

An unusual case of unicystic intramural ameloblastoma and review of the literature

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Abstract

Ameloblastoma is the most common tumor of odontogenic origin. There are various types of this tumor and confusion still exists among the clinicians about the correct classification. Multicystic ameloblastoma is the most frequent subtype while unicystic ameloblastoma can be considered as a variant of the solid or multicystic. This subtype is considered as a less aggressive tumor with a variable recurrence rate. However, its frequency is often underestimated. The aim of this article is reviewing the recent literature about unicystic ameloblastoma using our unusual clinical case as a starting point to illustrate this discussion. A 30-year-old man who had been complaining of slight pain in the premolar and molar area of the left side of mandible had a check up at our department. X-rays revealed a unilocular radiotransparency with radiopaque margins. The first histological diagnosis was an odontogenic cyst. Successive histological evaluations revealed that ameloblastic epithelial islands were present in lasso connective tissue. We think that our case report provides new insights into the approach to the ameloblastoma diagnosis. We agree with authors who have pointed out that a single small biopsy may often be inadequate for the correct diagnosis of ameloblastoma. Moreover, in the light of our experience, it should be kept in mind that ameloblastomas may have sometimes unusual presentations and this fact should induce surgeons and pathologists to consider carefully each lesion.

Keywords: Ameloblastoma, tumor, unicystic

Introduction

Ameloblastoma is the most common tumor of odontogenic origin.^[1] The World Health Organization (WHO) defines it as a locally invasive polymorphic neoplasia that often has a follicular or plexiform pattern in a fibrous stroma.

Clinicians emphasize that ameloblastoma is a benign tumor but is locally aggressive.^[2,3]

In 20% of cases, it is localized in the maxilla where it can be particularly dangerous because of the close proximity to vital structures and the great difficulty to obtain clean surgical margins. However, 80% of ameloblastomas are placed

in the mandible and, among these, 70% are detected in the ascendant ramus or molar region while 20% in the premolar area, and only 10% in the anterior region.^[2]

They are usually discovered around the fourth and the fifth decades of life, with the exception of the unicystic variant, which is most common between 20 and 30 year old.^[4]

The symptoms of this tumor are few, and swelling represents the most frequent.

From a radiological point of view ameloblastoma is observed as a radiolucent area which may present three different patterns. The most common form is the multilocular with various cysts which are in groups or separated by osseous septa giving a soap bubble appearance. The beehive form can be considered as second in frequency. The unilocular form is the third in frequency.^[5]

From a histological point of view, literature reports different entities, some of which have prognostic relevance. Multicystic and unicystic represent the two main biologic macroscopic subtypes.^[6,7] However, others rare varieties of this tumor such as peripheral ameloblastoma have been described.^[2,8,9, 10]

Unicystic and multicystic ameloblastoma

Multicystic ameloblastoma is the most frequent subtype and it generally brings about marked facial deformities and serious debilitation. Moreover, as it tends to infiltrate cancellous bone trabeculae, despite accurate curettage, the incidence of recurrence rates is up to 90%.^[6,7]

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Histologically, solid multicystic ameloblastoma is characterized by the occurrence of islands, strands and irregular configurations of tumor epithelium, consisting of a central mass of polyhedral cells resembling stellate reticulum surrounded by a layer of cuboidal or columnar cells, similar to pre-ameloblasts. When degeneration of centrally placed cells occurs in several tumor islands, the term multicystic is often used.

