

Calcifying odontogenic cyst associated with compound odontoma: A study on undemineralized material

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SUMMARY

In a minority of cases of calcifying odontogenic cyst (COC) it is possible to observe the formation of dental hard tissues in the cyst wall. The use of undemineralized sections has allowed an evaluation of the mineralized tissues normally lost with the use of demineralizing agents. All the dental hard tissues presented a high degree of morpho- and histodifferentiation. The histochemical staining for calcium salts (von Kossa) showed the presence of areas of low mineralization in the portion of the lesion, where the tissue maturation was not complete. In conclusion the appearance of the dental hard tissues in this case of compound odontoma arising in the cyst wall of a COC is similar to that already described in compound odontoma not associated with COC.

KEY WORDS:

Calcifying odontogenic cyst, Odontoma, Undemineralized sections.

RÉSUMÉ

Il est possible d'observer la formation de tissus dentaires durs dans la paroi d'une minorité de cas de kystes odontogènes calcifiants (KOC). L'utilisation de coupes de tissus non déminéralisés a permis une évaluation des tissus minéralisés qui sont habituellement perdus après décalcification. Tous les tissus dentaires durs présentent un degré important de morpho- en d'histodifférenciation. La coloration histochimique des sels calcaires (von Kossa) montre la présence de territoires de faible minéralisation dans les régions dans lesquelles la maturation tissulaire est incomplète.

En conclusion, l'apparence des tissus dentaires durs dans ce cas d'odontome composé développé dans la paroi d'un kyste odontogène calcifié est semblable à celle déjà décrite dans les odontomes composés non associé à un KOC.

MOTS CLÉS:

Kyste odontogène calcifiant, Odontome, Tissus non déminéralisés.

INTRODUCTION

Calcifying odontogenic cyst (COC) is extremely rare, comprising about 1% of all jaw cysts [13]. In a report it was found to represent 0.3% of 10.000 oral biopsies [10], while in another 20 cases of COC were identified out of 59.046 specimens studied in a 18-year period [12]. Wu and Chan [18] found, in a 20 year period, only 2 cases of COC out of 912 tumors and cysts of the jaws. It usually occurs in bone but it can also be located in the soft tissues of the tooth-bearing area [9]: the central lesions are five times more common than the peripheral ones [1]. The majority of the lesions are cystic, although approximately 15% are solid [16]. There is an equal sex distribution and a peak of incidence in the second decade [9, 13]. The lesion has a similar incidence in the maxilla and mandible [9, 13]: the most common site of occurrence is in the anterior part of the jaws. Radiographically the lesion usually appears as a radiolucency with well demarcated borders and a variable quantity of radiopaque material. COCs have characteristic histological features and usually they don't present problems in diagnosis.

In some cases the cyst is associated with the formation of dental hard tissues similar to those seen in complex and compound odontoma [9, 16]. Dental hard tissues are highly mineralized, especially enamel, and so in demineralized sections the most part of enamel is lost and it is not possible to evaluate its morphology or its degree of mineralization.

Aim of our study was a microscopical study, on undemineralized material, of the mineralization of dental hard tissues, calcifications, and ghost cells in a COC associated with a compound odontoma.

CASE REPORT

A 29-year-old male patient was seen in our dental school for the presence since six months of a painless swelling in the right premolar region. A radiographic examination showed a multilocular radiolucency containing some radiopaque masses. Under local anesthesia a mucoperiosteal flap was elevated and the lesion was enucleated.

The specimen was processed to obtain thin ground sections according to the cutting-grinding system [4]. Briefly the specimen was dehydrated in an ascending series of alcohols and embedded in a glycolmethacrylate resin (Technovit 7200 VLC, Kulzer, Wehrheim, Germany). After polymerization the

specimen was sectioned with a diamond saw at a thickness of about 200 μ and ground down to about 30 μ . After polishing, the slides were stained with basic fuchsin-methylene blue or with toluidine blue and observed under normal and polarized light in a Leitz Laborlux microscope (Leitz, Wetzlar, Germany). Von Kossa staining was also done to visualize the calcified structures. A double staining was performed on some slides, first with von Kossa and then with basic fuchsin. After polishing, the slides were then immersed in AgNO₃ for 30 minutes, and exposed to sunlight; the slides were then washed under tap water, dried and immersed in basic fuchsin for five minutes and then washed and mounted.

RESULTS

Microscopically it was possible to observe a cystic lesion with an associated formation of dental hard tissues (Fig. 1). There was an agglomeration of morphologically recognizable teeth of different sizes (Fig. 1). The thickness of enamel varied in the different specimens. In some portions there were Striae of Retzius while the Hunter-Schreger lined were absent. Enamel tufts and spindles were also present, and in some areas there was an enamel lamella, in the outer part of the enamel. The enamel prisms had an irregular orientation. Von Kossa staining revealed that the enamel presented different degrees of mineralization, and the areas of low mineralization were more numerous in the portions in which there was a not complete maturation of the tissues with an incomplete tooth morphodifferentiation (Fig. 2). Dentin presented an homogenous structure with dentinal tubules arranged in a regular way.

