# ODONTOMA: AN INSIGHT INTO THE MOST COMMON BENIGN ODONTOGENIC TUMOR

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Abstract: Odontomas are slow-growing hamartomas of aborted tooth formation which account for 22% of the odontogenic tumors. They are the most common benign odontogenic tumors of epithelial and mesenchymal origin, consisting of enamel, dentine, cementum and pulpal tissue. The diagnosis is based on clinical examination and radiographic images, and following surgical removal, it must be further confirmed by histological examination. This article presents an insight into odontomas of the oral cavity.

Keywords: odontoma, odontogenic tumor, benign

#### INTRODUCTION

The term 'odontoma', coined by Paul Broca in 1867<sup>1</sup>, was originally used for any tumor and/or tumor-like lesion of odontogenic origin. Over years, as a consequence of new insights, the term odontoma has changed its meaning, and since 2017 it has been classified according to the World Health Organization (WHO) in a group of benign mixed epithelial and mesenchymal odontogenic tumors. However, it is defined as tumor-like malformations (hamartomas) consisting of hard and soft dental tissues, enamel and dentine with different proportions of pulp and cement, not representing a true neoplasm.<sup>2</sup> These have been also called "composite odontomas" because they are composed of more than one type of tissues.<sup>3</sup> They are the most common benign odontogenic tumors<sup>4</sup> and are considered to be hamartomas rather than true neoplasms.<sup>5</sup> Buchner et al. in 2006 examined a sample of 1088 odontogenic tumors, in which odontomas accounted for 75.9% of the total. Previously, in 2002, Ochsenius et al. analyzed a sample of 362 odontogenic tumors in which odontomas represented 44.7%. In the year 1997, Mosqueda et al. evaluated a series of 349 odontogenic tumors, of which 34.6% corresponded to odontomas.<sup>6</sup> Odontomas arise from the growth of fully differentiated epithelial and mesenchymal cells that give rise to ameloblasts and odontoblasts and subsequently form the enamel and dentin tissues, respectively. Enamel and dentin are often deposited in an abnormal pattern due to the failure of the organization of odontogenic cells to achieve a normal state of morphological differentiation. Therefore, odontomas are considered as dental developmental anomalies rather than true odontogenic tumors.<sup>7</sup>

In 1914, Gabell et al. grouped odontomes according to their developmental origin into: epithelial, composite (epithelial and mesodermal) and connective tissue. According to 2005 WHO classification of odontogenic tumours, there are two types of odontomas, compound and complex odontomas. Clinically, they are classified as intraosseous (central), peripheral (soft tissue or extraosseous), and erupted odontomas. Compound odontomas appear as numerous miniature or rudimentary teeth. Complex odontomas appear as amorphous conglomerates of hard tissue. Mixed odontomas are a form of odontoma that combines the characteristics of a compound and complex odontomas. Most are found when a patient is evaluated radiographically for tooth eruption disturbances.

Differential diagnosis includes calcifying odontogenic cyst, calcifying odontogenic tumour, fibro-osseous lesion, ameloblastic fibro-odontoma and osteoblastoma. Some investigators have suggested that ameloblastic fibroma and ameloblastic fibro-odontoma both developmentally and histo-morphologically represent the early stages of formation of odontomes. Ossifying fibroma may closely mimic an odontoma but can be differentiated from it by the fact that it is well circumscribed and usually separates easily from its bony bed.

### **ETIOPATHOGENESIS**

Exact etiology od odontoma is known. Sources of cells for odontomas could be mature ameloblasts, cell rests of Serres, or extraneous odontogenic epithelial cells. These cells can be stimulated by either environmental or genetic factors. Genetic factors could be either due to inheritance of abnormal genes or mutation of the responsible genes and interference in the mechanism of genes controlling tooth formation.<sup>11</sup>

Papagerakis et al. stated that the differentiation of normal and tumor odontogenic cells is accompanied by the expression of some common molecules. The gene products present in some mesenchymal cells were also seen in the odontogenic tumor epithelium. The data may be related to a tumor-specific overexpression of the corresponding genes transcribed

at an undetectable level during normal development and / or to an epithelial-mesenchymal transition proposed to occur during normal root formation. A plausible explanation for the result is that odontogenic tumor epithelial cells are recapitulating genetic programs expressed during normal odontogenesis, but the tumor cells demonstrate abnormal expression patterns for these genes.<sup>12</sup>