Unicystic ameloblastoma can be considered as a variant of the solid or multicystic ameloblastoma. The two tumors show similar epidemiological features with a male-female ratio of 1:1.3. The most common affected sites are molar areas and the ascending ramus. Impacted teeth, especially third molars, are associated with multicystic ameloblastoma, although several reports underline that also unicystic ameloblastoma can be caused by impacted third molar teeth.^[2,5]

Solid multilocular ameloblastoma often have a soap bubble appearance, whereas there is a unilocular configuration in 24–47% of multicystic ameloblastoma.^[8]

The unicystic subtype is considered as a less aggressive tumor with a variable recurrence rate according to different authors. Unicystic ameloblastoma is a monocystic lesion and it usually shows quite a large cystic cavity with a lining composed of ameloblastic cells. It may also present one or more nodules arising from the cyst and projecting into the lumen of the cyst cavity comprising odontogenic epithelium with a plexiform pattern which may mimic a plexiform ameloblastoma.^[2]

Finally, a few unicystic ameloblastomas may have one more mural nodules or local thickenings of the cyst wall. Nodules comprise invasive islands or strands typical of solid multicystic ameloblastoma.^[2]

A variant without particular clinical relevance is the desmoplastic ameloblastoma characterized by a uniformly dense collagenous stroma with small nests and strands of compressed odontogenic epithelium.^[11,12]

In the scientific literature, other uncommon variants have been described, such as the kerato ameloblastoma and its papilliferous variant, as well as ameloblastoma associated with calcifying odontogenic cysts or diffuse mineralized dental tissue deposits similar to odonto-ameloblastoma.^[8,9,13]

In addition, some focal microscopic differentiations with no particular clinical relevance have been described in the literature. They include mucous cell differentiation, adenomatoid changes, and HPV 18-positive verrucous lesion in a cystic cavity of ameloblastoma.^[14-16]

It should be emphasized that although the concepts of unicystic and multicystic ameloblastoma, as described

above, were introduced more than 30 years ago by Robinson and Martinez,^[17] confusion still exists when clinicians and pathologists discuss this lesion.

Such confusion particularly involves the terminology of UA. The term unicystic comes from the macro and microscopic appearance since this lesion is a well-defined single cystic sac lined by odontogenic epithelium.

The term unilocular is used in a radiological sense in order to describe only one locus of radiolucency. The confusion derives from the fact that unicystic ameloblastoma can be observed as either unilocular or multilocular bone defect.

Therefore, this article has the aim of reviewing the recent literature about unicystic ameloblastoma using our unusual clinical case as a starting point to illustrate this discussion.

Case Report

A 30-year-old man who had been complaining of slight pain in the premolar and molar area of the left side of mandible for few weeks, had a check up at the department of dentistry at Versilia Hospital.

Extraoral examination revealed a slight swelling measuring 1 × 2 cm in the left area of mandible. The margins were not clearly distinct and the swelling was hard in consistency. The skin and the oral mucosa which covered the area were normal. Intraoral examination showed the presence of intercalated edentule areas. Teeth 3.2–3.3 and 3.8 appeared in good condition [Figure 1].

X-rays revealed a well-defined unilocular radiotransparency with radiopaque margins extending from the central incisor to the molar area but no signs of displacement of the roots were observed. Teeth 3.2 and 3.3 showed previous endodontic treatment [Figure 2].

A provisional diagnosis of radicular cyst was considered and the lesion was removed by osteotomy and curettage [Figures 3 and 4]. There were two specimens from a large grey irregular cyst with corrugate margins. The larger one measured 4 × 4 cm while the smaller 2 × 1 cm [Figures 5 and 6].

The first histological diagnosis was an odontogenic cyst.

The histological diagnosis indicated that the wall of the cyst was made up of pluristratified pavementous epithelium surrounded by fibrous tissue. An interesting finding was that inside the wall, islands of ameloblastic epithelium reactive to Calretinin and Cytocheratin 8 and 18 were detected.

Successive histological evaluations demonstrated other rarities. The ameloblastic epithelial islands were observed



Figure 1: Intraoral examination revealed a swelling area in the premolar and molar zone, where the patient has been complaining of mild pain. It should be observed that the oral mucosa which covered the area was normal

