Case Report

Large erupted complex odontoma in posterior mandible: a rare case report

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ABSTRACT

Introduction: Complex odontomas are hamartomas composed of randomly arranged dental tissues like enamel, dentin, cementum, and pulp. They typically have limited growth, with only 4.3% measuring more than 3 cm in size. Erupted odontomas, defined as intraosseous odontomas that erupt into the oral cavity, are exceptionally rare. This case report presents a rare case of a large complex odontoma in the posterior mandible of a 25-year-old female. **Case report**: A 25-year-old female patient was referred to the Radiology Department with a chief complaint of left jaw swelling that had persisted for 6 months. Facial asymmetry was noted due to a swelling in the left jaw. Intraoral examination revealed a yellow-brown, irregular, hard mass resembling dental calculus. Radiographs showed a well-defined, irregular radiopaque mass approximately 6 x 3 cm in size, surrounded by a radiolucent rim. The opacity was similar to the density of enamel and dentin in certain regions of the lesion. The mandibular left molars were missing. Radiographic findings confirmed a diagnosis of odontoma. The patient was then referred to the Department of Oral Surgery for further management and surgical intervention. **Conclusion**: Odontomas rarely erupt, but their eruption and large size can lead to significant complications like pain, inflammation, infection, and facial asymmetry. Dentists should be familiar with the clinical and radiographic characteristics of odontomas to ensure prompt and effective management.

KEYWORDS

Odontoma, erupted complex odontoma, large odontoma, mandible, panoramic radiograph

INTRODUCTION

Odontomas are a type of benign odontogenic tumor composed of epithelial and ectomesenchymal odontogenic tissues.[1] They are the most common odontogenic tumors, consisting of enamel, dentin, cementum, and pulp.[2] However, some researchers consider odontomas to be hamartomas rather than true neoplasms, as they exhibit well-differentiated dental tissues and limited, gradual growth.[1–3] The term "odontoma" was first introduced by Paul Broca in 1867 and was later classified by Thoma and Goldman in 1946.[4] Currently, the World Health

Organization classifies odontomas as benign mixed epithelial and mesenchymal odontogenic tumors, along with ameloblastic fibro-odontoma, which is regarded as an immature precursor of complex odontoma.[5] Odontomas are frequently asymptomatic and are often discovered during routine radiographic examinations, usually when investigating issues related to tooth eruption, malformation, impaction, or delayed eruption.[6,7]

According to the World Health Organization classification, odontomas are divided into two subtypes based on their radiographic, histologic, and clinical characteristics: complex and compound. Compound odontomas exhibit organized dental tissues and may contain tooth-like structures known as denticles, whereas complex odontomas consist of randomly arranged dental tissues.[3] On panoramic radiographs, complex odontomas appear as homogeneous, radiopaque lesions with an oval or irregular shape.

The lesion density is greater than that of the surrounding bone and tissue. Additionally, a well-defined radiolucent rim surrounds the radiopaque lesion, representing the connective tissue capsule.[8] Odontogenic tumors have a prevalence ranging from 1% to 32%[1], with odontomas accounting for the majority at 67% and complex odontomas comprising 33% of cases.[6,7] These lesions can occur at any age, with a peak incidence in the second decade of life. Patients typically ranges from 6 to 46 years old, with an average age of 23. There is no apparent predominance in the sex distribution of these lesions, although some studies report compound odontomas being more common in males and complex odontomas slightly more prevalent in females.[1,2]

The etiology of these tumor lesions remains unclear.[1] Several theories have been proposed regarding their cause, including localized trauma during primary dentition development, infectious and/or inflammatory processes, hereditary abnormalities, and genetic alterations that influence tooth formation.[2,6,7] All odontomas exhibit a degree of histodifferentiation, resulting in the formation of recognizable dental tissues. Compound odontomas also demonstrate morphodifferentiation, whereby they resemble the shape of teeth.[3] In contrast, complex odontomas are an agglomerate of all dental tissues, displaying abnormal morphodifferentiation alongside normal histodifferentiation.[4]

