Case Report

Multiple Complex Odontomas of the Mandible: A Rare Case Report and Literature Review

Abstract

A 53-year-old female appeared with pain in the right mandible ramus, for the past 9 months, after tooth extraction. Clinical and radiological examination using conventional and advanced computerized tomography diagnostic imaging led to a provisional diagnosis of multiple complex odontomas. Complete conservative excision of the lesion was performed. The clinical diagnosis was confirmed histopathologically. Postoperative period was uneventful with no evidence of recurrence. According to an extensive literature review, this report describes the oldest patient ever diagnosed with multiple odontomas in the literature.

Keywords: *Complex odontoma, hamartoma, mandible, multiple odontomas, odontogenic tumor*

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Introduction

Odontomas are mixed odontogenic tumors since they are composed of both epithelial and mesenchymal dental hard tissues.[1,2] Single odontomas are one of the most common odontogenic tumors and can be classified as compound or complex. Complex odontoma is a hamartoma in which enamel and dentin, and sometimes cementum and pulp, are present.[2] It is primarily diagnosed in children, adolescents, and young adults. Both compound and complex odontomas mostly occur as solitary lesions in the jaw. On the other hand, multiple odontomas (MOs) are characterized by numerous odontomas involving from one to all four quadrants of the jaws. A literature review disclosed 15 cases of MOs, of them 9 were complex. Because it is a rare pathologic entity, little is known about clinical features of MOs. This report describes a rare case of multiple complex odontoma in a 53-year-old female. This is the first report of MOs in a 53-year-old patient.

Procedure

A 53-year-old female patient presented with a noncontributory medical and dental history, and there was no significant medical history. Her chief complaint was pain in the right mandible ramus, for the past 9 months, after tooth extraction. The intraoral examination showed swelling on the buccal side of the right horizontal body of the mandible with permanent mandibular right first premolar mesially displaced. The swelling was hard on palpation, the overlying mucosa was normal, the adjacent premolar had mobility, and there was no neck lymphadenopathy.

Panoramic radiography and computerized tomography [Figures 1-3] revealed two well-defined radiopaque lesions on the right mandible's body. The posterior lesion slightly expanded the premolar and molar external and lingual mandibular wall with thinning of the lingual cortex and small disruption of the buccal cortex. The major lesion measured 31 mm × 23 mm in the larger sectional axes. The remaining path of the inferior alveolar nerve canal seemed to be below the lesion. Both lesions were intraosseous and independent. The anterior one slightly disrupts and surrounds the mental foramen. Taking into account the pathologic and radiologic presentations, a provisional diagnosis of multiple complex odontomas was made. Compound odontoma, osteoma, ameloblastic fibro-odontoma, and cemento-ossifying fibroma were considered for differential diagnosis.[1,2]

It was performed a total conservative excision procedure under general anesthesia

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[Figures 4 and 5]. The posterior lesion was excised *en bloc*. It was not possible to maintain the second left lower premolar due to the lesion extension and periodontal status. The anterior lesion was separated into two portions to enable the excision [Appendix 3]. After excision, a thorough curettage was made, and the lesions were submitted for histopathological analysis.

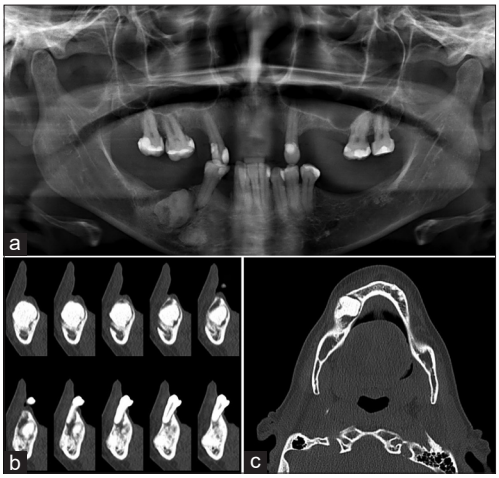

Figure 1: Preoperative panoramic radiograph and computerized tomography images. (a) Panoramic radiograph. (b) Coronal plane. (c) Axial plane

As a potential rare pathological entity in a 53-year-old patient, histopathological analysis was made in two independent laboratories. Both laboratories confirmed the diagnosis of complex odontoma.