Cementum was acellular for the most part. In the cyst wall it was possible to observe ghost cells, with various degrees of nuclear pyknosis and a granular cytoplasm (Fig. 3). Von Kossa showed that most of these cells were calcified (Fig. 4). The lesion was diagnosed as a COC associated with a compound odontoma.

Fig. 2: With an histochemical staining (Von Kossa) it is possible to observe different areas of mineralization.
C=cyst. E=enamel.

Silver nitrate-basic fuchsin-toluidine blue. 6 \times .
Fig. 2: La coloration histochimique de von Kossa permet d'observer les différents territoires de minéralisation. C=kyste. E=émail.
Nitrate d'argent. Bleu de toluidine-fuchsine basique. 6 \times .

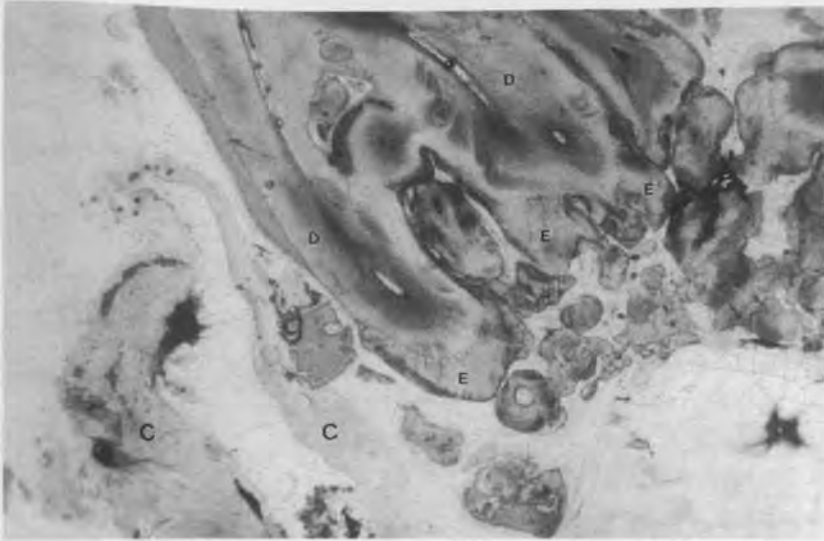


Fig. 1: It is possible to observe in the left corner the lumen of the cyst (C), while in the right side it is apparent the formation of dental hard tissues with a high degree of morpho- and histodifferentiation. E=enamel. D=dentin.

Basic fuchsin-toluidine blue (6×).

Fig. 1: On observe dans le coin inférieur gauche la lumière du kyste (C), tandis que la partie droite de la figure est occupée par la formation de tissus dentaires durs montrant un degré élevé de morpho- et histodifférenciation. E=émail. D=Dentine.

Bleu de toluidine-fuchsine basique. 6×.

