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CASE REPORT

Synchronous ameloblastoma and orthokeratinized odontogenic cyst of the mandible

Eduardo Rodrigues Fregnani[1], Danyel Elias da Cruz Perez[1], Fernando Augusto Soares[2], Fábio Abreu Alves[1]

Departments of [1]Stomatology and [2]Pathology, A. C. Camargo Cancer Hospital, Sao Paulo, Brazil

The simultaneous occurrence of ameloblastomas with odontogenic cysts or other non-odontogenic lesions have already been described as combined lesions. However, we are unaware of any report in the English literature of simultaneous occurrence of ameloblastoma and orthokeratinized odontogenic cyst (OOC) occurring as completely distinct lesions. This report shows a case of synchronous ameloblastoma and OOC, located on posterior regions of the mandible, but in distinct sides.
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Case report

A 21-year-old white man was referred to the Department of Stomatology, AC Camargo Cancer Hospital, Sao Paulo, Brazil, complaining of a painful swelling on the right posterior mandible for 4 months of evolution. On extraoral examination, there was a facial asymmetry due to swelling on the region of right body of the mandible, painful to palpation. Intra-oral examination revealed an expansion of all vestibular bone cortical of the right body and ascending ramus of the mandible, measuring 7.0 × 5.0 cm of extension. The lesion presented hard consistency and the superficial mucosa was intact and presenting normal colour.

The panoramic radiography showed a large multilocular radiolucency of the right angle and ascending ramus of the mandible, which included a displaced impacted third molar and apparently caused disruption of cortical bone in the region of angle. Interestingly, on the left side of the mandible, it was observed a well-defined unilocular radiolucency on the body of the mandible that was also associated with the impacted third molar (Fig. 1). Computerized tomography showed both lesions to be well circumscribed and defined, characterized by low-density masses involving the right and left ascending ramus, causing enlargement of lingual and vestibular bone cortical (Fig. 2). The main clinical and radiographic hypothesis of diagnosis was multiple odontogenic lesions, probably multiple odontogenic keratocysts (OK).

Incisional biopsy of both lesions was performed and histopathological diagnoses of the lesions were plexiforme ameloblastoma (right side) and OK (left side). Under general anaesthesia, the patient underwent bilateral tumour resection and both were carried out conservatively with extensive bone curettage associated with exodontias of the right and left inferior second molars and both impacted third molars, without trans- or post-operative complications.

Microscopically, the lesion localized on the right mandible presented multiple coalescent cords and islands of epithelial cells where the peripheral cells resemble ameloblasts with inverted polarity of their nuclei, embedded in a loosely stroma (Fig. 3). The other lesion, sited on the left side, showed a cystic cavity lined by squamous and stratified epithelium with a thick layer of orthokeratin in all its extension. Prominent keratohyaline granules were observed adjacent to orthokeratin layer and no prominent basal cell layer was observed (Fig. 4). According to clinical, radiographic and histopathological features, the diagnosis of synchronous plexiform ameloblastoma (right mandible) and orthokeratinized odontogenic cyst (OOC; left mandible) was established.

After 8 years of clinical and radiographic follow up, no signs of recurrences are observed (Fig. 5).

Comments

According to the World Health Organization (1), OOC is a developmental cyst that could be compared with the uncommon orthokeratinized type of OK but only based on histology. However, OOC occurs mainly in patients during the fourth and fifth decades of life (OK occurs

Correspondence: Eduardo Rodrigues Fregnani, Department of Stomatology, A. C. Camargo Cancer Hospital, Rua Professor Antônio Prudente, 211 – Liberdade – CEP: 01509-010, São Paulo/SP, Brazil. Tel: +55 11 2189 5129, Fax: +55 11 2189 5083, E-mail: erfreg@terra.com.br
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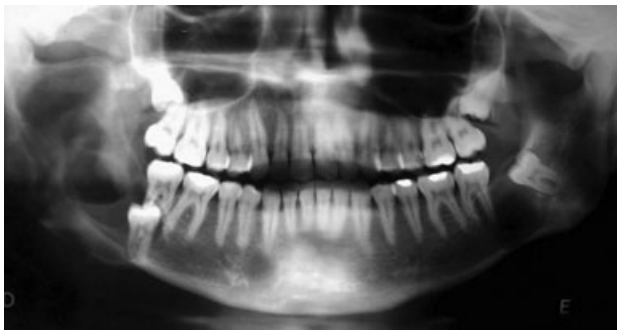

Figure 1 Panoramic radiography showing a multilocular radiolucency of the right posterior mandible and on the left side a unilocular radiolucency on the body of the mandible associated with the impacted third molar.

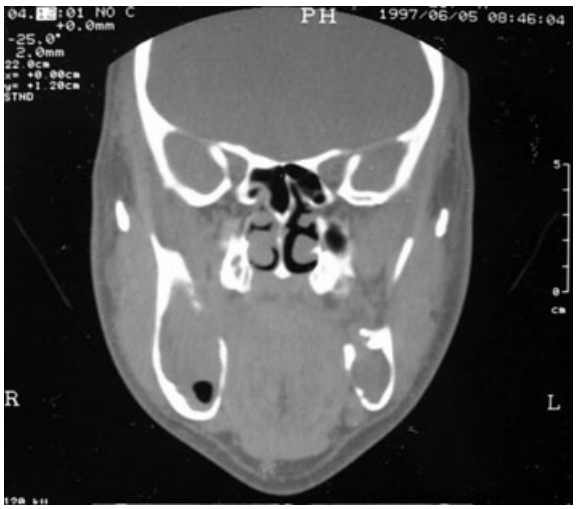

Figure 2 Computerized tomography (coronal view) showing well circumscribed and defined lesions causing enlargement of lingual and vestibular bone cortical.

