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Case Report

A New Lesion? A True Cystic Odontoma, Completely Enclosed within Its Own "Dental Follicle"

Jerry E Bouquot[1]*, Melissa Luna[2], Brett Barnette[3] and Yassmin AM Mohamed[4]

[1]*Clinical Professor and Past Chair of Oral Pathology, Department of Oral and Maxillofacial Surgery, School of Dentistry, West Virginia University, Morgantown, West Virginia*

[2]*Assistant Professor, Director of Maxillofacial Diagnostics, Department of Oral and Maxillofacial Surgery, School of Dentistry, West Virginia University, Morgantown, West Virginia*

[3]*Resident, Department of Oral and Maxillofacial Surgery, School of Dentistry, West Virginia University, Morgantown, West Virginia*

[4]*Lecturer, School of Dentistry, University of Khartoum, Khartoum, Sudan*

***Corresponding Author:** Jerry E Bouquot, Clinical Professor and Past Chair of Oral Pathology, Department of Oral and Maxillofacial Surgery, School of Dentistry, West Virginia University, Morgantown, West Virginia.

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Abstract

Background: The odontoma is the most common of all odontogenic tumors but one of its variants, the cystic odontoma, is so infrequent that it is typically not even mentioned in published reports and reviews of odontomas. We present what we believe to be the first "true" cystic odontoma, defined as a compound or complex odontoma surrounded entirely by its own epithelium-lined cystic sac, essentially found within a fluid-filled cystic lumen, with no associated impacted tooth and no attachment of the cyst lining to the inner structures. We suggest that the term cystic odontoma be reserved only for this type of odontoma, and that the descriptive term "dentigerous cyst with embedded odontoma" be used for those entities comprised of odontomas in the stroma of a cyst. We present herein a detailed account of the first true cystic odontoma, with a brief review of a second one from our files.

Case Reports: A 19-year-old male presenting with an asymptomatic, slowly enlarging cortical expansion of the facial aspect of the right maxillary incisor region. No unerupted teeth were in the area and the lesion had slightly rotated both right incisors. Radiographs showed a 19x14 mm well-demarcated mixed radiopaque/radiolucent lesion with an even, thin sclerotic rim surrounding an irregular zone of radiolucency. Diminutive tooth-like structures could be seen. At surgery a lobulated mass was seen to be completely covered by a thin, semitranslucent membrane. The lesion shelled out easily and the surface membrane peeled away completely, with no attachment to underlying structures. Microscopically, the outer capsule was similar to a dental follicle, with a cuboidal epithelial layer overlying dense fibrous tissue and facing a centrally located mass of small, often mature teeth with areas of immature or embryonic dental tissues in a fibrous background. There was no recurrence. An additional cystic odontoma from our files was located in the anterior maxillary midline of a 16-year-old female. It was asymptomatic and non-expansile and 11 x 11 mm in radiographic diameter; it proved to be a composite odontoma "floating" in a clear fluid, surrounded by an epithelial-lined cyst.

Conclusions: We present the first reported example of a true cystic odontoma: an odontoma completely surrounded by a follicle-like cyst lining. The calcified portion of the odontoma, in this case a combination of compound and complex odontomas, was located within a fluid-filled cystic lumen; there was no associated impacted tooth. The odontoma shelled out easily and did not recur.

Keywords: Cystic Odontoma; Odontoma; Odontogenic Tumors

Introduction

Odontogenic tumors are among the most varied of human tumors and the odontoma is the most common of all such tumors, representing up to 75% in some oral pathology biopsy services [1-4]. First reported as a "stony growth" by Fouchard in 1728, the odontoma is now believed to be more a developmental hamartoma than a genuine neoplasm of odontogenic origin, but some can become very large (Figures 1A, 1B) and the true etiology remains elusive [5-9]. Although occasional familial cases or associations with Gardner syndrome, basal cell nevus syndrome, familial colonic adenomatosis, Tangier disease, Hermann syndrome and osseous dysplasia of the jaws have been reported, the vast majority of cases are associated with no identifiable cause [3,10-12]. They can develop at any age, but usually are diagnosed in the teen and young adult years [3].

