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Calcifying Cystic Odontogenic Tumor (CCOT-Gorlin tumor) of the alveolar part of the mandible - a case report

Adrian Gnatek Dr, Piotr Osica PhD, Anna Janas-Naze PhD Assoc. Prof.

Adrian Gnatek ORCID ID:0000-0002-1702-5162 email:gantekadrian@gmail.com Piotr Osica ORCID ID:0000-0002-6436-5442 Anna Janas-Naze ORCID ID:0000-0001-6885-4457

Department of the Oral Surgery, Central Clinical Hospital, Medical University of Lodz Head of the Department: Anna Janas-Naze, PhD, Assoc. Prof.

Corresponding author: Adrian Gnatek Department of Oral Surgery, Medical University of Lodz 92-213 Łódź, ul. Pomorska 251, tel. 42 675 75 29 The article was financed by Medical University of Lodz as a part or statutory activity nr 503/2-163-01/503/01

Abstract

Odontogenic tumor – like lesions are rarely occurring pathological processes in jawbones. The article presents a case of a 39-year-old-patient that had been treated surgically because of Gorlin tumor of alveolar part of mandible. Clinical examination showed alveolar ectasia in the region of the tumor in mandible. Due to patient's mental impairment taking any X-rays and surgery photographic documentation was impossible.

Key words: Calcifying Cystic Odontogenic Tumor; CCOT; Gorlin tumor; tumor-like lesions; surgery; analgosedation

First reports related to Calcifying Odontogenic Cyst (COC) are dated 1962. The newest tumors classification established by WHO in 2005, described the lesion as Calcifying Cystic Odontogenic Tumor (CCOT). Due to its clinicopathologic features, CCOT was classified as ghost cell tumors group, which involves: earlier mentioned CCOT, DGCT – dentigerous ghost cell tumor and GCOC – ghost cell odontogenic carcinoma[1].

Because of morphological and histological complexity of the tumor, Gorlin's classification published in 1962, already described CCOT as a lesion associated with dental apparatus[2].

Dentigerous tumors are pathological lesions deriving from the remnants of developing dental bud components. CCOT or COC are benign neoplasm lesions. Presence of ghost cells and amelobastoma-like epithelium are characteristic features of the tumor. CCOT occurs in two clinical forms: peripheral and intraosseus. Calcifications inside ghost cells of the tumor might be observed. The most common localization of CCOT is the anterior segment of maxilla and mandible[3].

Occurrence of pathological lesions such as Calcifying Cystic Odontogenis Tumor (CCOT) is relatively rare. It represents about 1,6 to 4,3% of all odontogenic tumors. Nomenclature related to CCOT varies. CCOT is described as: calcifying odontogenic cyst, Gorlin cyst, calcifying ghost cell odontogenic tumor, keratinizing and calcifying odontogenic cyst, dentinogenic ghost cell tumor, epithelial odontogenic ghost cell tumor[4].

Sometimes clinical and radiological features of the tumor might imitate dentigerous cyst. Namely, its histological and clinical development inside oral cavity tissues reveals features of the tumor[5].

Despite the fact, that CCOT occurs in anterior segment of jaws, its presentations in posterior parts of maxilla involving maxillary sinus is also known[6].

Case report

39-year-old patient was referred to the Oral Surgery Department for the consultation and treatment of the mandible body tumor in the region of missing tooth 33.

Patient's medical history revealed deep congenital mental impairment, no drug allergies. no significant surgery procedures in the past and no drug treatment.

In extra oral examination no abnormalities were found.

Intra oral examination showed numerous missing teeth and an alveolar ectasia in the region of the mandible tumor. After a deep analysis of the patient's medical history, clinical examination and an assessment of possible complications, surgery was planned – resection of the mandible body tumor in the region of the missing tooth 33.

The surgery operation plan along with possible complications was presented to the patient and his guardian. All the surgery permissions were collected. Pre-operative medications and laboratory blood tests were recommended and the surgery scheduled.