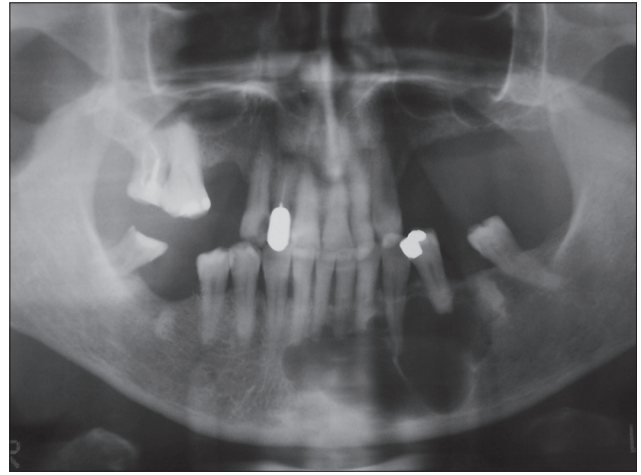


Figure 2: X-rays indicated a well-defined unilocular radiotransparency with radiopaque margins extending from the central incisor to the molar area. No signs of displacement of the roots were observed

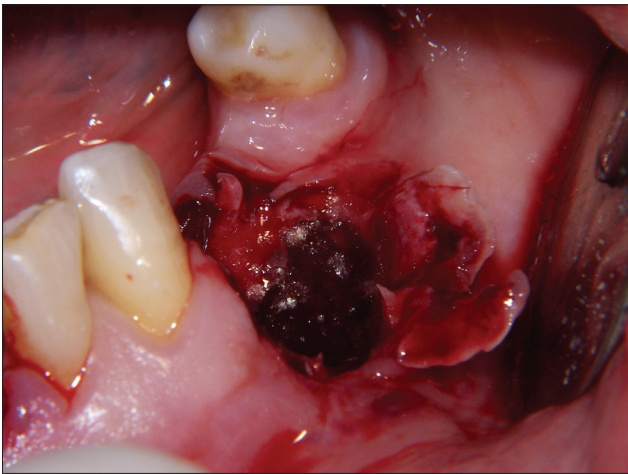


Figure 3: This image shows the lesion after flap elevation. It should be noted that the cortical plate has been partially destroyed and the cyst is in direct contact with soft tissues

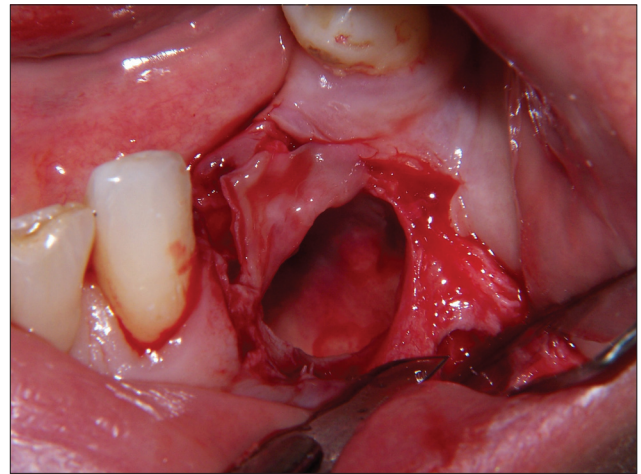


Figure 4: The surgical intervention has been carried out so as to remove integrally the cyst. Therefore a trap door has been created and, using a spoon, the surgeon has carefully removed the lesion. This fact allows a macroscopic evaluation of the cyst by a pathologist



Figure 5: This image shows the larger specimen from a large grey irregular cyst with corrugate margins. It measured 4 x 4 cm



Figure 6: This image shows the smaller specimen from the cyst. It measured 2 x 1 cm

in lasso connective tissue and in some areas they presented the typical fenced epithelium.

At the time of writing, the patient has had a 6-month follow up check without any evidence of recurrence.

Discussion

Literature considers two different types of intraosseously located ameloblastomas, the solid or multicystic variant and the unicystic form. As described above, the multicystic variety appears as a solid tumor or multicystic as a result of degeneration of central islands of the tumor while, the unicystic form is a single well-defined lesion made up of odontogenic epithelium with ameloblastic appearance and stratified squamous epithelium in remaining areas. Several attempts to classify unicystic ameloblastoma have been made but there is still some confusion.

