Coll. Antropol. **28** (2004) 1: 357–362
UCD 616.314-007.64
Original scientific paper

Calcifying Odontogenic Cyst – Gorlin's Cyst – Report of Two Cases

Goran Knežević[1], Klara Sokler[1], Pavel Kobler[2] and Spomenka Manojlović[3]

[1] Department of Oral Surgery, University Hospital »Dubrava», Zagreb, Croatia
[2] Department of Oral Surgery, School of Dental Medicine, University of Zagreb, Zagreb, Croatia
[3] Department of Pathology, School of Medicine, University of Zagreb, Zagreb, Croatia

A B S T R A C T

The authors present two cases of calcifying odontogenic cysts, which were confirmed by histological examination. In the first case the radiographic findings and clinical status did not indicate the presence of a calcifying odontogenic cyst. In the second case, differential diagnosis included COC. The histopathological findings showed that what appeared to be simple cases of bone translucencies was in fact an unusual odontogenic lesion. The two cases point out the possibility of incorrect assessment of deceptively banal cases in daily specialist practice.

Key words: *calcifying odontogenic cyst, Gorlin's cyst, keratinizing epithelial odontogenic cyst, calcifying ghost cell odontogenic tumor, epithelial odontogenic ghost cell tumor*

Introduction

A calcifying odontogenic cyst (COC) is rarely found in everyday practice in oral surgery. It was thoroughly described by Gorlin and coworkers in 1962[1] and in 1963 Gold introduced the term keratinizing and calcifying odontogenic cyst[2]. He believed that this specific type of cyst had not been described previously in the literature as an entity. The World Health Organization (WHO) according to Kramer and Pindborg classification from 1992[3] and the majority of the authors favored the use of the term calcifying odontogenic cyst[3–22] and described it as a cystic or neoplastic-like odontogenic pathological lesion of the jaw and classified it as a benign odontogenic tumor[3].

Calcifying odontogenic cyst represents 2% of all odontogenic pathological changes of the jaws[4], although it can be found together with other odontogenic tumors, most frequently with odontoma in 24% of cases[5].

Received for publication October 22, 2002

It most often occurs as a central (intraosseous) lesion[3,4,6,7,9], whereas peripheral (extraosseous) localization in the soft tissue is rare[8,14], their incidence in the total number is 13–25%[3,7,10,9]. In 65% of cases calcifying odontogenic cyst occurs in the anterior part of the jaw in the incisor and canine region[5,6,7,13]. Central and peripheral forms of calcifying odontogenic cyst occur equally in the upper and lower jaws[4,8,10,12]. Johnson[14] reported the occurrence of 60% in the mandible, 30% in the form of peripheral calcifying odontogenic cysts, while the anterior part of the jaw was involved in 53% of cases. On the basis of 52 examples of calcifying odontogenic cysts connected with an odontoma Hirshberg et al.[15] concluded that the upper jaw was affected in 61.5% and the anterior region of the jaw in 75%.

COC cyst can occur in very young patients, even in the first year of life[17]. Cases have also been recorded of patients in their eighties[5,7]. However, in the majority of cases it occurs in the second decade of life[5,7,17,18].

COC may clinically be diagnosed as calcifying epithelial odontogenic tumor[11], adenomatoid odontogenic tumor, ameloblastic fibroodontoma, complex or compound odontoma, dentigerous cyst or as other types of odontogenic cysts. It is unclear whether it should be regarded as a separate entity or as a stage in development of another odontogenic tumor[11–13,23–25].

The reasons why in the past we did not have the diagnosis of COC were probably in clinical and histological similarity of the lesion with some odontogenic tumors or cysts. The lesion differs histologically from the odontogenic cysts and epithelial tumors as ameloblastoma, but could be similar to the calcifying epithelial odontogenic tumor that presents more aggressive growth. Presentation of following two cases diagnosed in last two years could be our contribution for better understanding of this lesion in our clinical practice.

Results

Case 1

A 49-year-old man was referred to the Department of Oral Surgery for a swelling in the vestibulum of the left side of the mandible. No visible facial alterations could be seen, but a mass of the outer wall of the mandible was detected by palpation.

Intraorally a sharply circumscribed swelling could be seen on the left side of the mandible, 6.8 × 2.5 cm. The swelling was covered with unchanged mucosa.

The lesion stretched from the left lateral incisor to the lower third molar on the same side of the jaw. Both premolars and the first and second molar of the jaw were missing. The loosening of the cortical bone could be felt by palpation. The jaw appeared distended. The left canine, the root of which protruded into the cyst, did not react to electrical stimulus, although the tooth was not movable. No paresthesia of the lower lip was present.

The radiographic findings showed sharply circumscribed bilocular translucency along the lower edge of the mandible. The root of the left canine was not resorbed (Figure 1).