No evidence of recurrence was observed at 14-month follow-up. The patient was requested to make regular follow-up visits to our hospital.

Discussion and Conclusion

In the past few years, numerous epidemiological studies were made on the prevalence of odontogenic tumors. According to the published greatest series, odontomas are one of the most common odontogenic tumors, along with ameloblastoma and keratocystic odontogenic tumor.[3-7] In spite of the high prevalence of odontomas, MOs are extremely rare in humans with an unknown prevalence.[8] A review of the English language literature found 15 cases

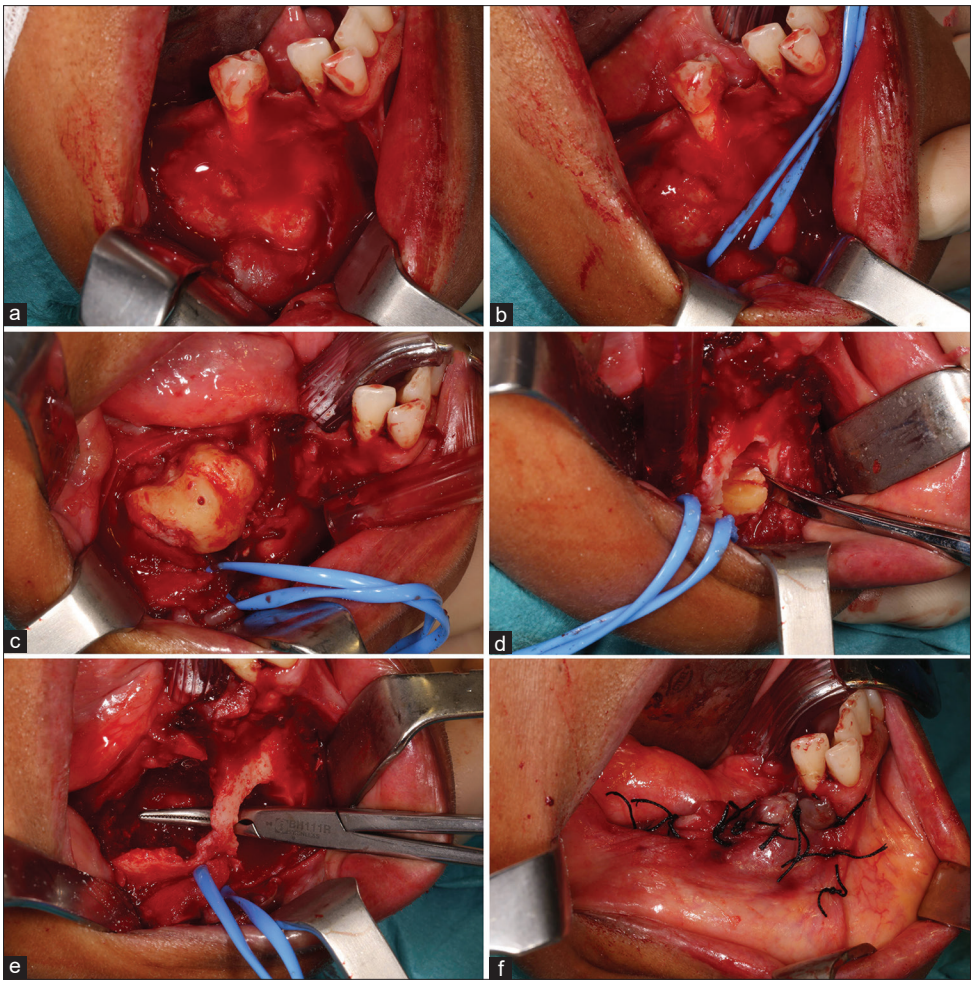

Figure 2: Excision procedure. (a) Exposure of both lesions. (b) After nerve localization, it was wrapped with a silicon tube. (c) Excision of the posterior lesion *en bloc* and extraction of the second left lower premolar. (d) Separation of the anterior lesion in two portions to ease the excision. (e) After removing both lesions, it was possible to preserve a thin portion of buccal bone. (f) Sutures and final aspect