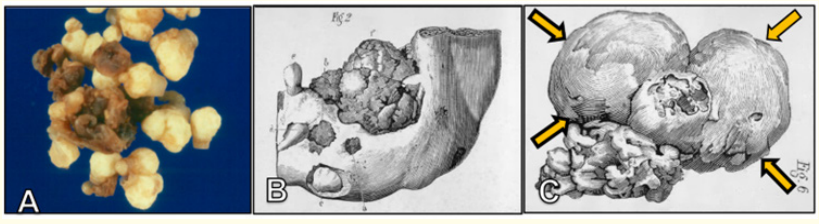

Figure 1: Examples of compound and complex odontomas. A) Gross appearance of compound odontoma, with many tooth-like structures present; B) 1860 (Forget) drawing of a massive complex odontoma, with no actual tooth-like structures, which has perforated the cortex in several places; C) Possible first example (Forget, 1860) of a likely cystic odontoma partially covered by a soft tissue capsule (arrows) [6].

Two types of odontoma are currently identified by the WHO: 1) The compound odontoma (Figure 2A) and the complex odontoma, with the first containing primitive or mature but diminutive tooth-like structures or "denticles" and the latter containing irregularly formed, immature and mature dentin, cementum, enamel and pulp tissues with minimal actual tooth formation; additional subcategories include central and peripheral, depending on whether the tumor is inside or outside the bone [3,13-15].A third microscopic diagnosis is sometimes used: composite odontoma, with microscopic features of each of the first two [3,14].

Regardless of the histologic type, many odontomas are found near the crown of an impacted tooth (Figure 2A), typically in the stroma of a dental follicle or dentigerous cyst, occasionally perforating through the epithelial lining to protrude into the fluid of the cyst lumen, but originating outside of that cyst lumen [16-25]. So far, authors have referred to these as "cystic odontomas," but in reality they are a combination of two different lesions occurring in a common site: an odontoma in the wall of a dentigerous or other cyst. We present the first two cases of a "true" cystic odontoma, i.e. an odontoma *not* associated with a tooth and located *entirely within* a fluid-filled cyst lumen, not in the fibrous wall of a cyst lining (Figure 2B).

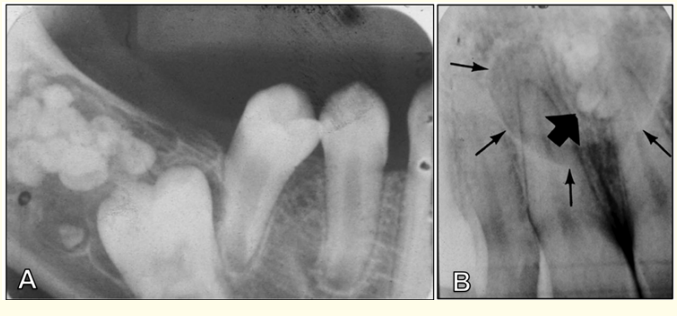

Figure 2: Examples of faux and real cystic odontomas. A) A compound odontoma in the wall of a dentigerous cyst; B) A true cystic odontoma proven microscopically (not shown) to represent a composite odontoma completely located inside an odontogenic cyst, with small arrows outlining a well-demarcated radiolucency which was fluid-filled; large arrow pointing to an aggregate of small tooth-like structures inside the cyst.

While one of our cases appears herein to be the first reported with complete clinicopathologic information, there may have been a true cystic odontoma reported, without histology but with a clinical drawing, as early as 1860 (Figure 1C).[6].Our examples, one in detail, were derived from the authors' clinical practices, and one has been reported at a national meeting [26].

Case Report

A 19-year-old male presented with the complaint of a "swelling" of the anterior right maxillary area which was bulging his upper lip causing an esthetic concern (Figure 3). This asymptomatic mass had been slowly enlarging for at least seven years. The patient could recall trauma to the region at age 11, causing small incisal edge fractures of both central incisors, but was unaware of a local infection or of another family member with a similar problem. His medical history was unremarkable.