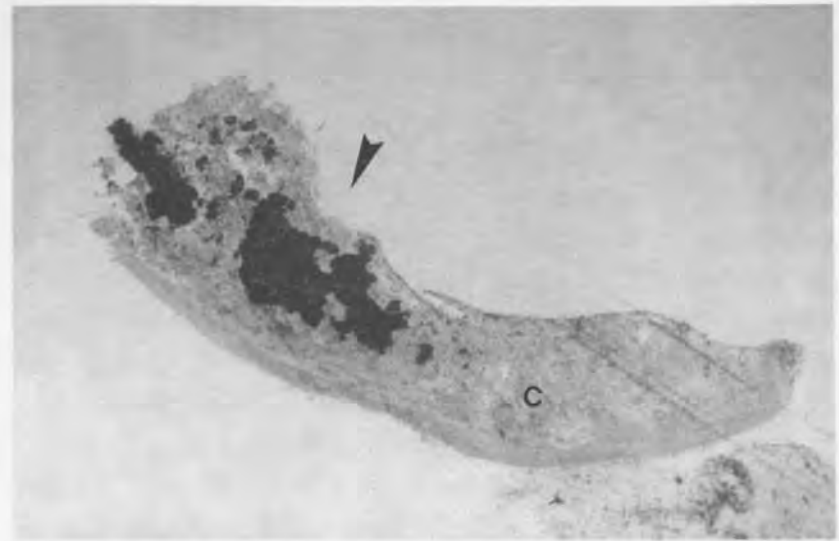
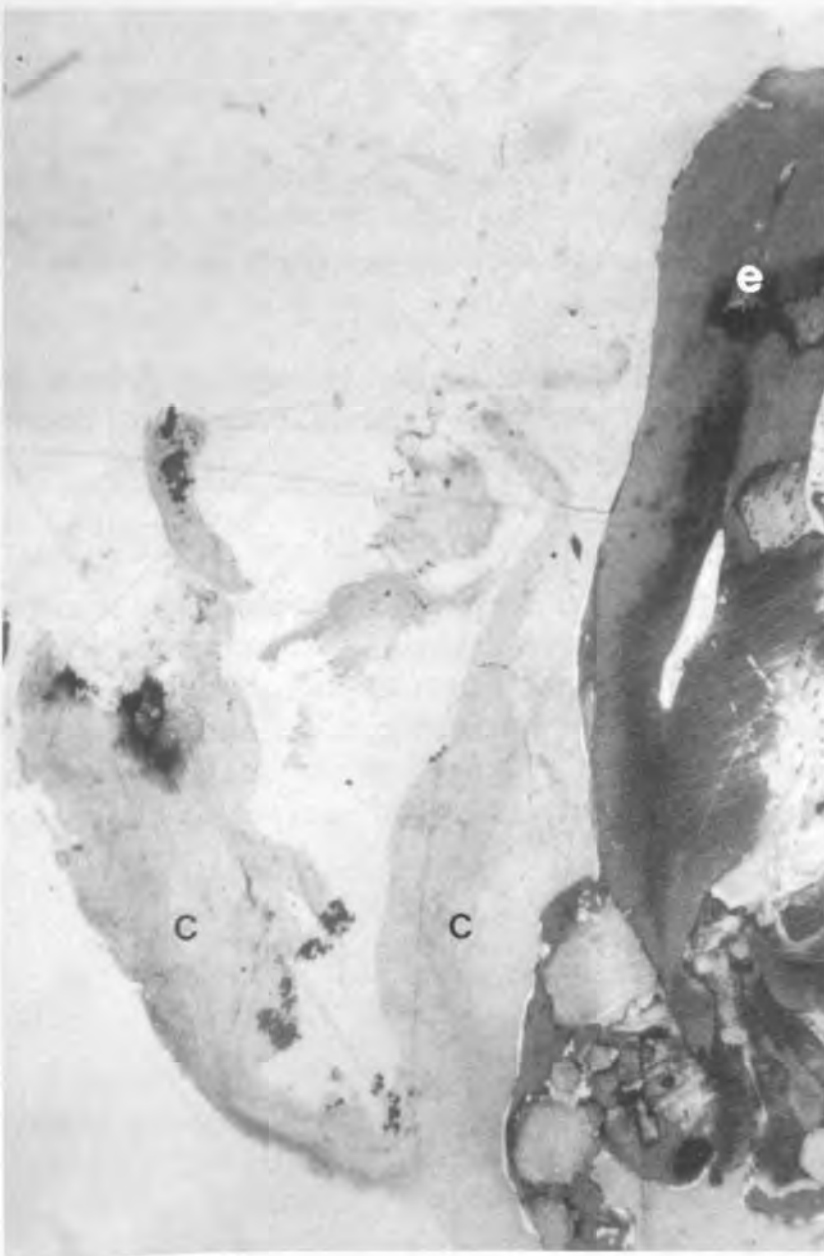


Fig. 3: The histochemical staining (Von Kossa) shows in the cyst wall (C) the presence of calcified structures (arrowhead). Silver nitrate-basic fuchsin-toluidine blue. 40×.

Fig. 3: La coloration histochimique de von Kossa montre la présence de structures calcifiées (tête de flèche) dans le paroi du kyste. Nitrate d'argent. Bleu de toluidine-fuchsine basique. 40×.

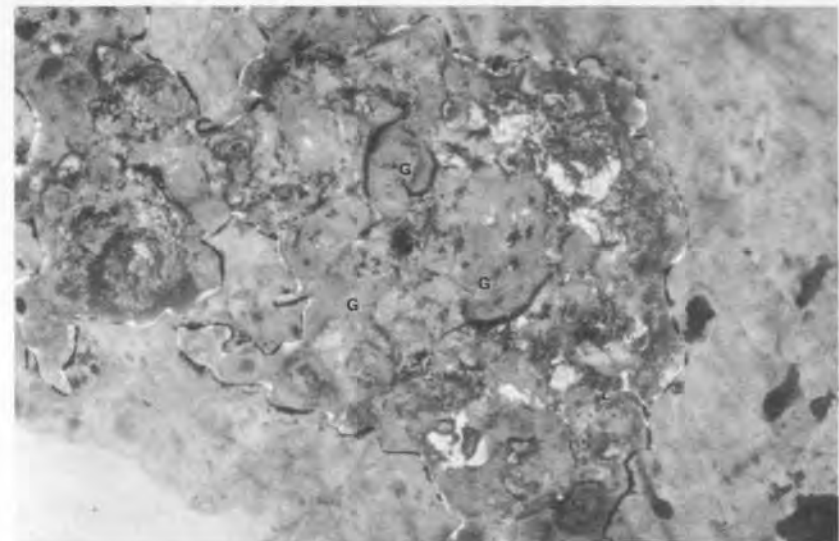


Fig. 4: Higher magnification of the previous field: most of the ghost cells (G) are calcified.