Odontomas typically exhibit limited growth, with the largest dimension generally reaching up to 3 cm. Nonetheless, in rare instances, sizeable odontomas, commonly referred to as "large odontomas," which exceed 3 cm in diameter, have been observed on panoramic radiographs.[1,9] A study conducted by Miki et al. reported that only 4.3% of odontomas were larger than 3 cm.[2] Odontomas with extensive jaw involvement are infrequently reported.[6] Notably, most large odontomas were classified as complex odontomas and occurred predominantly in patients under 30 years of age.[9]

These lesions are characterized by their expansive growth, which can result in cortical bone expansion, facial asymmetry, and displacement or impaction of adjacent teeth. The literature indicates that all reported cases of large complex odontomas have shown a tendency to expand the buccal and/or lingual bone plates, with a significant percentage being associated with facial asymmetry.[10]

Junquera et al. clinically categorized odontomas into three types: 1) intraosseous odontomas, 2) extraosseous odontomas occurring solely in the soft tissue, and 3) erupted odontomas resulting from an intraosseous odontoma's eruption into the oral cavity due to its location coronal to an impacted/erupting tooth or its superficial position within the bone.[2,4] Central odontomas are the most prevalent odontogenic tumors.[7] Peripheral odontomas are limited to the soft tissue covering the mandible and maxilla. Erupted odontomas, which are intraosseous odontomas that exceptionally emerge in the oral cavity, are exceedingly rare occurrences. The eruption of odontomas was observed in only 1.6% of all odontoma cases.[2]

Complex odontomas rarely erupt into the oral cavity.[11] However, in some cases, an odontoma may spontaneously erupt. To date, more than 50 cases of erupted odontomas have been reported. Most of these patients are women in their second or third decades of life.[12] Furthermore, the surgical management of these lesions requires careful planning to avoid damage to vital structures, such as the inferior alveolar nerve particularly in mandibular cases.[13,14]

The presented case is unique due to the large size and posterior mandible location of the erupted complex odontoma, which is uncommon. Odontomas typically present in the maxilla, and their eruption is relatively rare, while their similarities to other radiopaque lesions such as cemento-ossifying fibroma, cemento-osseous dysplasia, ameloblastic fibro-odontoma, osteoid osteoma, and odonto-ameloblastoma make this case an interesting addition to the literature.This case report presents an uncommon case of a large complex odontoma in the posterior mandible of a woman.

CASE REPORT

A 25-year-old female in generally good health presented to the Radiology Department with a chief complaint of left-sided jaw swelling that had been present for the past 6 months. Extraoral examination revealed facial asymmetry caused by swelling on the left side of the jaw. Extending from the corner of the mouth to the posterior border of the ramus and from the orbital rim to the inferior mandibular border. No signs of inflammation were observed (Figure 1).

Figure 1. The extraoral photograph demonstrates facial asymmetry due to swelling

Intraoral examination revealed a yellow-brown, irregular, hard mass measuring approximately 1.5 cm x 2 cm, resembling dental calculus, located in the left posterior mandibular region, distal to the second premolar. The first, second, and third molars were not clinically visible. Ulcers were observed on the mucosa posterior to the mass, likely caused by chronic irritation due to occlusion

with the maxillary teeth. Upon palpation, the lesion was firm and asymptomatic (Figure 2).

Figure 2. The intraoral photograph shows a swelling with an ulcer

Panoramic, lateral cephalometric, and posteroanterior cephalometric radiographs were performed. The radiographs revealed a well-defined, irregular radiopaque mass measuring approximately 6 x 3 cm, exhibiting anteroposterior expansion from the distal premolar to the left mandibular ramus, and superoinferior expansion from the occlusal plane to the inferior mandibular cortical bone. Lateral expansion of the lesion was evident on the posteroanterior view, resulting in mandibular asymmetry.