At examination, an asymmetry was noted in the right upper lip region. A bony hard 16x11x5 mm sessile mass of the facial cortex was seen beneath the lip, with deviation of the midline frenum toward the left. The right lateral incisor was slightly rotated, and a diastema was noted between this tooth and the canine; the right central incisor was somewhat over erupted (Figure 3A). Adjacent teeth were all vital to routine pulp testing, and the surface mucosa had a normal appearance. Gingival margin asymmetry was observed when right maxillary incisors were compared with the left ones.

- **Radiology:** Pantographic and periapical radiographs revealed a well-demarcated mixed radiopaque/radiolucent lesion with an even, thin sclerotic rim surrounding an irregular zone of radiolucency (Figure 4). Diminutive tooth-like structures could be seen. The root of the right maxillary lateral was slightly deviated towards the distal.
- **At surgery:** When a full thickness buccal flap was raised for surgical removal, much of the facial alveolar cortex was perforated or missing (Figure 3B). Once exposed, the lobular mass, with its surface nodules, shelled out easily in one piece (Figure 5A), and the outer semitransparent covering could be peeled off completely, with no obvious connection to underlying calcified tissues, and separated from them by a small amount of clear fluid. Several much smaller, independent "dental follicles" were found around 4 of the individual "teeth" centrally in the lesion (Figures 5B,5C), but none connected to the surface capsule.
- To avoid ridge deficiency after lesional removal, a bovine-derived bone graft (PepGen P15, DENTSPLY Tulsa Dental Specialties, Tulsa, OK) was used in the defect area.
- **Histopathology:** Microscopy demonstrated that the outside capsule consisted completely of dense and rather avascular fibrous tissue with a cyst lining of cuboidal epithelium facing the internal calcified portions of the mass (Figure 6A). This epithelium was similar to but not identical to that seen in dental follicles or dentigerous cysts. Within the mass, multiple small tooth-like figures (Figure 6B) and rounded calcified structures were seen in a background fibrous stroma, along with sheets and islands of dentinoid, osteodentin and cementoid tissues, often intermixed with globules of enamel matrix (Figure 7). Four small teeth had classic dental follicles around their crowns and the pulp tissue in them was normal and mature. Occasional small islands of benign odontogenic epithelium were scattered throughout the mature fibrous background stroma; the stroma was not consistent with the more pritive stroma of an ameloblastic fibroma.
- **Follow-Up:** Six months after odontoma removal, and without further dental or surgical treatment, the right maxillary incisors had repositioned, resolving the patient's esthetic concerns. No recurrence was seen after 2 more years of follow-up.

Discussion

As earlier mentioned, odontomas are, by far, the most common odontogenic tumors in oral pathology biopsy services [3,28]. The prevalence rate in the general population, as determined by evaluating 1,250 pantographic radiographs from persons in the second and third decades of life, is 1.4/1,000 [27]. Compound odontomas represent about 2/3 of all odontomas submitted for biopsy and most often develop in the anterior maxilla, while the complex odontoma is seen most frequently in the posterior mandible [3]. Overall,

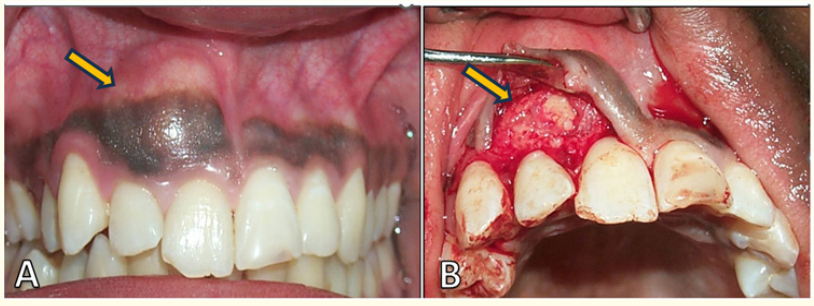

Figure 3: Clinical presentation of lesion. A) Large circular mass was bone hard on palpation (arrow), with the underlying central incisor overerupted; B) After removing overlying soft tissue, a firm mass of individual "calcified globules" was seen, with no overlying cortex.

