CASE REPORT

An Erupted Silent Tumour

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Abstract Odontomas are the most common odontogenic tumors of the oral cavity which are nonaggressive, hamartomatous in nature consisting of enamel, dentin and cementum. They are called as composite because they contain more than one type of tissue. They are generally asymptomatic, hence recognised on routine radiologic examination. The compound odontoma is composed of multiple small tooth like structures, whereas the complex odontoma consists of a conglomerate mass of enamel and dentine, which bears no anatomical similarity to the tooth. The eruption and infection of odontoma are uncommon, only few cases of erupted complex odontoma are reported in the literature. We report a case of silent erupting complex odontoma.

Introduction

Odontomas are the most common odontogenic tumors of the oral cavity which are nonaggressive, hamartomatous in nature, consisting of enamel, dentin and cementum. They are called as composite because they contain more than one type of tissue. They are generally asymptomatic, hence identified on routine radiological examination. The compound odontoma is composed of multiple small tooth like structures, the complex odontoma consist of a

conglomerate mass of enamel and dentin, which bears no anatomical resemblance to the tooth. The eruption and infection of odontoma are uncommon, but only a few cases of an erupted complex odontoma are reported in the literature.

We report a case of a large erupted complex odontoma associated with an impacted molar (Figs. 1, 2, 3).

A 23 year old healthy female visited the department of oral and maxillofacial surgery, Manipal with the complaint of pain and swelling in the lower right back region of the jaw since 6 months. She had consulted a local dentist for pain and swelling in the same region 2 weeks ago where she was prescribed some medicines and was diagnosed with pericoronitis for which operculectomy was done. It revealed a yellowish white tooth like material and she was referred to our hospital for further management. She had associated numbness along the distribution of the inferior alveolar nerve on the right side since 2 weeks. On clinical examination mouth opening was adequate, swelling of the buccal and lingual gingiva was obvious in relation to the missing molar region, and exposed yellow colored tooth irregular in structure with inflamed gingival margin was observed. Palpation of the third molar region revealed tenderness. There were no teeth with decay. Orthopantomograph showed a dense rounded radio opacity (about 3.0×3.5 cm), distal to the mandibular right second premolar and overlying the coronal portion of the impacted mandibular molar, which was displaced mesio-inferiorly. A uniform, well-defined radiolucent halo surrounded the radio opacity except in the superior area where it erupted into the oral cavity. The right mandibular canal was displaced inferiorly with the impacted first molar on it. There was clear evidence of bone between the second premolar and the lesion. Considering the clinical and radiologic features, a diagnosis of erupting complex odontoma was

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Fig. 1 Clinical photograph

considered. CT scan was advised for the surgical planning. It showed clear relationship of the inferior alveolar nerve with the lesion. Under general anesthesia excision of the odontoma through intraoral approach was accomplished along with the fibrous capsule and the extraction of impacted first molar. Surgical site was packed with antiseptic gauze pack (BIPP), left to heal by secondary intention. The excised material was sent for histopathologic examination (Figs. 4, 5, 6).

Study of the specimen exhibited haphazard deposition of enamel, dentin and cementum. The capsule showed premature mesenchyme with odontogenic rests arranged in the form of rosettes and strands. The final diagnosis was erupted complex odontoma (Fig. 7).

Patient reported every week after surgery. The size of the defect reduced to the size of the extraction socket (Fig. 8).

Discussion

The term odontoma was first coined by Broca in 1866, who defined it as a tumor formed by overgrowth of complete dental tissue [1]. Odontoma constitute about 22 % of all

Fig. 3 CT Scan image

odontogenic tumors of the jaws [2]. In studies using the 2005 WHO classification, the most frequent odontogenic tumors follow the sequence, KCOT, ameloblastomas, and odontomas. They are asymptomatic hamartomatous, nonaggressive benign tumors commonly diagnosed by routine radiological examination. Studies that employed the 1992 classification usually reported odontomas as the highest prevalent odontogenic tumor in USA, Italy, Chile and Estonia and second highest in Iran, Turkey and third highest in India. In studies using the 2005 WHO classification, odontoma is second highest in Mexico and third highest in China, Libya, Brazil and Egypt [3].

The World Health Organization classifies odontomas as a benign odontogenic tumor composed of odontogenic epithelium and odontogenic ectomesenchyme with dental hard tissue formation. In 1992 based on the degree of morphodifferentiation WHO classified odontomas as

Fig. 2 Radiographic image

Fig. 4 Surgical procedure showing the removal of the odontoma

Fig. 5 Surgical specimen

Fig. 6 Impacted molar

Fig. 7 Histopathologic image depicting the haphazard collection of enamel, dentin, pulp

compound odontoma with at least superficial anatomic resemblance to teeth as all the dental tissues are represented in an orderly fashion. Compound odontomas are of three types; *denticular* type composed of two or more separate denticles, each having a crown and root; *particulate* type composed of two or more separate masses of particles bearing no macroscopic resemblance to tooth and consisting of hard dental tissues abnormally arranged and *denticuloparticulate* type where denticles and conglomerate masses or particles are present side by side [4], while in a complex odontoma there is little or no morphologic resemblance to normal tooth formation.