The radiopaque mass was surrounded by a radiolucent rim anteriorly, posteriorly and inferiorly, with a corticated border, particularly in the anterior region. Cortication was absent at the superior aspect of the mass. The degree of opacity in some regions resembled the density of enamel and dentin. Inferior displacement of the mandibular canal was noted due to the lesion's expansion (Figure 3, 4, 5).

Based on the radiographic features of the amorphous radiopaque with a radiolucent border, a provisional diagnosis of complex odontoma was made. However, other radiopaque lesions such as cemento-ossifying fibroma, cemento-osseous dysplasia, ameloblastic fibro-odontoma, osteoid osteoma, and odontoameloblastoma were also considered in the differential diagnosis. Surgical enucleation was the recommended treatment approach for this lesion. The lesion was successfully enucleated without complication.

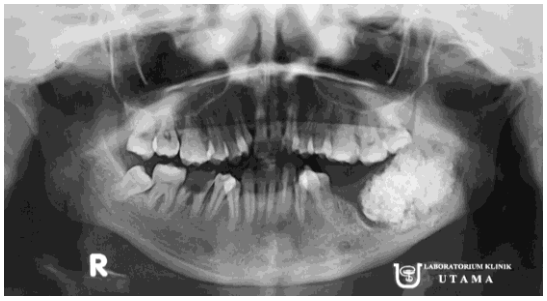

Figure 3. Panoramic radiograph shows as a well-defined radiopacity surrounded by a radiolucent halo with inferior displacement of mandibular canal

