Cystic Mandibular Lesion in the Antiquity. A Rare Finding

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ABSTRACT Ameloblastoma is a not uncommon tumour reported in the clinical literature and is characterised by a multilocular cavity with ‘soap bubble’ pattern usually in the posterior body of the mandible. A review of the palaeopathology literature did not reveal any examples of ameloblastoma. In this paper, a probable case of ameloblastoma in a male skeleton, 45–55 years at age of death, from Casserres, Barcelona, Spain, (ca AD V–IX centuries) is presented. This case was identified during the anthropological study of the remains in the laboratory of biological anthropology in the Universitat Autònoma de Barcelona. The mandible of this individual (AEC07-C2-139) exhibits unusual features, like cavitated lesions in the posterior side of the right gonion and the expansion of the right ramus of the mandible. All these features are consistent with ameloblastoma. Copyright © 2011 John Wiley & Sons, Ltd.

Key words: ameloblastoma; antiquity; Catalonia; keratocyst; multilocular; Spain; tumour

Introduction

Cavitated lesions in jaws are frequently clinical radiological findings. The most usual lesions in jaws are those of inflammatory origin, being radicular cysts and lateral periodontal cysts the commonest. However, mandibular cavitated lesions could have a tumoural or pseudotumoural, traumatic, infectious or idiopathic origin.

The skeletal remains allow the finding of this kind of lesions directly. Nevertheless, it is unusual to find them in the antiquity. The most frequent palaeopathological conditions related to the jaws and teeth are fistulae, ante mortem tooth loss, calculus, caries, hypoplasia, hypercementosis, pulp chamber exposure and alveolar resorption, but tumour or cystic lesions are very uncommon.

We present an ancient case of a mandibular osteolytical lesion and perform a differential diagnosis among the most frequent entities that can cause it.

Material and methods

The lesion analysed here was found in a mandible from a mature (45–55 years old at death) male from the early Medieval Age (AD V–IX centuries) coming from the ‘Accés Est Casserres’ archaeological site, a necropolis near the village of Casserres (Barcelona, Catalonia, Spain). Sex and age were determined according to the skeletal morphological traits (Ferembach et al., 1980; Krogman & Iscan, 1986). The AEC07-C2-139 individual was buried in a stoned tomb near a basilica.

The skeleton is almost complete and the bone tissue is relatively well preserved. The cranium is rather complete although the upper maxilla appeared partially broken. No signs of pathology affecting the bone were observed. The mandible presents a unilateral lesion.

To discuss the possible aetiology and patogenicity of the lesion in the mandible of the individual, macroscopical and radiological analysis were performed (Whaites, 2007). Individual criteria referred to biological profile had also been used (age and sex) (Neville et al., 2002; Coulthard et al., 2003; Sapp et al., 2004; Cawson & Odell, 2008).

Results

The individual from Casserres shows several skeletal and oral lesions. In this sense, the right elbow shows some signs compatible with degenerative osteoarthritis. It was probably due to a traumatic lesion on the epicondyle of the humerus, and it could happen in an.
early age of his life. The vertebrae also show signs of slight arthritis, and the calcaneus presents osteophytes on the Achilles tendon attachment.

In relation to the oral lesions, four caries affecting the mandibular teeth were detected: one located in the enamel–cemental junction (tooth 47) and three in the dental crown (teeth 34, 35 and 45). The first mandibular molars (36 and 46) were lost ante mortem, and the bone surrounding was resorbed. No fistulae were observed. Almost all the teeth in the mandible present calculus on their surface.

The mandible presents a unilateral and multilocular lesion (4 × 3.5 cm), with internal septa forming a ‘soap bubble’ pattern, in the posterior side of the right gonion (Figures 1 and 2). The limits of the lesion present a normal tissue, indistinguishable from the rest of the bone, and the inner bone is rough. In the radiological image, three differenced radiolucent lobes with some radiopacities can be observed (Figure 3). The lack of bone in the anterior wall of the lesion does not let know if more lobes could form part of the tumour.

The morphological analysis suggests that the lesion did not display a cortical disruption. Therefore, a malignant aetiology could be discarded. The size and shape of the lesion could also discard inflammatory, traumatic, infectious and pseudotumoural injuries.

The most frequent causes of mandibular lesions have been reviewed. Taking into account the previous considerations, differential diagnosis was done among the following entities described in the most significant oral medicine-related and pathology-related books (Neville et al., 2002; Coulthard et al., 2003; Sapp et al., 2004; Cawson & Odell, 2008).