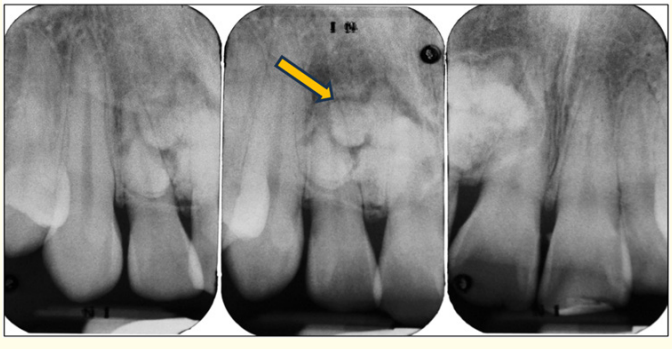

Figure 4: Radiographs of the mass show it to be comprised of numerous calcified globular constructions, sometimes forming into tooth-like structures (arrow).

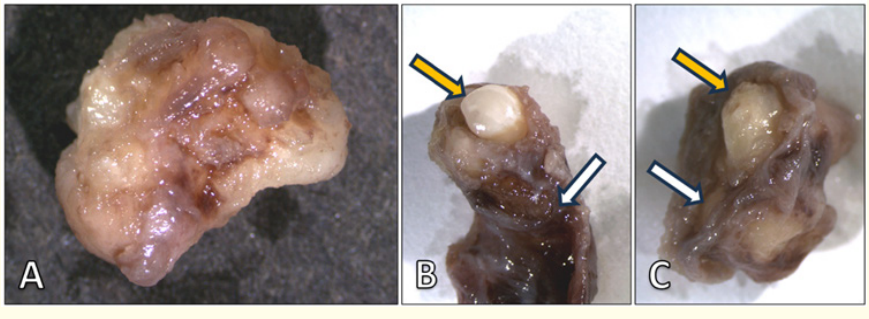

Figure 5: Gross specimen after removal. A) Entire mass with the nodular facial surface noted in Figure 1B, it is covered on all sides by a cyst-like membrane; B) Small cuspid-like structure (yellow arrow) immediately after removal of a smaller or secondary follicle (white arrow); C) Small premolar-like structure (yellow arrow) after removal of its own individual follicle (white arrow).

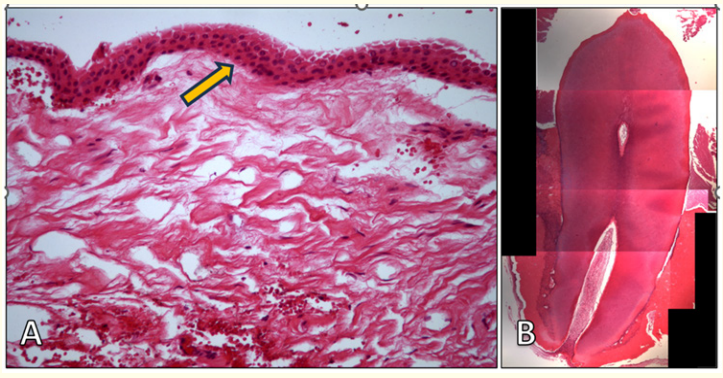

Figure 6: Histopathology of lesion. A) Fibrous capsule completely surrounded the mass, with lining epithelium of cuboidal cells (arrow) on the surface facing the tooth structures; B) Cuspid like structure in Figure 3B showing that root and pulp tissues were all present, well formed and mature.

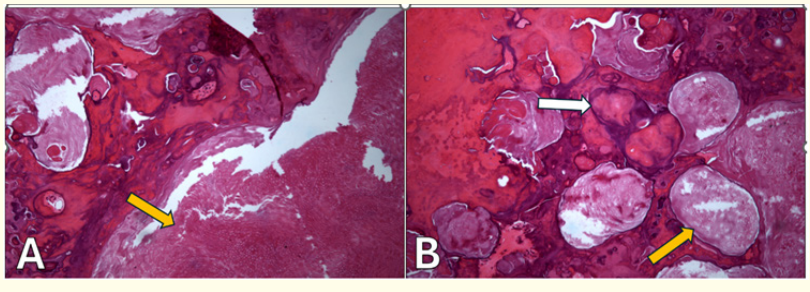

Figure 7: Additional histopathology of lesion. A) Much of the mass was comprised of large and small globules of enamel matrix (arrow); B) Throughout the mass were islands and sheets of dentinoid and cementoid material with embedded rounded dentin globules (white arrow) and rounded globules of enamel matrix (yellow arrow).