Table 1: Multiple odontomas reported in the literature

Author (year)	Gender/ age	Location	Radiographic diagnosis	Histological diagnosis	Size	Other abnormality	Treatment
Bader 1967	Female/0-5	4 quadrants	Compound and complex	Confirmed	Extensive in maxilla; localized in mandible	Stenosis of esophagus	Two-staged operation
Schmidseder and Hausamen 1975	Male/4	4 quadrants	Compound	Confirmed	Localized	Stenosis of esophagus	Two-session surgical excision
Melnick 1975	Male/20	4 quadrants	Compound	Confirmed	Localized	No	Excision
Iwamoto 1999	Female/15	2 quadrants (Bilateral mandible)	Compound and developing complex	Confirmed	Localized	No	Excision
Ajike 2000	Female/15	4 quadrants	Compound	Confirmed	Extensive	No	Excision
Bordini 2008	Male/17	4 quadrants	Compound	Confirmed	Extensive in right maxilla, localized in other 3 quadrants	No	Excision
Hammoudeh *et al*., 2009	Female/4	2 quadrants (Bilateral mandible)	Complex	Confirmed	Localized	Pierre-robin syndrome	Excision
Maleewong *et al*., 2011	Male/15	4 quadrants	Complex	Confirmed	Extensive	No	Excision
	Female/19	4 quadrants	Complex	Confirmed	Extensive	No	Excision
Srivastava 2012	Female/42	2 quadrants	Complex	Confirmed	Localized	No	Excision
Kumazawa *et al*., 2014	Female/4	4 quadrants	Complex and Compound	Confirmed	Extensive	No	Excision
Erdogan 2014	Male/27	4 quadrants	Compound	Confirmed	Extensive	Mild mental retardation sight disorder, severe myopia	Partial excision
Guledgud *et al*., 2014	Male/13	4 quadrants	Complex	Confirmed	Extensive	No	Excision
Gujjar *et al*., 2015	Male/45	4 quadrants	Compound	Confirmed	Localized	No	Excision
Liu *et al*., 2017	Female/9	1 quadrant (left mandible)	Complex	Confirmed	Localized	Otodental syndrome	Excision
Botelho *et al*., 2018	Female/53	1 quadrant (right mandible)	Complex	Confirmed	Localized	No	Excision

of MOs [Table 1],[8-20] of them only 9 were complex odontomas. The cases presented by Browne,[21] Malik and Khalid,[22] and Mani[23] were not included in this analysis due to the absence of a histological diagnosis. MOs are mainly diagnosed in the first two decades, with only one case of a 42-year-old female.[16] Therefore, this rare case in a 53-year-old female patient represents the oldest patient ever diagnosed with MOs and the tenth case of multiple complex odontomas described.

In general, odontomas are asymptomatic, although they can appear in any age they are primarily found in the second decade, and they are not gender related.[24] The most prevalent symptoms are impaction of permanent tooth, swelling of the jaw, and adjacent teeth displacement. Pain and tooth malpositioning are not so common.[24] In this case report, the patient complained of jaw's swelling and pain that started after the extraction of the lower right first molar, 9 months before her first appointment with us.