Differential clinical diagnosis included an odontogenic residual cyst, odontogenic keratocyst, monocystic ameloblastoma[12] or some other unusual soft odontogenic tumor[3,7,12,21,26,27].

Because of the size of the cyst the operation was performed under general anesthesia. The cystic lesion was enucleated and a permanent postoperative drain by plastic tube was used[28].

The material was a hollow formation 4.2 cm in diameter with brown content in the lumen. Histologically the wall was composed of fibrous tissue, and the inter-

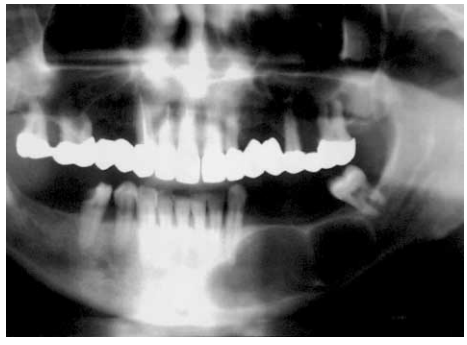

Fig. 1. Calcifying odontogenic cyst – case 1. Orthopantomogram shows bilocular translucency in the left body of the mandible.

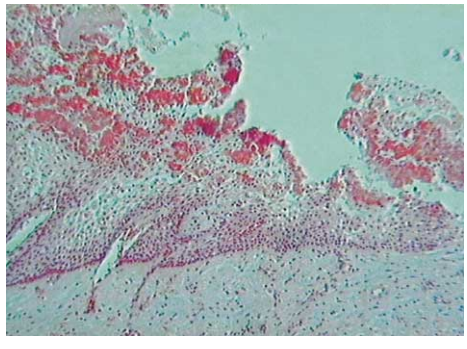

Fig. 2. Calcifying odontogenic cyst – case 1. The cystic formation, covered by normal epithelium, the basal layer of which is composed of tiny cubic cells with dark nuclei, and surface layers of large, bright, loosely distributed cells, similar to a star-like reticulum. On the surface are accumulations of large eosinophilic masses containing keratinized »shadow« cells (HE × 20).

nal surface covered in places by a thin layer or in others by a thick layer of epithelial cells. There were hilic, and in places calcified areas, and ghost cells[26]. Tiny islets of odontogenic epithelia could be seen in the connective stroma (Figure 2).

Case 2

A 21 year-old girl was referred to the Department of Oral Surgery after an X-ray examination showing a cyst of the upper jaw.

Clinically, the deformations could be seen on the vestibular wall of the upper jaw in the region of the right impacted canine extending to the right first molar. The patient was diabetic. Sharply circumscribed translucency could be seen on the X-ray bite image, which stretched from the region of the right impacted canine (the deciduous canine was also pres-

Fig. 3. Calcifying odontogenic cyst – case 2. The bite image of the maxilla shows translucency in the bone at vestibular swelling with a thinned cortical bone.

Fig. 4. Calcifying odontogenic cyst – case 2. Translucency visible on the intraoral image in the premolar and canine area. The shadow of the crown of the impacted canine and shadow of calcified dental tissue under the crown of the canine can be seen on the edge of the translucency.

ent) up to the region of the first right molar (Figures 3 and 4).

No fluctuation of a cystic content was noticed. Loosening of the cortical buccal wall was observed as well as swelling of the palate.

Differential clinical diagnosis included follicular cyst and odontoma of the upper jaw[12], with the possibility of one of the odontogenic tumors in which calcified dental tissue occurs[11,12,26].

The operation was performed under general anaesthesia by enucleation of the lesion, in agreement with the principle of clinical method for treating small cystic lesions of jaws[28].

The material was an oval irregularly developed firm mass, resembling a tooth, and a long hollow formation 2.5 cm in diameter. Histologically the lesion was consisted of cellular fibrous tissue covered by odontogenic epithelium which in places strands composed of ghost cells and foci of calcification. In places islets of odontogenic epithelium could be seen in the connective stroma (Figures 5 and 6).

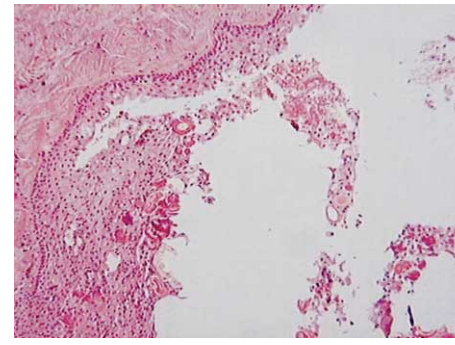

Fig. 5. Calcifying odontogenic cyst – case 2. The lumen of the cyst is covered by cubical and cylindrical odontogenic epithelium with accumulations of keratinized and calcified large eosinophilic cells. (HE ×100).