Odontomas have been associated with trauma during primary dentition as well as with inflammatory and infectious processes, hereditary anomalies (Gardner syndrome, Hermann's syndrome), odontoblastic hyperactivity and alterations in the genetic components responsible for controlling dental development.<sup>13</sup>

Studies by Glasstone (1952) and Rushton (1957) have supported the role of trauma in the development of odontoma. Levy states that the stage of development during which the trauma has occurred determines the development of hypoplastic teeth, odontomas, and supernumerary tooth. Presence of odontoma in sites other than tooth bearing regions suggests that trauma could have possibly displaced the developing tooth germ leading to its malformation.<sup>11</sup>

#### **CLINICAL FEATURES**

Notably, odontomas are usually slow-growing and painless intra-bony lesions. <sup>10</sup> Most of odontomas do not cause clinical symptoms, and many are detected incidentally or after investigating the reason for delayed eruption of a permanent tooth. <sup>3</sup> In general, the clinical indicators of odontoma may include eruption disturbance (non-eruption of permanent teeth, retention of deciduous teeth), expansion of the cortical bone, teeth malposition and pain. <sup>14</sup> In 70% of odontomas, pathological anomalies are observed in relation to the neighbouring teeth such as devitalisation, malformation, aplasia, malposition and affected teeth. <sup>15</sup>

This tumour is usually seen in the second and third decades of life, and rarely seen below 10 years of age.<sup>3</sup> The odontoma generally affects the permanent teeth more than the primary teeth.<sup>16</sup> Katz has reported that odontomas rarely involved the primary dentition and found only five of 396 odontomas (2%), with the youngest being a 2-year-old patient.<sup>17</sup> They are seen more frequently in the maxilla than the mandible.<sup>9</sup> There is a slight predilection for occurrence in males (59%) compared to females (41%).<sup>5</sup>

The majority of odontomas are found to be between 1 and 2 cm in diameter, with those greater than 3 cm being referred to as giant odontomas.  $^{10}$  The size of these large odontomas ranged from 29 to 60 mm with a mean diameter of  $41.08 \pm 9.94$  mm. The largest giant complex odontomas were reported in the literature by Widayanti et al. and Bueno et al.  $^{3}$ 

The compound type is approximately twice as common as the complex type. The compound type tends to occur in the anterior part of the jaws, and the complex type tends to present in the posterior part. The compound composite odontome most frequently occurred in incisor cuspid region of the upper jaw in contrast to the complex odontomes which were commonly found in molar and premolar region of the mandible. Kaugars reported that the percentage of odontomas in the molar region gradually increased with each successive decade of life. Interestingly both type of odontomas occurred more frequently on the right side of the jaw than on the left, (compound 62%, complex 68%). Some reports have reported presence of both the types of odontomes in different locations, such as maxillary sinus, mandibular ramus, subcondylar region or mental foramen, mid palate and the middle ear.

Interestingly, compound odontomas are diagnosed earlier than the complex type, probably due to involvement of upper anterior permanent teeth in most cases. <sup>18</sup> Kemmerer et al. (2015) concluded that 6 % of the odontomas were found in tomographic exams, 16.8% were a coincidental finding, 6.6% due to late eruption of permanent teeth, 1.8% were found due to signs and symptoms of pain and tumefaction. Levi-Duque et al. (2019) concluded that 63.3% of the cases were found in routine radiographic exams, 30% due to the lack of permanent tooth eruption, 5% due to infectious symptomatology and only 1.7% as a consequence of increase in volume. <sup>16</sup>

#### HISTOPATHOLOGIC FEATURES

Histologically the odontoma is not a diagnostic dilemma. It is composed of dentin, cementum, pulpal tissue and enamel. However, mature enamel is lost during the decalcification processing and will not be seen on conventional hematoxylin and eosin stained slides. <sup>13</sup> In compound odontoma, enamel, dentin, and pulp are all normally organized and enclosed in follicular connective tissue like a normal tooth. <sup>10</sup>