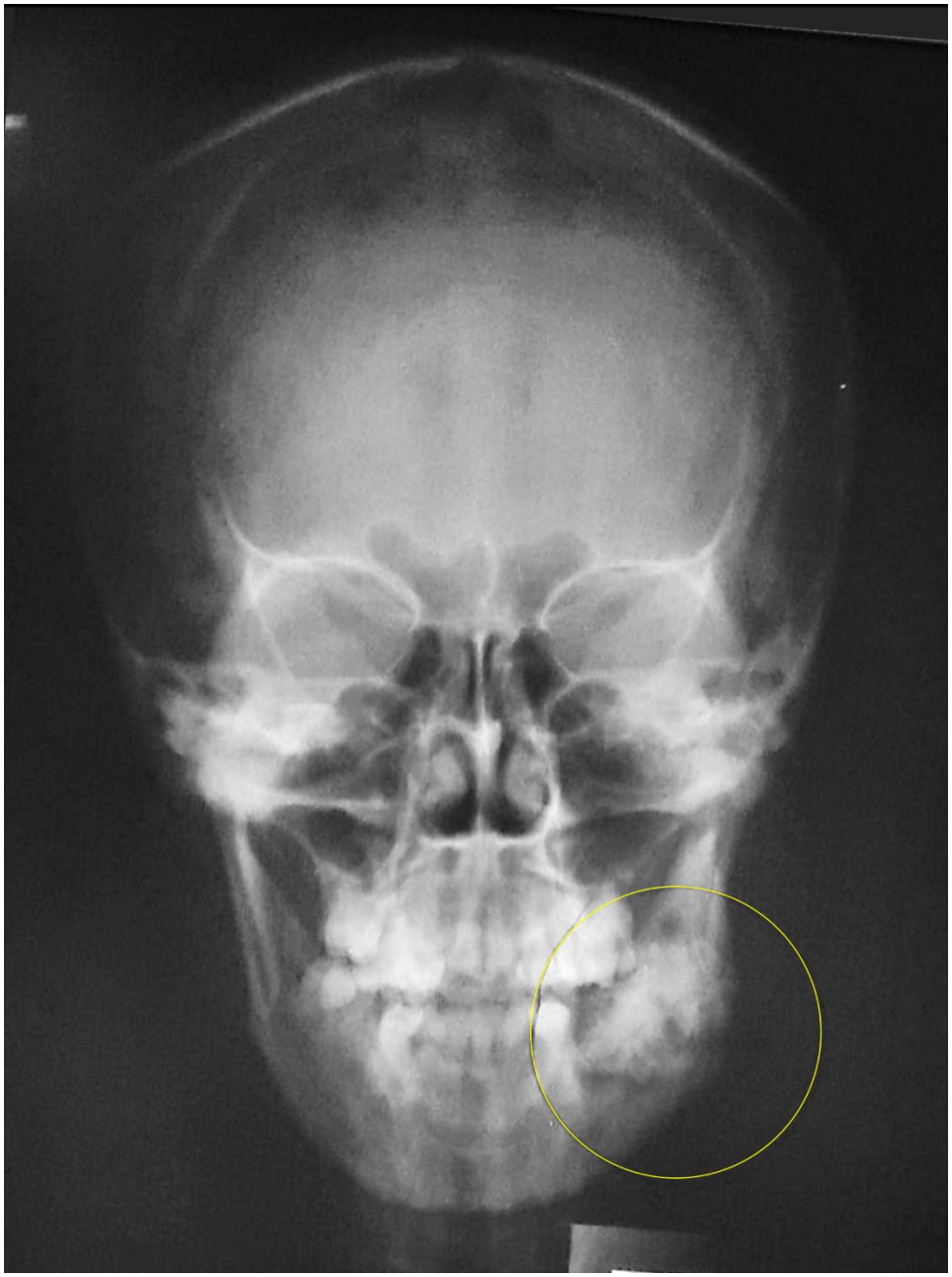

Figure 4. Posteroanterior cephalometric radiograph shows lateral expansion of the lesion causes mandibular asymmetry

Figure 5. **Lateral skull radiograph shows the lesion as a well-defined radiopacity surrounded by a radiolucent halo**

DISCUSSION

Complex odontomas are slow-growing, expansile, and typically asymptomatic lesions.[11] The reported frequency of complex odontomas ranges from 5% to 30% among odontogenic tumors. Although complex odontomas demonstrate a slight predilection for the maxilla, the current case occurred in the mandible.[2,11] Only 1.6% of complex odontomas erupt into the oral cavity.[4] Complex odontomas account for 42.4% of erupted odontomas. A comprehensive search of PubMed and Google Scholar covering the past 10 years (2014-2024) using the keywords "ERUPTED" "COMPLEX ODONTOMA" AND "MANDIBLE" revealed 14 reported cases of erupted odontomas in the mandible Of these, 6 measured 3 cm or larger, qualifying them as giant odontomas. This case represents one of the largest reported erupted complex odontomas. Erupted odontomas tend to occur more frequently in younger individuals, as exemplified by the 25-year-old patient in this case.[2] The exposure of a large erupted odontoma to the oral microflora may contribute as a predisposing factor for local inflammation.[15] Additionally, the rough surface of an erupted odontoma can promote dental plaque accumulation, and the absence of a periodontal ligament may facilitate the invasion of oral microorganisms into the surrounding bone.[5]

Table 1. A summary of erupted complex odontoma in mandible from 2014 to 2024.

