxillary tuberosity (Fig. 3A). A full thickness mucoperiosteal flap was raised and the mass was removed completely. After haemostasis, primary closure was achieved by advancing the flap buccally. Gross examination consisted of a large hard tissue specimen (Fig. 3B and C) with attached soft tissue fragments and a tooth. The large hard tissue specimen measured approx $5 \times 4 \times 3$ cm in size, with an irregular surface and stony hard in consistency. The surface of hard tissue showed the presence of round to elongated calcified structures which was whitish to creamish in color. Ground sections of the large hard tissue mass shows haphazardly laid enamel, dentin, and focal cementum-like tissue (Fig. 4A and B). Decalcified sections show denticles embedded in a fibrous connective tissue. Microscopic examination showed a grid of tubular dentin with holes containing enamel matrix and pulp spaces

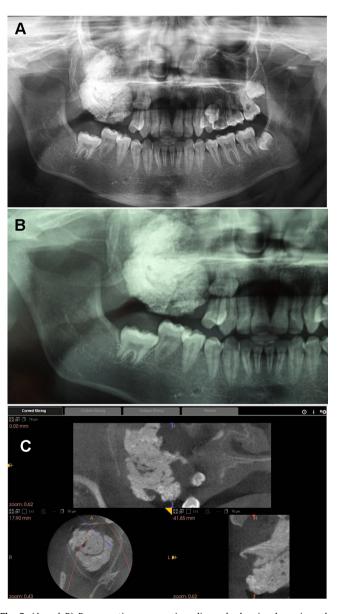


Fig. 2. (A and B) Pre-operative panoramic radiograph showing large irregular radiopaque mass having density similar to enamel and dentin. The mass was occupying right maxillary tuberosity and encroaching sinus. With impacted and displaced 15. (C) Digital volume tomography showing the extension of the lesion. Curvilinear view demonstrated impacted 15 in relation to the mass.

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