Complex odontomas are often spherical in shape and consist primarily of a disordered mixture of odontogenic tissues. Cementum or cementum-like substances are often admixed with dentinoid structures. Small spaces with pulp tissue, enamel matrix, and epithelial remnants may be observed within the calcified mineralised masses of dentin. A thin fibrous capsule or, occasionally, a cyst wall is seen surrounding the lesion. Small islands of eosinophilic-staining epithelial ghost cells are present in about 20% of com plex odontomas. These may represent remnants of odontogenic epithelium

that have undergone keratinization and cell death from the local anoxia. A thin layer of cementum is often present about the periphery of the mass.<sup>20</sup> Loose, myxoid connective tissue with odontogenic epithelial rests may be seen in close association with the lesion, and most often represents normal dental follicular tissue. Fibrous connective tissue with a cystic lining representing a dentigerous cyst may also be seen.<sup>13</sup>

Chang JY, et al did a clinicopathologic study of 81 cases of odontoma and the histologic examination revealed enamel matrix in 90%, dentin in 100%, cementum in 88%, pulp tissue in 96%, fibrous capsule in 93%, ghost cells in 83%, reduced enamel epithelium in 86%, and nests of odontogenic epithelium in 58% of odontomas. Dentigerous cyst was associated with 9% of odontomas.<sup>21</sup>

#### RADIOGRAPHIC FEATURES

Radiographically, odontomas present as well-defined, radio-opacities situated in the bone with density greater than bone and equal to that of a tooth. A radiolucent halo, typically surrounded by a thin sclerotic line, surrounds radio-opacity. The radiolucent zone represents a connective tissue capsule similar to that of the normal tooth follicle. In compound odontoma, multiple teeth-like structures of varying size and shape are seen. Complex odontomas are seen as irregular radio dense masses with no resemblance to teeth. <sup>15</sup> The radiographic findings of odontomas are determined by their stage of development and degree of mineralization. The first stage is characterized by radiolucency due to the lack of calcification. Partial calcification appears in the intermediate stage, while in the third stage, the lesion usually presents as radiopaque masses surrounded by radiolucent areas corresponding to the connective tissue histologically. <sup>22</sup>

#### **MANAGEMENT**

In earlier days; odontoma as tumor of odontogenic origin, was treated with radical resection of the affected area. However, changed concept of odontomas as hamartomatous malformations has modified treatment plan to conservative modality including selective removal of denticles with more emphasis on enucleation of connective tissue capsules, as its remnants left behind can predispose to cystic change, interfere with eruption of permanent teeth and cause considerable destruction of bone due to recurrence.<sup>23</sup>

To avoid tooth eruption abnormalities, early detection and surgical removal of these hamartomas are recommended. <sup>10</sup> Kaban states that odontomas are easily enucleated, and displaced adjacent teeth by the lesion are seldom traumatized during surgical excision because they are usually separated by a septum of bone. <sup>8</sup>

Odontomas may continue to enlarge or cause problems to the adjacent teeth; therefore, surgical removal is the treatment of choice for large odontomas. Depending on the size of the odontoma, various surgical techniques such as removal of cortical plate, segmental osteotomy via extraoral excision, and sagittal split osteotomy may be necessary. The surgical defect after excisional removal may be reconstructed by grafting and plating.<sup>24</sup>

During surgery, it is necessary to be careful not to damage the impacted tooth, adjacent teeth, and anatomic structures of the jaw. Therefore, it is important to accurately localize the odontoma preoperatively by radiography and to determine its relationship with the adjacent structures.<sup>2</sup> Morning examined 42 cases of impacted teeth in relation to odontomas and reported that 17 of 35 impacted teeth erupted after removal of odontomas with or without exposure of the impacted teeth.<sup>17</sup>

#### **SUMMARY**

Odontomas are benign tumor-like lesions which originate from epithelial and ectomesenchymal components. They are one of the most frequent odontogenic lesions in the series of odontogenic tumors. Typically asymptomatic, they are revealed on routine radiographs or upon assessing the origin of delayed tooth eruption. Early diagnosis and correct treatment are essential to avoid any complications, such as prolonged retention of primary teeth and delayed eruption of permanent teeth.

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