there is a slight female predilection and almost 80% of cases are diagnosed during the second and third decades of life, with an average patient age at diagnosis of only 15 years [1-3,8,14,15]. Since odontomas are typically asymptomatic and slow growing, they are usually found during a routine radiographic examination. Occasional cases are diagnosed because of delayed eruption of a tooth under an odontoma, or because of cortical expansion, secondary infection, with resultant pain, or actual eruption through the alveolar mucosa, apparently being pushed by the underlying tooth [1-3,14,15]. Individual lesions are seldom larger than the size of a single tooth in the area from which it arises. Cortical perforation, as seen in the present example, appears to be a rare phenomenon, except in the largest lesions, where it is more common than not.

The biological behavior of each variant of odontoma (including the cystic odontoma, we assume) is similar. They are all minimally aggressive, enlarging very slowly over years, typically reaching a size of 2 cm. or less, then remaining the same indefinitely. Occasionally one will reach a size of 6-7 cm and some prefer to refer to odontomas larger than 3 cm. as "giant odontomas."

The few reported "cystic odontomas" (dentigerous cysts with an odontoma in the wall of the cyst) have all been less than 2 cm. in size, all have been composite types, and all have shelled out very easily at surgery. Also, none of the reported cases, including our own, could be predicted by radiographic appearance, although no report has yet included cone beam CT images. The cystic area of such cases are comprised of fibrous stroma, not cystic fluid.

The teeth in an odontoma are usually quite diminutive but occasional lesions contain normal-sized crowns, usually with small or unformed roots. In our case, several such teeth had true dental follicles surrounding their crowns, not attached to or in any way associated with the larger surrounding cyst wall, but not remarkable from other tooth structures in routine odontomas. Additionally, it should be mentioned that the several islands of benign odontogenic epithelial islands in the odontoma stroma, which is normal for any odontoma, were not found in the stroma of the surrounding "follicle."

There is little or no malignant potential to an odontoma, and we assume that our cystic odontoma has the same biological behavior as other odontomas [3,28,43]. Usually the lesion can be shelled out, even when not encased in a cyst, but the lobular nature of many may force the surgeon to remove it in fragments. While our cases were not associated with erupting teeth, even a small odontoma can prevent the eruption of an underlying tooth and should be removed carefully so as not to interfere with normal eruption. Surgical orthodontics may be required to assist the impacted tooth in its eruption.

The favorable esthetic orthodontic response after surgical removal of the cystic odontoma in the present case suggests that the odontoma was probably "pushing" the teeth into unnatural positions, but this did not produce other injuries to adjacent teeth. The diastema between the maxillary right cuspid and lateral incisor closed within 6 months after the odontoma removal. The authors suggest delaying orthodontics or further treatment to allow natural response as noted in the present case.

One final point should be made. The authors have no idea how an odontoma became centrally located within a dental cyst, but the fact that the surface capsule peeled off easily and completely from the gross specimen suggests that the disease process involved two different lesions, like the old cystic odontomas with their odontomas located in the stroma of an otherwise routine dental cyst or follicle. In our true cystic odontoma, the odontoma happens to develop within the cyst itself, not in its stroma. This perhaps originated from a small primitive odontogenic hamartoma which somehow found its way into a fluid-filled cyst, but we have no real explanation for this development. This true cystic odontoma is in no way similar histologically to cystic adenomatoid tumor, cystic ameloblastoma or calcified cystic odontogenic tumor.

Conclusion

We present what appears to be the first lesion of its kind: a routine composite odontoma with small follicles around several diminutive teeth and a much larger "follicle" surrounding the entire calcified portion of the odontoma, that is, located completely inside a cyst lumen. In every other regard, including biological behavior, it was unremarkable. We also briefly describe a second case of a true cystic odontoma, but without the same detail.

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