The etiology of odontomas remains unclear. However, they have been associated with environmental (traumas, infections, or inflammation) and genetic causes (cleidocranial dysostosis, Gardner's syndrome, Hermann's syndrome, and Pierre–Robin syndrome).[14,25-27] Recently, a study suggested a possible genetic etiology for MOs since the partial duplication of chromosome 11 q13.3 may confer a gain of function of the FGF3 and FGF4 genes.[28] In this case, there was no familiar history, and she was the first known case of MOs in the family.

Radiographically, complex odontomas appear as a spherical or ovoid radiopacity with a fine radiating periphery, surrounded by a radiolucent zone, which may be broader in a developing complex odontoma. Differential diagnosis from a compound odontoma or even an osteoma may not be possible radiographically.[2]

Mandible multiple complex odontomas constitute a therapeutic challenge,[29] since, in some cases, lesions are located nearby vital anatomic structures. In this case, to prevent the injury of the mental nerve, after the detachment and lesion recognition, we localized and wrapped the nerve with a silicon tube [Figure 5]. Thus, we readily

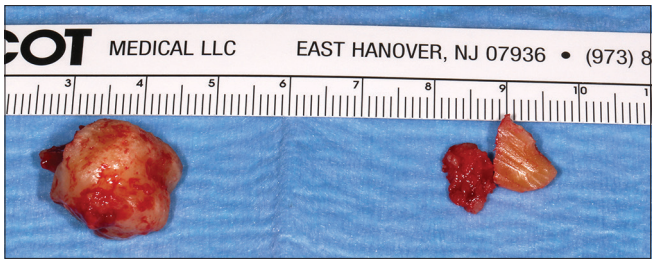

Figure 3: Gross specimens excised

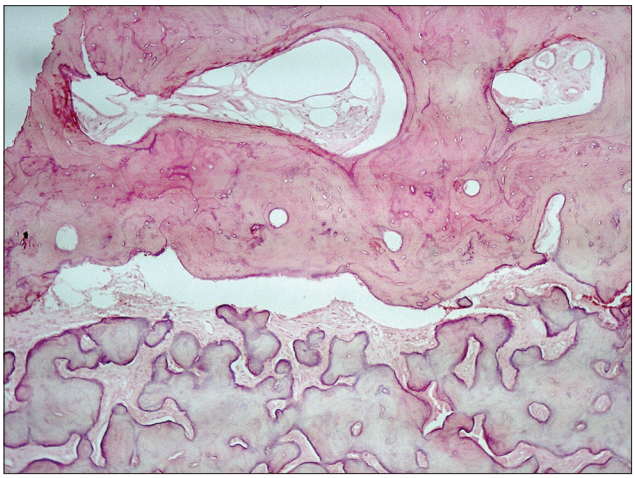

Figure 4: Multiple complex odontomas, microscopic findings. The core exhibits decreased cellularity, smaller fibroblastic nuclei, dispersed and amorphous dentin, dispersed enamel and cementum, and low vascularization (H and E, ×400)

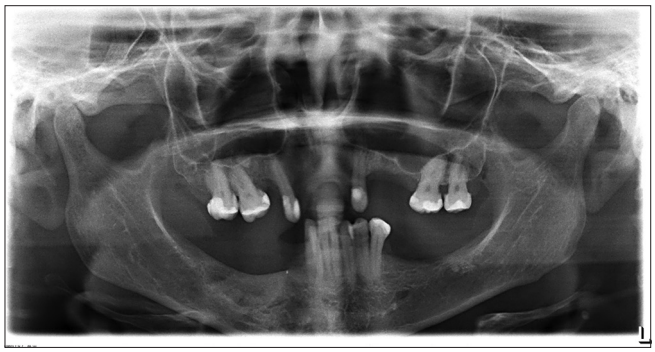

Figure 5: Panoramic radiograph of 14-month follow-up showing no recurrence

identified this structure during all surgeries, decreasing the postsurgical risks. The patient had no complication after surgery.

In conclusion, after an extensive literature review and in light of the existing information, this report describes the oldest patient ever diagnosed with MOs in the literature.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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