No	Authors	Presenting time	Age	Sex	Location	Size	Symptoms	Associated with impacted teeth	Treatment
1	Pamukçu, et al.[2]	2021	21	male	Left posterior mandible	2.9 cm × 1.4 cm × 1.8 cm	swelling	yes	excision
2	Pamukçu, et al.[2]	2021	23	male	Left posterior mandible	1.3 cm × 1.5 cm × 1.8 cm	swelling and exudate	yes	excision
3	Leódido, et al.[6]	2015	18	female	Right posterior mandible	4.2 cm	swelling and pain	yes	excision
4	Bagewadi, et al.[11]	2015	22	male	Right posterior mandible	3.5 cm × 4 cm	swelling	yes	excision
5	Pró, et al.[7]	2016	22	male	Left posterior mandible	3.6 cm × 3 cm	swelling	yes	surgical removal
6	GV, et al.[12]	2022	17	male	Left posterior mandible	2 cm × 1 cm	swelling and extraoral fistule	yes	surgical removal
7	Katoumas, et al.[15]	2018	23	male	Left posterior mandible	2,5 × 2 × 2 cm	swelling	yes	surgical removal
8	Niazmand, at al.[16]	2019	18	female	Right posterior mandible	5 cm × 1 cm	intraoral swelling	yes	excision
9	Guledgud, et al.[17]	2014	13	male	Anterior mandible	2.2 × 1.8 cm	swelling and pain	no	enucleation
10	Ahmed, et al.[18]	2015	24	male	Right posterior mandible	N/A	pain and limited mouth opening	yes	surgical removal
11	Gupta, et al.[19]	2017	19	female	Left posterior mandible	1.4 cm × 0.9 cm	swelling and pain	yes	N/A
12	Bhattacharya, et al.[4]	2015	N/A	female	Right posterior mandible	4 cm × 3 cm	swelling	yes	surgical removal
13	Lone, et al.[20]	2014	15	male	Left posterior mandible	3.5 cm	swelling and pain	yes	enucleation
14	Kudva, et al.[21]	2016	23	female	Right posterior mandible	3.0 cm × 3.5 cm	swelling and pain	yes	excision
15	Presenting case	2024	25	female	Left posterior mandible	6 cm × 3 cm	swelling	no	enucleation

Genetic factors play a crucial role in the development of odontomas, working through various mechanisms. First, the inheritance of abnormal genes can contribute to odontoma formation. According to Hitchin, odontomas may be inherited postnatally through a mutant gene that alters the genetic control of tooth development. The dental lamina, which gives rise to dental germs, typically disappears after fulfilling its function. However, remnants of this structure, known as rests of Serres, may have a significant role in the etiology of both compound and complex odontomas, which can appear where a tooth should be. A mutation

in the epithelial cells that form the dental lamina or germ rests can impair the capacity of the odontogenic epithelium to progress through the necessary stages of tooth formation, while still maintaining the ability to stimulate mesenchymal differentiation required for dentin formation and the development of functional ameloblasts and odontoblasts, leading to the formation of an odontoma. Second, mutations in specific genes, such as LHX8 and PARP1, have also been implicated in the morphogenesis of compound and complex odontomas. Lastly, the abnormal expression of genes that control normal tooth formation and structure can interfere with the mechanisms governing tooth development, contributing to the formation of odontogenic tumors.[4,7]

Odontomas, despite often being asymptomatic when small, can expand and lead to facial asymmetry as the jaw enlarges.[11] The patient exhibited swelling and buccal cortical plate expansion, resulting in facial asymmetry. Clinical features of odontomas may include the retention of primary teeth, the non-eruption of permanent teeth, pain, cortical bone expansion, and tooth displacement.[2] The mass was located in the left posterior mandibular region, where all molars were missing. This association between an odontoma and the absence of one or more adjacent teeth is an uncommon occurrence.[22] Erupted odontomas can be symptomatic, potentially causing complications such as pain, swelling, adjacent soft tissue inflammation, or suppuration from infection of surrounding structures.[2,11] Clinically, erupted odontomas may resemble a solidified calculus deposit on the occlusal surfaces of teeth, as observed in this instance. They may also be mistaken for necrotic bone associated with osteomyelitis when erupting to the level of the alveolar ridge.[4] An ulcer was present on the adjacent gingiva, potentially due to chronic irritation from the mass's rough, hard surface in contact with the mucosa.[11] Secondary infections around erupted odontomas have been attributed to the lack of adhesion between the odontoma and the surrounding soft tissue/bone interface, increasing the risk of microbial invasion.[21]