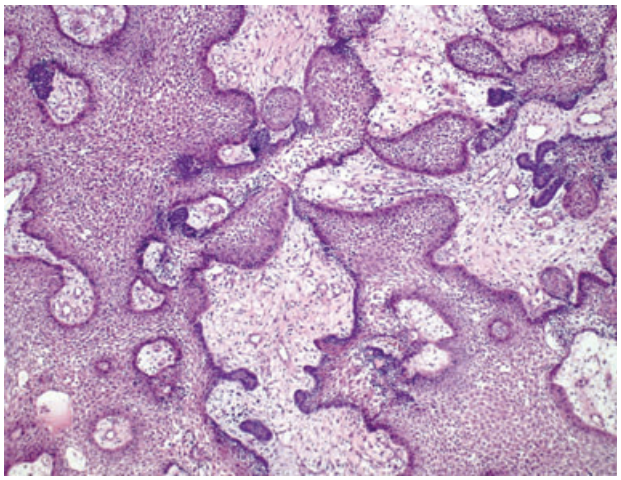

Figure 3 Plexiform ameloblastoma presenting multiple coalescent epithelial cell cords where the peripheral cells resemble ameloblasts (haematoxylin and eosin, original magnification, ×100).

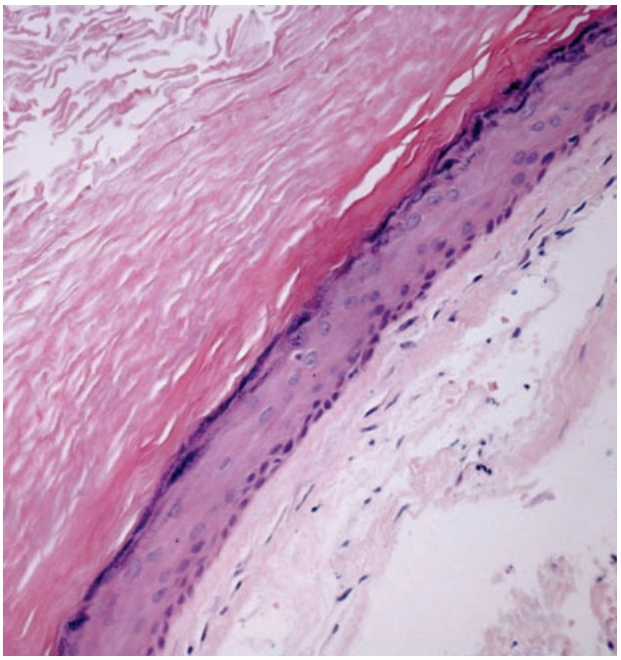

Figure 4 Histopathology of orthokeratinized odontogenic cyst showing a cystic cavity lined by squamous epithelium with a large layer of orthokeratin in all its extension (haematoxylin and eosin, original magnification, ×100).

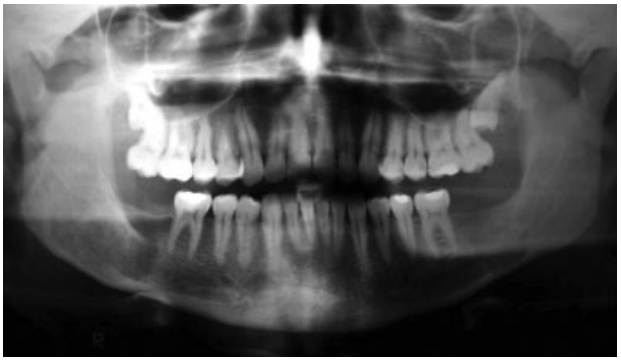

Figure 5 Panoramic radiography showing bone formation and no signs of recurrences after 8 years.

commonly in the second and third decades), and it has no tendency to recur and is not related to the nevoid basal cell carcinoma syndrome, as it is observed in OK. Moreover, OCC presents histopathological and immunohistochemical features different from OK, strongly suggesting that these are distinct entities (2). In our case, different from OK, the epithelial lining was fully recovered by non-corrugated orthokeratin, with evident granular layer extended through all epithelium, and no prominent basal cell layers arranged in palisaded were observed.

All previous reports that showed the occurrence of simultaneous odontogenic lesions or simultaneous odontogenic and non-odontogenic lesions, described combined lesions, some times called hybrid lesions (3–5). Differently, the case described here represents the

occurrence of two completely independent lesions. In our case, the presence of two mandibular radiolucent lesions led us to suspect multiple OKs. However, an incisional biopsy allowed a definitive diagnosis as two pathological distinct lesions before surgery.

The patient was treated by surgical excision associated with extensive curettage. The conservative approach to treatment of solid ameloblastomas is still a matter of critics, although recent studies showed good results (6). In our case, a long follow-up period (8 years) without recurrence confirms its success. Occurrence of two distinct and simultaneous odontogenic lesions in the same patient is extremely uncommon. To our knowledge, synchronous ameloblastoma and OOC occurring as distinct lesions have not been previously reported in the English language literature.

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