Classifications

The first attempt to classify unicystic ameloblastoma was that of Robinson and Martinez^[17] who divided into three different subtypes. They considered ameloblastoma only if one or more of the following criteria were present:

- in the lining epithelium the basal cells were clearly columnar with hyperchromatic nuclei and the overlying cells were only loosely textured with the absence of “cohesiveness”
- Downgrowth of ameloblastic epithelium into the connective tissue portion of the cyst wall
- Presence in the connective tissue portion of the cyst wall of islands composed of a periphery of columnar epithelial cells and a centre identical to stellate reticulum
- Intraluminal nodules composed of anastomosing cords and islands of epithelium

Several years later, Ackermann *et al.* first divided these entities into the following three histologic groups^[18]:

- Luminal UA: Tumor confined to the luminal surface of the cyst
- Intraluminal/ plexiform UA: Nodular proliferation into the lumen without infiltration of tumour cells into the connective tissue wall

- Mural UA invasive islands of ameloblastic epithelium in the connective tissue wall not involving the entire epithelium

More recently the classification of Ackermann *et al.* has been modified by Philipsen and Reichart,^[19] considering:

- Subgroup 1: Luminal UA
- Subgroup 1.2: Luminal and intraluminal
- Subgroup 1.2.3: Luminal, intraluminal and intramural
- Subgroup 1.3: Luminal and intramural

These authors indicate that it is well known that unicystic ameloblastomas show a combination of various histological features so their classification might be more appropriate. Moreover, they underline that it can be useful in order to plan the treatment. Thus, they indicate that a tumor in subgroup 1 and 1.2 can be treated conservatively, while a more invasive approach should be followed in the other two categories.^[19]

Literature survey

Literature reports several cases of ameloblastomas apparently arising from what was wrongly considered an odontogenic cyst. Although more than 90 publications have described one or more cases of ameloblastomas at least, most of these publications are difficult to find or lack adequate radiographical images and microscopic analysis.

In addition, confusion in ameloblastoma nomenclature makes any comparison impossible.

Our literature review analyzed Unicystic ameloblastoma considering 233 cases [Table 1].

The mean age at the time of diagnosis of UA is strongly related to the possible association between an impacted tooth and the tumor.

Philipsen *et al.*^[19] divided their review into cases with an initial presentation of dentigerous cyst, where the mean age was 16.5 years old and non-dentigerous cysts with a mean age of 35.2 year old.

Li *et al.*^[20] described their 15 years experience in treating

Table 1: Unicystic ameloblastoma cases in the scientific literature

Authors	Number of cases	Age	Location
Philipsen, <i>et al.</i>	193	(initial presentation as dentigerous cyst)16.5, 35.2	Maxilla /Mandible 3/13 (ratio)
Li, <i>et al.</i>	33	25.3	Maxilla 3 Mandible 30
Gordon, <i>et al</i>	1	Not found	Maxilla
Navarro, <i>et al</i>	1	17	Maxilla
Patel, <i>et al.</i>	1	14	Not found
Sivapathasundharam, <i>et al</i>	1	28	Mandible
Paikkatt VJ, <i>et al</i>	1	11	Maxilla
Oliveira-Neto HH, <i>et al</i>	1	11	Mandible
Quereshi SS, <i>et al</i>	1	10	Mandible

ameloblastomas, considering 33 unicystic ameloblastomas, and the mean age was 25.3 year old although peaks were observed in the second and third decades.

Quereshi *et al.*^[21] reported an ameloblastoma in a 10-year-old girl which was masquerading a cancer lesion.

Moreover, both Oliveira-Neto *et al.*^[22] and Paikkatt *et al.*^[23] described two cases in 11-year-old children.

More recently two cases in a 17-year-old male and in a 14-year-old female with a Gardner syndrome were reported.^[22,24]

On these bases, it is confirmed that association of an impacted tooth with the ameloblastoma is crucial in determining the age of diagnosis.