**Differential diagnosis**

**Dentigerous cyst:** the most frequent is the unicystic and solid type, whereas Casserres mandible is multicystic. It is always associated to the crown of a tooth in development or without outbreak. It is adhered to the cervical portion of the tooth. Most of them manifest in adolescents and young adults.

**Adenomatoid odontogenic tumour:** it is frequent in the second decade of life, more in women than in men. It appears more frequently in the anterior lower jaw and maxilla. It is related to a retained tooth. Neither position nor tooth retention coincide with our specimen.

**Ameloblastic fibroma:** represents about 2% of all odontogenic tumours. It predominates in children and young adults. It appears more commonly in the mandible (80–90%). It is a radiolucent lesion. Neither the radiological pattern nor the age coincides with our specimen.

**Traumatic bony cyst:** it is usually a unilocular cyst. The cause of this injury is unknown and because of the lack of an epithelial cover, it cannot be classified like a true cyst. Traumatism is not supported by radiological analysis. The lesions are usually revealed in the second decade of life. Neither the shape nor the age agrees with our specimen.

**Central granuloma of giant cells:** it affects more women than men in a proportion of 2:1. It is manifested as a solitary injury (early lesions) with a mixed radiolucent

Figure 1. Superior view of the mandible of the individual from Casserres showing a big bone expansion in the body of the right hemimandible.

Figure 2. Detail of the multilocular lesion.
and radiopaque pattern. It is frequent in children and young adults (75%) more often located in the jaw previous to molars. The specimen of Casserres belongs to a mature male individual, and the radiological pattern is completely different.

*Lateral periodontal cyst*: it is located adjacent or lateral to the root of a tooth. Its more frequent location is in premolar and canine areas, sometimes in the incisive zone. The situation of the lesion in our case discards this entity.

*Cemento-ossifying fibromas* are benign lesions affecting the jaws and other craniofacial bones. They commonly affect adults between the third and fourth decade of life. Radiographically, they appear as well-defined unilocular or multilocular intraosseous masses, commonly in the premolar/molar region of the mandible. The lesion is invariably encapsulated and of mixed radiolucent densities.

![Radiological image](image_url)

Figure 3. Radiological image of the lesion. Multilocular morphology can be appreciated.

<table>
<thead>
<tr>
<th>Ameloblastoma</th>
<th>OKC</th>
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<tbody>
<tr>
<td>Odontogenic epithelial neoplasia from any tissue</td>
<td>Neoplasia from dental sheet</td>
</tr>
<tr>
<td>Wide range of histological patterns</td>
<td>Parakeratocyst neoplastic form</td>
</tr>
<tr>
<td>Incidence between 30 and 50 years old</td>
<td>Incidence at any age</td>
</tr>
<tr>
<td>Locally invasive</td>
<td>Locally invasive</td>
</tr>
<tr>
<td>Asymptomatic</td>
<td>Asymptomatic</td>
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<tr>
<td>Mainly multilocular, radiolucent</td>
<td>Mainly unilocular, radiolucent</td>
</tr>
<tr>
<td>Frequently in the posterior zone of the mandible</td>
<td>Frequently in the posterior zone of the mandible</td>
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<tr>
<td>Treatment: enucleating</td>
<td>Treatment: enucleating</td>
</tr>
<tr>
<td>Recidivist, lethal if it achieves the cranial base</td>
<td>Recidivist</td>
</tr>
<tr>
<td>Radiolucent</td>
<td>Radiolucent</td>
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The *odontogenic myxoma* is an odontogenic tumour arising from the dental papilla. It is more common in young adults and affects the mandible more than the maxilla. Usually, the odontogenic myxoma is painless but can cause a swelling. On radiographs, it appears as a multilocular radiolucency (dark area) with short, straight trabeculae. As a result, this appearance is usually described as ‘honeycomb-like’.

*Clear cell odontogenic carcinoma* is a rare odontogenic tumour that tends to occur in the mandible of older adults with a predilection for women. Clear cell odontogenic carcinoma is a rare odontogenic tumour with female predilection and peak age incidence in the fifth to seventh decades of life. In contrast with the case being reported where the lesion occurred in the posterior region of the jaw, studies have shown that the anterior portions of the jaws especially the mandible are most frequently affected.