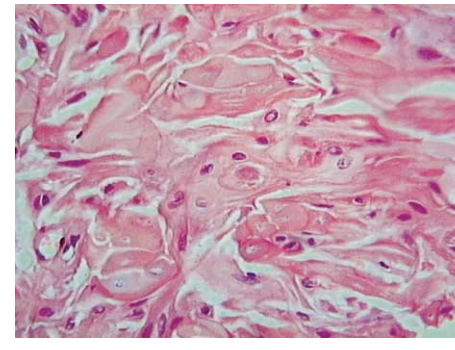

Fig. 6. Calcifying odontogenic cyst – case 2. Eosinophilic masses composed of accumulations of keratin and rests of ghost cells (HE ×250).

Discussion

We have described here first two cases of calcifying odontogenic cysts found at clinical material of the Department of Oral Surgery.

In the first case, although the clinical image indicated an odontogenic residual cyst, odontogenic keratocyst or monocystic ameloblastoma, a calcifying odontogenic cyst was found. In the second case the possibility of a calcifying odontogenic cyst was suspected and histopathologically confirmed. The specificity of these two cases is that the clinical diagnosis shown in the radiographs appeared to be relatively simple and clear, particularly in the first case. However, the histopathological diagnosis proved to be a rare odontogenic lesion.

The presented cases are examples of two types of calcifying odontogenic cyst (monocystic and monocystic odontoma creative).

In 1981 Praetorius and coworkers[6] tried to classify calcifying odontogenic cyst by dividing it into two entities: a cyst and a neoplasm. The cystic entity was classified into three types. Type 1: a simple monocystic type of typical Gorlin's cyst, with or without dentinoid calcified tissue, Type 2: monocystic odontoma creative type,

with all the characteristics of the previous type, except that the hard tissue was complex or compound odontoma, and a presence of ameloblastic fibroma tissue in the cystic wall extending into the surrounding tissue, Type 3: monocystic ameloblastomatous proliferating type which was marked by ameloblastomatous proliferation both in the walls and in the lumen, and hard dental tissue which consisted of dentinoid formation in connection with islets of epithelia in the connective wall. The neoplastic form was described as an odontogenic tumor with so-called shadow-like cells »ghost cells«. The epithelial elements consisted of numerous ameloblastomatous proliferations of tissue in the connective tissue of the stroma. Within the epithelial islets were present different amounts of ghost cells, and the hard tissue was composed of different amounts of dentinoid in direct contact with the epithelium[6,12].

According to Pretorius[6] our first case could be classified as Type 1 and the second as Type 2.

In 1992 Bucher[5] described a multicystic form of calcifying odontogenic cyst as a separate entity, and on the basis of an analysis of 215 clinical and histological features of central calcified odontogenic cysts[5]. Instead of calcifying odontogenic cyst Langlais and coworkers suggested the term calcifying odontogenic lesion including cystic and tumorus form as separate forms, and a combined lesion when elements of both forms are observed[12].

Calcifying odontogenic cyst is most frequently radiographically seen as a unilocular translucency[5,17,19] with sharply circumscribed edges[7,17] occurring in the form of a multilocular lesion in a very small number of cases, from 5% to13%[18].

A definite diagnosis of calcifying odontogenic cyst can be reliably made on the basis of a histological examination.

With regard to the very small number of recurrences, only eight cases of recurrences have been documented in English literature[1,22,23,24].

Treatment of calcifying odontogenic cyst is usually enucleation of the cyst. In his most recent publication on pathology Barnes mentions the possibility of malignant transformation, and in differential diagnosis points out that COC must be differentiated from calcifying odontogenic tumor and ameloblastoma, that are essentially more aggressive and require an extensive surgical approach[26].

The diagnosis of pathological alterations of odontogenic etiology appears very simple. However, only to those with a superficial knowledge of the matter. The further one considers the problem of odontogenic tumors, so an excellent knowledge of the structure is necessary for correct diagnosis, both on the part of the clinician and the pathologist, who need to have subspecialist training in this field. An optimal solution would be the existence of a clinical pathologist, who would arrive at the final diagnosis in cooperation with an oral or maxillofacial surgeon.

COC may mimic numerous odontogenic or not lesions therefore the clinical and histological diagnosis is difficult. These two cases demonstrate in addition that a specific knowledge in oral histopathology is required to differentiate odontogenic lesions.