With regard to the location of the tumor, the literature underlines a marked prevalence for mandible. Philipsen *et al.*^[19] in their review indicated a ratio of 3 to 13 in favor of mandible.

Li *et al.*^[20] reported only 3 cases of tumour in maxilla versus 30 cases in mandible.

Up to now, three other cases are described in the maxilla. Gordon *et al.*^[25] evaluated a strange association between an osteoblastoma at the apex of a molar associated with a posterior maxillary ameloblastoma.

Navarro *et al.*^[24] showed a unicystic ameloblastoma in the anterior maxillary area underlining the importance of a differential diagnosis when it occurs in that area.

Recently, Paikkatt *et al.*^[23] observed an ameloblastoma in the area of premaxilla in a young teenager. A provisional diagnosis of radicular cyst was made.

Oliveira *et al.*^[22] presented a unicystic ameloblastoma involving an unerupted inferior second premolar in a girl under orthodontic treatment, while both Quereshi *et al.*^[21] and Sivapathasundharam *et al.*^[26] evaluated two tumors in the posterior mandible.

All authors agree about the fact that posterior mandible including ascending ramus are the most affected areas.

Concerning histology, it is impossible to compare data due to the fact that histological classifications were made in few publications.

Philipsen *et al.*^[19] in their review showed that two thirds of unicystic ameloblastomas showed invasive ameloblastic tissue in the wall of the cyst. They underlined that this histological pattern occurred more frequently in the “no impaction” category.

It is worth observing that in the case series reported by Ackermann *et al.*^[18], Leider *et al.*,^[27] and Wang^[28] a large number of the tumors presented intramural nodules.

The article of Ng *et al.*^[29] reveals an interesting aspect to evaluate, which is the possible correlation between tooth impaction and infiltrative potential. This should represent a future topic of the research.

A case of unusual histological presentation was described by Sivapathasundharam *et al.*,^[26] who observed an unicystic ameloblastoma with luminal and intramural plexiform epithelial proliferation with typical dentin in the connective capsule.

Ngwenya *et al.*^[30] reported two cases where the histological analysis was crucial to determine the true nature of unicystic ameloblastomas.

In our case report, the lesion looked like an odontogenic cyst, but a careful histological analysis revealed an unusual type of mural ameloblastoma, with ameloblastic cells only in few areas. These cells were surrounded by a thick layer of fibrous connective tissue [Figures 7 and 8]. This could be due to mesenchymal induction mediated by neoplastic ameloblasts [Figure 9].

Although Philipsen *et al.*^[19] recommended performing an enucleation after a biopsy failed to show mural invasion, literature lacks general agreement about a surgical approach.

The article of Ngwenya^[30] *et al.* emphasized that macroscopic examination of serial sections of ameloblastomas provide information that an examination of randomized biopsies cannot give. Evans *et al.*^[15] also described an interesting case report where microscopic changes similar to an adenomatoid odontogenic tumor on a small incision biopsy confused the correct diagnosis of ameloblastoma.

On these bases, it should be kept in mind that the first histopathologic analysis of our specimen revealed a harmless odontogenic cyst although further investigations discovered that it was an unusual presentation of ameloblastoma. In the light of our case report, we believe that a careful histological analysis using different biopsies by the same specimen is crucial to determine true nature of the lesion.

Gardner *et al.*^[31] reported that the recurrence rate for conservative surgical treatment is less than 25%, whereas other authors have described a recurrence rate of 10–20%.^[25] However, it should be noted that few case reports evaluated the follow-up period. In their review, Philipsen *et al.*^[19] underline that there seem to be differences in recurrence rates between the intraluminal subtype and the intramural subtypes although sufficient data to support this hypothesis are not available yet.