*Odontogenic keratocyst (OKC)*: it represents about 5–15% of all jaw cysts. It has a high recurrence rate (50%) and develops more aggressively than any other jaw cysts. Patients in their second to fourth decades of life are affected most commonly. Odontogenic keratocyst usually appears as a unilocular lucent lesion, with smooth corticated borders, but could be multilocular too. It is associated with an impacted tooth. Odontogenic keratocyst may cause cortical thinning, tooth displacement and root resorption. It is most commonly located in the body and ramus of the mandible. In spite of some morphological differences (Table 1) with the Casserres specimen, OKCs cannot be completely discarded without a histological analysis.

*Ameloblastoma*: it is a benign epithelial neoplasm and represents about 10% of odontogenic tumours. It manifests in the third to fifth decades of life. It occurs in the ramus and posterior body of the mandible (80% cases). It could be unilocular or multilocular. It is the most probably diagnosis for the presented case.
Discussion

Despite the not uncommon occurrence of the lesion today, apparently, in palaeopathological literature, very few references of mandibular cyst have been found. It is probable that the multicystic cyst found by Salama & Hilmy (1951) in the mandible ramus of a 2800 BC Egyptian mummy (V dynasty) was a keratocyst. Also, a composed cystic lesion in a 1200-year-old prehistoric American Indian was reported as a possible multilocular cyst or ameloblastoma (Neiburger, 1977). On the other hand, Chimenos-Küstner & Agustí-Farjas (2006) described a similar case belonging to a male individual of Santa Magdalena d’Empuries of the same chronology of that of Casserres.

Differential diagnosis of the lesion in the mandible of the individual from Casserres can be carried out among keratocyst and ameloblastoma by similarity in the age of appearance, sex of the individual, recurrence and site of appearance (Table 1). Taking into account the most possible alternatives, the age and sex of the individual, the multicystic form with the ‘soap bubble’ pattern, the specific location and the radiological characteristics, we suggest ameloblastoma as the first option of diagnosis.

Ameloblastoma is the most common clinically significant odontogenic tumour. Its relative frequency equals the combined frequency of all other odontogenic tumours. Ameloblastomas are tumours of odontogenic epithelial origin. Theoretically, they may arise from rests of dental lamina, from a developing enamel organ, from the epithelial lining of an odontogenic cyst or from the basal cells of the oral mucosa.

Ameloblastomas are slow growing, locally invasive tumours that run a benign course in most cases. They occur in the three following different clinical radiographic situations:

Conventional solid or multicystic (about 86% of all cases), unicystic (about 13% of all cases) and peripheral (extraosseous) (about 1% of all cases).

Conventional solid or multicystic intraosseous ameloblastoma (presented case) is encountered in patients over a wide age range. It is rare in children younger than age 10 and relatively uncommon in the 10- to 19-year-old group. The tumour shows an approximately equal prevalence in the third to seventh decades of life. There is no significant gender predilection. About 85% of conventional ameloblastoma occur in the mandible, most often in the molar-ascending ramus area. About 15% of ameloblastoma occur in the maxilla, usually in the posterior regions. The tumour is often asymptomatic, and smaller lesions are detected only during a radiographic examination. A painless swelling or expansion of the jaw is the usual clinical presentation. If untreated, the lesion may grow slowly to massive or grotesque proportions. Pain and paraesthesia are uncommon, even with large tumours. The most typical radiographic feature is that of a multilocular radiolucent lesion. The lesion is often described as having a soap bubble pattern when the radiolucent loculations are large and as being honeycombed when the loculations are small. Buccal and lingual cortical expansion is frequently present. Resorption of the roots of teeth adjacent to the tumour is common. In many cases, an unerupted tooth, most often a mandible third molar, is associated with the radiolucent defect.

Conclusions

This study describes a probable case of ameloblastoma in a male, aged 45–55 years at death, from Casserres, Barcelona, Spain (ca AD V-IX centuries). Through the comparison with other cases and the clinical aspects of the lesion, it was demonstrated that the individual exhibits a pattern of odontogenic tumour similar to that seen in ameloblastoma patients. Specifically, multilocular lesions and a radiolucent radiological pattern combine to provide compelling evidence for the diagnosis of ameloblastoma, although without a histological screen test, OKC cannot be dismissed.

Oral lesions and pathology are used in anthropology as elements of characterization of different populations. The most common lesions analysed in this context are caries, alveolar bone loss, fistulae and other entities related to teeth. However, it is interesting to note that all kinds of diseases and disorders could be found in ancient remains, so the Casserres case could be the most ancient case of ameloblastoma published.

Acknowledgments

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References


Ameloblastoma in a Late Roman Individual