Radiographically, complex odontomas are characterized by amorphous, radiopaque masses with a surrounding thin radiolucent zone.[6] The radiographic appearance of odontomas varies depending on their developmental stage and degree of mineralization. In the initial stage, complex odontomas appear as radiolucent areas due to incomplete calcification of dental tissues. In the intermediate stage, they show a mixed radiographic appearance from partial calcification. In the final, mature stage-the third stage-the mass typically manifests as a radiopaque amorphous mass with varying densities of hard dental tissues, encapsulated by a radiolucent rim that corresponds histologically to a connective tissue capsule lined by a thin radiopaque sclerotic border.[2,4,11] The radiopaque appearance with amorphous dental hard tissue masses and the surrounding radiolucent zone indicates the mass was completely mature and in the third stage.[2,11] The absence of cortication at the superior aspect of the mass in the radiograph suggests the eruption of the mass into the oral cavity. Complex odontomas must be differentiated from cemento-ossifying fibromas, as odontomas are more radiopaque, while cemento-ossifying fibromas are characterized by a diffuse, less dense radiopaque lesion when mature, and lack a surrounding radiolucent halo.[4,11] Periapical cemento-osseous dysplasia/focal cemento-osseous dysplasia are mature fibro-osseous lesions that may be solitary with dense radiopacities and a radiolucent rim, but PCOD has a wider, uneven sclerotic border. PCOD is situated deep in the alveolar bone, whereas a complex odontoma often extends high into the alveolus towards the crest of the ridge.[4,11] An ameloblastic fibro-odontoma is more radiolucent compared to an odontoma because it has a greater soft tissue component; even when the hard tissue component increases, the complex odontoma has a single mass of disorganized tissue in the center, whereas the ameloblastic fibro-odontoma has multiple scattered mature pieces of dental hard tissues.[4,11] Osteoid osteomas are characterized by a small, ovoid or round radiolucent area surrounded by a rim of sclerotic bone, with some calcification in the central radiolucency.[21]

Odontoameloblastoma is an extremely rare condition involving the simultaneous occurrence of an ameloblastoma and a complex odontoma.[11]

Intraosseous or central odontomas can subsequently become extraosseous or erupt.[18] The impaction of teeth beneath an odontoma occurs due to obstruction of the eruptive pathway. Approximately 37-87% of patients with odontomas exhibit delayed eruption of a permanent tooth, with this probability increasing to 86% in erupted odontomas.[2,15] As suggested by Junquera et al., most erupted odontomas are associated with an impacted tooth, and the eruptive force of this tooth may play a significant role in the eruption of the odontoma.[2,7,21]However, no impacted tooth was identified in this case. Ragalli et al. proposed that the reactive growth of the circumambient capsule of the odontoma may contribute to its eruption.[2,21] It is inaccurate to describe the process as "odontoma eruption," as it differs from tooth eruption, since the lesion lacks a periodontal ligament and root. Without the contractility of fibroblasts, an odontoma cannot erupt.[7,11] The absence of a periodontal ligament in an odontoma precludes its eruption mechanism from being similar to that of normal teeth.[4,11]

Several eruptive mechanisms for odontomas have been proposed in the literature, including pressure resorption, bone remodeling, and trauma.[4,11] The enlargement of the odontoma and the consequent pressure it exerts may lead to sequestration of the overlying bone, resulting in occlusal displacement or eruption of the odontoma.[4,11] Furthermore, odontoma eruption within the oral cavity can be attributed to bone remodeling in an edentulous area, where the bone height decreases until the odontoma is exposed.[7] Additionally, the increasing size of the odontoma and the associated gingivitis may promote resorption of the overlying bone.[5]