They have also been classified clinically as:

- Intra-osseous (central)
- Extra osseous (peripheral)
- Rarely, intra-osseous odontomas may facilitate their eruption into the oral cavity when located coronally to an impacted or erupting tooth or superficially in bone. Here they are referred to as erupted odontomas. Of the two forms of odontoma complex odontoma erupting into the oral cavity is very rare [5].

Rumel et al. in 1980 described the first case of erupted odontoma [2]. To our knowledge there are only 40 cases of erupted odontomas reported including our case. Of the 40 cases 20 cases corresponded to compound odontomas and 20 to complex odontomas.

Hitchin suggested that odontomas are inherited through a mutant gene or interference, possibly postnatal, with genetic control of tooth development. The odontogenic tumour epithelial cells are recapitulating genetic programs expressed during normal odontogenesis, but the tumour cells demonstrate abnormal expression of these genes [6].

Fig. 8 Radiograph after 3 months showing the bone formation

Odontomas can also manifest as part of some syndromes: Gardner syndrome, Hermann syndrome, familial colonic adenomatosis, and basal cell nevus syndrome [7].

Radiographically odontomas have characteristic features which depend on their stage of development and degree of mineralization.

- First stage: A radiolucency due to lack of calcification
- Intermediate stage: Partial calcification is observed
- Third stage: The lesion usually appears as radiopaque masses surrounded by radiolucent areas corresponding to the connective tissue histologically [5]

They differ from cemento ossifying fibromas, as they are more radiopaque and have a tendency to associate with unerupted molar teeth. Odontomas occur more among younger patients as compared to cemento-ossifying fibromas. Although periapical cemental dysplasia might resemble the complex odontoma, they are usually multiple and centered on the periapical region of the teeth. In addition, the periphery of cemental dysplasia usually has a wider uneven sclerotic border.

Differentiating an ameloblastic fibro-odontoma (AFO) from a developing odontoma might be difficult, but generally these tumors have a greater soft tissue component (radiolucency) than the odontoma. A complex odontoma usually has a mass of disorganized tissue in its center, whereas, the AFO usually has multiple, scattered, mature, and small pieces of dental hard tissue. Also, the AFO occurs in older individuals as compared to the complex odontoma. Regions of enostosis, although radiopaque, do not have a soft tissue capsule, as seen in odontomas [8].

A cementoblastoma presents as a well-defined radiopaque mass attached to the tooth root and surrounded by a radiolucent rim.

Osteoid osteomas are characterized by a small ovoid or round radiolucent area surrounded by a rim of sclerotic bone; the central radiolucency exhibits some calcification [6].

Odontoameloblastoma is another very rare sort of mixed odontogenic tumor commonly affecting children and adolescents, whose aggressive growth is usually accompanied by bone expansion and root resorption [9].

Various theories have been proposed for the eruption of odontoma. The mechanism of odontoma eruption appears to be different from tooth eruption because of the lack of periodontal ligament and root in odontoma. Therefore the force required to move the odontoma is not linked to the contractility of the fibroblasts, as in the case for teeth. An increase in the size of the odontoma over time produces a force sufficient to cause bone resorption. Another reason may be bone remodeling.

Immunohistochemical investigations have suggested that the cellular activity involving the reduced enamel epithelium and the follicle is associated with eruption. However, for this to occur dental follicle is required although indirectly, as it provides both the conductance and chemoattraction for the osteoclasts necessary for tooth eruption [8].

Junquera et al. [10] suggested that most of the erupted odontomas are related to unerupted teeth, so he postulated that the eruptive force of these teeth plays an important role in odontoma eruption.

Ragalli et al. [11] suggested that reactive growth of capsule may contribute to eruption of odontoma.

Secondary infections around an erupted odontoma have been attributed to the lack of adhesion between the odontoma and the surrounding soft tissue/bone interface resulting in increased chances of microbial invasion. Bertolai et al. reported a case of Ludwig's angina associated with an erupted odontoma in a 29-year-old female, which was managed by antibiotic therapy, intraoral incisions to drain the swelling and surgical excision of the odontoma [12].

A rare case of an odontoma occurring in the cranium near the pituitary gland has been reported by Faria et al.

[13] demonstrating that an odontogenic lesion may arise in brain tissues due to the embryological relationship between primordial stomodeum and Rathke's pouch, which induced hypophysis disruption, and therefore, hypothyroidism, diabetes, and growth retardation.

Treatment

Reports of odontoma removed in sections prevent the fracture of the jaw [8]. Goran et al. performed the operation by two-stage procedure intraorally [4]. Blinder et al. described an intraoral buccal and lingual approach and discussed the advantages for the removal of large odontomas [14]. Savitha and Cariappa recommended a bony lid technique for enucleation of a large ameloblastic fibro-odontoma, encroaching on the left ramus of the mandible by using an extraoral approach [15]. Casap et al. in 2006 have described a case of a large odontoma in mandibular ramus region which was removed using sagittal split technique [16]

The present case was also treated by removal of the odontoma and the impacted tooth and left to heal by secondary intention without any grafts because of the following reasons:

1. To prevent any compression over the inferior alveolar nerve.
2. The defect was unable to close by primary closure.

Conclusion

Odontoma is a nonaggressive lesion usually identified by routine radiological examination. The clinical features may vary from no symptoms to nerve paresthesia. The management of which is usually conservative. Early diagnosis and prompt treatment could prevent the possible complications.

Compliance with Ethical Standards

Conflict of interest None.

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