Silver nitrate-basic fuchsin-toluidine blue. 100×.

Fig. 4: Vue à un plus fort grossissement des territoires précédents: la plupart des cellules fantômes (G) sont calcifiées.

Nitrate d'argent. Bleu de toluidine-fuchsine basique. 100×.

DISCUSSION

More than 200 cases of COCs have been reported [2, 3, 15].

The COC can frequently present in association with or can contain areas with a microscopic appearance similar to a variety of tumours (adenomatoid odontogenic tumour, calcifying epithelial odontogenic tumour, ameloblastic fibroma and fibro-odontoma, complex and compound odontoma) [7, 8, 10, 13, 14], due to the ability of the epithelial lining of the COC to induce the formation of dental tissues in the connective tissue wall [2, 3, 13]. Hong *et al.* [6] in a review of 92 cases of COCs found an association

with odontoma in 16 cases, while Buchner *et al.* [2] found presence of an odontoma in 35% of 17 cases of central COC, and Shamaskin *et al.* [12] in 47% of 15 cases of central COCs. Takeda *et al.* [15] found in a histologic study of 13 COCs that 9 were of the odontome producing type and Buchner [3] in a review of 215 cases of central COC found that 26% of the cases were of the odontoma-associated type. Toida *et al.* [16], on the other hand, reported that only about one-tenth of described COCs have occurred in association with an odontoma, and according to them only a few reports of COC found concurrently with true odontoma have been published. The histogenesis of COC is probably related to the odontogenic epithelium [16] but the true nature of the COC, whether it is a cyst or a tumour, is still controversial. The cases of COCs in which are present other features of odontogenic tumours are controversial in that some investigators [16] think that the COC develops secondarily from the odontogenic epithelium that contributed to the formation of the odontoma, while other authors believe that COC arises *de novo* and is not a secondary phenomenon in pre-existing odontogenic tumours or cysts [15]. In some cases COCs have features that suggest a neoplastic rather than a cystic nature (i.e. presence of solid areas, presence of an epithelium with a striking resemblance to ameloblastoma, resorption of adjacent roots or teeth, erosion of cortex and moderate amount of bone destruction [14, 15].

A classification of COCs has been proposed by Preatorius [17]:

Type 1A: simple unicystic type

Type 1B: odontome-producing type

Type 1C: ameloblastomatous proliferating type

Type 2: neoplasm which possesses some of the histological features of COC.

In the latter case it is possible to observe microscopically the presence of an ameloblastomatous epithelium in which the development of cysts is a secondary feature and the term dentinoameloblastoma has been proposed [13]. For this type of neoplasm it has been suggested also the term of «dentinogenic ghost cell tumour» [5], but Ellis and Shmookler [5] prefer the term of «epithelial odontogenic ghost cell tumor (EOGCT)» due to the fact that the ghost-nucleated keratinizing cells are the most distinctive histologic feature.

More recently Hong *et al.* [6] in their review of 92 cases of COCs divided the lesions between cysts (85.9%) and neoplasms (14.1%).

The cysts were classified as:

- 1) nonproliferative COC (simple unicystic structure);
- 2) proliferative COC (multiple daughter cysts, extensive ghost cell formation, marked tendency for calcification);
- 3) ameloblastomatous COC (ameloblastoma-like cyst lining epithelium with ghost cells and calcifications);
- 4) COC associated with odontoma.

The neoplasms were divided in

- 1) ameloblastoma ex COC;
- 2) peripheral epithelial odontogenic ghost cell tumour;
- 3) central epithelial odontogenic ghost cell tumour.

Buchner [2-3] proposed on the other hand the following clinicopathologic classification:

A. Peripheral (extraosseous) COC

1. cystic variant
2. neoplastic (solid) variant

B. Central (intraosseous) COC

1. Cystic variants
 - a. simple
 - b. associated with an odontoma
 - c. associated with odontogenic tumours (other than odontoma)
 - d. other variants (such as clear cell variant, pigmented variant)
2. Neoplastic (solid) variant (known at present as dentinogenic ghost cell tumour or epithelial odontogenic ghost cell tumour)

3. Malignant COC

In conclusion our results in undemineralized sections showed that in our case of COC associated with a compound odontoma the morpho- and histodifferentiation of the dental hard tissues found in the cyst wall had features strikingly similar to those found in undemineralized sections of compound odontoma [11].

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