The eruption of odontomas can also be attributed to remodeling of the jawbone. However, this process requires the presence of a dental follicle, which indirectly provides the necessary conductance and chemoattraction for the osteoclasts involved in the eruption.[4,11,18] Immunocytochemical studies have indicated that the pattern of cellular activity involving both the reduced dental epithelium and the follicles is associated with tooth eruption. The reduced dental epithelium initiates a cascade of intercellular signals by expressing epidermal growth factor-β and transforming growth factor. These factors then stimulate the follicular cells to produce colony-stimulating factor (CSF), which recruits osteoclasts to the follicle. Additionally, the reduced dental epithelium secretes proteases that assist in the breakdown of the follicle, creating a path of least resistance. This epithelial signaling could explain the remarkable consistency of eruption times, as it is likely that the dental epithelium is programmed as part of its functional life cycle.[18]

In cases where erupted odontomas are not associated with impacted teeth or are located in edentulous ridges, it has been hypothesized that resorption of the edentulous portion of the alveolar process may play a role. Additionally, the reactive growth of the capsule surrounding the odontoma could also contribute to its eruption.[18] Eruption at a younger age may be facilitated by bone remodeling that results from the presence of dental follicles.[18] The pressure resorption theory has been proposed to be more prevalent in older individuals, whereas bone remodeling appears to be a significant factor in younger patients.[4,11,23] In this case report, the lack of impacted teeth and the remodeling and resorption of the surrounding bone appear to be key factors contributing to the eruption of the odontoma. The etiology of odontoma eruptions is multifactorial in nature, and its underlying mechanisms have not been fully elucidated.[2,21]

Regardless of their eruptive characteristics, odontomas can become infected due to the replacement of large areas of normal bone with avascular tumor tissue.[4] Rarely, there may be additional manifestations, including the formation of dentigerous cysts and calcifying epithelial odontogenic cysts.[23] These

complications emphasize the importance of early diagnosis and surgical removal of this lesion.[4]

Odontomas generally have a low recurrence rate.[5] Removing the lesion and surrounding soft tissue through curettage is the preferred treatment to prevent cystic degeneration.[24] Across the reported cases, the treatment approach consistently involved surgical excision or enucleation of the lesion. The specific surgical technique selected is typically based on factors such as the tumor's size, its anatomical location within the maxilla or mandible, and the thickness of the surrounding buccal or lingual cortical bone. For complex odontomas located in the mandible, numerous studies have described successful management through a simple enucleation procedure.[25] However, the treatment of giant odontomas can present higher complication rates due to their size and proximity to adjacent structures.[1,6] In cases of more extensive lesions, the removal process may involve significant bone loss, increased risk of jaw fracture, and a risk of damage to the inferior alveolar nerve.[6] Pathological fracture of the mandible is a concern when managing expanded lesions with a very thin cortical plate remaining.[26] Sectional removal of the odontoma can help prevent jaw fracture.[21] The odontoma was successfully removed via enucleation without complications. Subsequently, the patient was provided with a prosthetic device fabricated in the prosthodontics department, aimed at restoring normal physiological function.

The limitations of this case report include the absence of histopathological analysis and three-dimensional imaging. While the existing data provide a thorough examination of the patient's clinical presentation and diagnostic findings, additional investigations could enhance the understanding of the disease's characteristics and outcomes.

CONCLUSION

Odontomas rarely erupt into the oral cavity. Despite their benign nature, their eruption and large size can lead to complications such as pain, inflammation, infection, ulceration, and facial asymmetry. Dentists should be alert to delayed eruption or missing teeth, and utilize two-dimensional radiography for early diagnosis. This case highlights the crucial role of comprehensive radiographic evaluation. While odontomas are commonly asymptomatic, their capacity for substantial growth, as demonstrated here, underscores the need for early detection. This case has important implications, serving as a reminder for clinicians to maintain a high index of suspicion for odontomas, even when overt symptoms are absent, and to employ appropriate imaging techniques to enable prompt diagnosis and intervention, thereby reducing the risks associated with delayed treatment. Partial resection of large odontoma is recommended to prevent surgical complications.

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