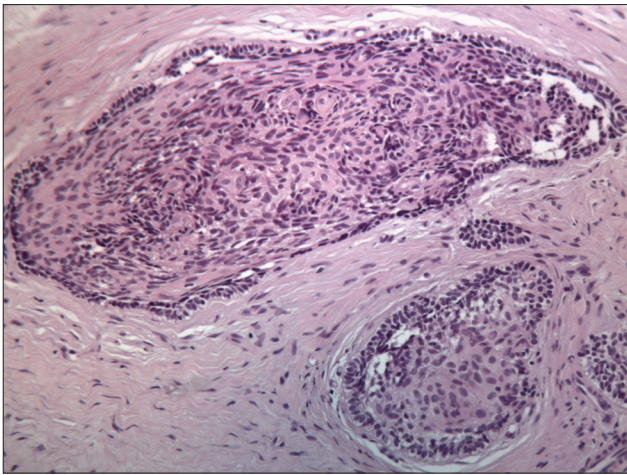


Figure 7: Typical islands of ameloblastic epithelium surrounded by connective tissue

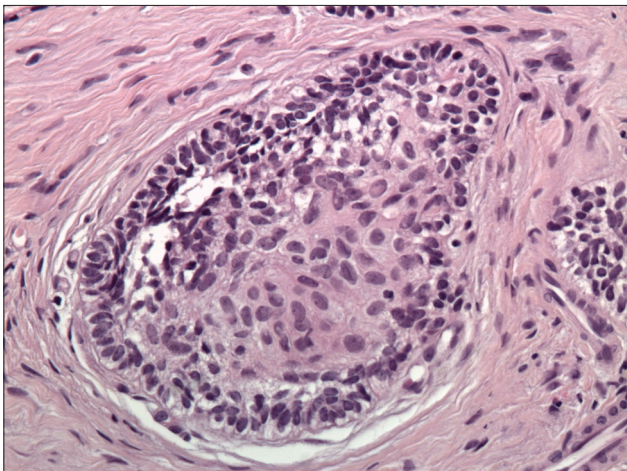


Figure 8: This is a histological image of islands of ameloblastic epithelium presenting the typical peripheral fenced epithelium

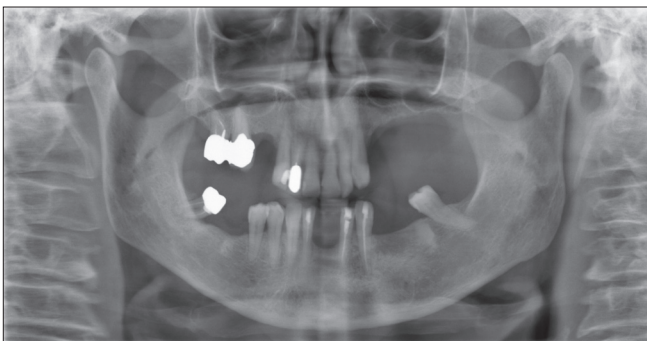


Figure 9: Radiological evaluation after 6 months indicates no evidence of recurrence of the lesion

Unicystic ameloblastoma has a better prognosis with a lower recurrence than multicystic ameloblastoma, although it should be remembered that the tumor epithelium may break the periphery of the capsule infiltrating the surrounding cancellous bone.

All in all, this literature review supports the idea that unicystic

ameloblastomas may appear in different histologic subtypes, and further studies need to clarify what the behavior of each different tumor is.

We think that our case report provides new insights in general knowledge of unicystic ameloblastomas. It is unusual to find an unicystic lesion with few islands of tumor epithelium in the thick of a cyst, and this fact should induce surgeons and pathologists to consider carefully each lesion. A preoperative biopsy can only be representative for the entire lesion in extremely few instances and will probably result in incorrect classification and diagnosis. Thus, the true nature of the lesion can be evident when the entire specimen is available for microscopy. It should be emphasized that this approach was expressed by Ackermann^[18] *et al.* first several years ago. Pathologists should make multiple or serial sectioning to search cell and tissue configurations in cyst wall nodules.

Thus, if tumor islands are found intramurally, a more aggressive surgical approach should be carried out, considering removal of adjacent bone and a follow up period of at least 10 years.

However, further studies are necessary to clarify both what approach the surgeon in relationship with the specific ameloblastoma subtype must consider and the exact period of follow up of each subtype of tumor.

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