REFERENCES

1. GORLIN, R. J., J. J. PINDBORG, F. P. CLAUSEN, R. A. VICKERS, Oral Surg. Oral Med. Oral Pathol., 15 (1962) 1235. — 2. GOLD, L., Oral Surg. Oral Med. Oral Pathol., 16 (1963) 1414. — 3. KRAMER, I. R. H, J. J. PINDBORG, M. SHEAR: Histological typing of odontogenic tumors. (Springer Verlag, Berlin 1992). — 4. ALTINI, M., A. G. FARMAN, Oral Surg. Oral Med. Oral Pathol., 40 (1975) 751. — 5. BUCHER, A., J. Oral Maxillofac. Surg., 49 (1991) 330. — 6. PRAETORIUS, F., E. HJORTING-HANSEN, R. J. GORLIN, R. A. VICKERS, Acta Odontol. Scand., 39 (1981) 227. — 7. FREEDMAN, P. D., H. LUMERMM, J. K. GEE, Oral Surg. Oral Med. Oral Pathol., 40 (1975) 93. — 8. SWAN, R. H., G. D. HOUSTON, S. P. MOORE, J. Periodontol., 56 (1975) 340. — 9. ERASMUS, J. H., I. O. C. THOMPSON, L. J. VAN RESENBERG, A. J. VAN DER WESTHUIJZEN, Dentomaxillofac. Radiol., 27 (1998) 30. — 10. NEVILLE, B. W., D. D. DAMM, C. M. ALLEN, J. E. BOUQUOT: Oral and maxillofacial pathology. (W. B. Saunders, Philadelphia, 1995). — 11. PINDBORG, J. J., Cancer, 11 (1958) 838. — 12. LANGLAIS, R. P., O. E. LANGLAND, C. J. NORTJE: Diagnostic imaging of the Jaw. (Williams & Wilkens: Malvern, 1995). — 13. HONG, S. P., G. L. ELLIS, K. S. HARTMAN, Oral Surg. Oral Med. Oral Pathol., 72 (1991) 56. — 14. BUCHER, A., P. W. MERRELL, L. S. HANSEN, A. S. LEIDER, Oral Surg. Oral Med. Oral Pathol., 72 (1991) 65. — 15. JOHNSON, A., M. FLETCHER, L. GOLD, S.–Y. CHEN, J. Oral Maxillofac. Surg., 55 (1997) 679. — 16. HIRSHBERG, A., L. KAPLAN, A. BUCHER, J. Oral Maxiflofac. Surg., 52 (1994) 555. — 17. LELLO, G. E., M. MAKEK, Int. J. Oral Maxillofac. Surg., 15 (1986) 637. — 18. NAGAO, T., T. NAKAJIMA, M. FUKUSHIMA, T. ISHIKI, J. Maxillofac. Surg, 11 (1983) 174. — 19. TANIMOTO, K., S. TOMITA, M. AOYAMA, Y. FURUKI, M. FUJITA, T. WADA, Int. J. Oral Maxilofac. Surg., 17 (1988) 29. — 20. DOMINGUEZ, F. V., E. G. ESPINAL, Acta Odontol. Latinoam., 1 (1984) 77. — 21. McGOWAN, R. H., R. M. BROWNE, Br J. Oral Surg., 20 (1982) 203. — 22. SLOOTWEG, P. J., R. KOOLE, J. Maxillofac. Surg., 8 (1980) 143. — 23. STOELINGA, P. J., F. B. BRONKHORST, J. Craniomaxfac. Surg., 16 (1988) 184. — 24. SWIMSON, T. W., Br J. Oral Surg., 13 (1976) 217. — 25. HOFFMAN, S., J. R. JACOWAY, S. O. KROGS: Intraosseous and parosteal tumors of the jaws. (ArMed. Forces Institute of Pathology, Washington, 1987). — 26. BARNES, L.: Surgical pathology of the head and neck. (Mareel Dekker Inc, New York-Basel, 2001). — 27. RUSHTON, E., K. HORNER, Br. J. Oral Maxillofac. Surg., 35 (1997) 196. — 28. GRUREVIĆ, J., G. KNEŽEVIĆ: Alternative u liječenju velikih cista donje čeljusti. In: Proceedings. (9. kongres Udruženja stomatologa Jugoslavije, Ljubljana, 1988).

G. Knežević

Department of Oral Surgery, University Hospital »Dubrava«, Av. G. Šuška 6, 10040 Zagreb, Croatia
e-mail: knezevic@sfzg.hr

KALCIFICIRAJUĆA ODONTOGENA CISTA – GORLINOVA CISTA – PRIKAZ DVAJU SLUČAJEVA

SAŽETAK

Autori prikazuju dva slučaja kalcificirajućih odontogenih cista koje su potvrđene histološkom raščlambom. U prvom slučaju rentgenski nalaz i klinička slika nisu nagovještavali postojanje kalcificirajuće odontogene ciste. U drugom slučaju diferencijalna dijagnoza uključivala je i mogućnost kalcificirajuće odontogene ciste. Histopatološki nalazi pokazali su, da su ono što se činilo da su bili jednostavni primjeri koštanih prosvjetljenja zapravo bile neobične odontogene patološke promjene. Prikazana dva slučaja ističu mogućnost netočne procjene prividno banalnih slučajeva u svakodnevnoj specijalističkoj praksi.