The patient and his guardian were informed about the details of postoperative recommendations and obligatory follow-up visits.

In general anesthesia the tumor of mandible body in the region of the missing tooth 33 was excised. Pathological lesion was excised with clinical margins. Excised tissues were sent to histopathological examination.

Intra- and postoperative process were uneventful. Patient in a general good condition was discharged from the clinic in guardian's care.

Control examination was performed the day after surgery. Proper healing of the wound was observed. The follow-up and sutures removal were recommended after one week.

Histopathological examination described the tumor as a calcifying cystic odontogenic tumor (CCOT) – Gorlin tumor. Currently the patient stays under Oral Surgeon's care.

Discussion

Calcifying cystic odontogenic tumor (CCOT) is an uncommon lesion. It accounts for about 2% of all odontogenic tumors. It was earlier referred to as Calcifying Odontogenic Cyst. Renamed and reclassified in 2005, after the new WHO tumors classification was introduced. It may occur simultaneously with odontomas, ameloblastomas and ameloblastic fibromas. CCOT exists in two forms: intra- and extraosseus. Most frequently it involves anterior part of jaws. In some cases CCOT occurs bilaterally[7].

CCOT is a benign odontogenic cystic lesion. The presence of ameloblastoma-like epithelium, calcification and ghost cells being its characteristic feature. Most often the discovery of the lesion, particularly its intraosseus form, is observed on routinely taken OPGs.

Odontogenic ghost cell tumors represent large group of pathological odontogenic lesions. They occur in different forms – from the cysts up to solid neoplasms. The character of these lesions also varies – from benign variants to locally aggressive and metastatic forms. The common treatment method of these sorts of lesions is surgical excision. In some cases bone resection is performed[8].

During the diagnostic process it is significant to draw attention to coexisting CCOT and other odontogenic tumors.

Simultaneous occurrence of CCOT and other lesions is rare. However, co-occurrence of CCOT, odontoma, supernumerary tooth and follicular cyst in the maxilla was observed[9].

CCOT is rare benign cystic odontogenic tumor. The choice of treatment methods always depends on the localization and histological type of the tumor. In particular cases to avoid trauma and tooth loss in young patients, marsupialization of the tumor is performed[10].

Surgical treatment of CCOT absolutely has to be associated with total excision of the lesion. Then, follow-ups ought to be planned. Despite the fact that Ghost Cell Odontogenic Carcinoma is a rare neoplasm tumor with aggressive clinical process, malignant transformation of CCOT into GCOC was described[11].

In the presented case the tumor was excised completely. Histopathological examination revealed Calcifying Cystic Odontogenic Tumor, which is a relatively rare lesion in the region of oral cavity and maxillofacial region. Due to their infrequent occurrence, radical excision of the tumor and thoroughly performed follow-ups are key factors in CCOT treatment.

Patients with oral cavity lesions are still an increasing group of surgical patients. All odontogenic pathologies ought to be taken into consideration. Each variant of the same tumor requires individual treatment plan. It seems that an accurate histological examination and diagnosis are priceless and should be always related to clinical symptoms. Knowledge associated with odontogenic tumors ought to be deepened. Each odontogenic tumor may be treated as a challenge for oral surgeons in surgery proceedings.

Conclusions

Patients after excision of benign tumor-like lesions such as CCOT should be observed periodically. Clinical and radiological phases of bone remodeling ought to be assessed.

Accurate histopathology examination and diagnosis are key factors for further proceeding assessment and planning.

REFERENCES:

- C. Ledesma-Montes *et al.*, "International collaborative study on ghost cell odontogenic tumours: Calcifying cystic odontogenic tumour, dentinogenic ghost cell tumour and ghost cell odontogenic carcinoma," *J. Oral Pathol. Med.*, vol. 37, no. 5, pp. 302–308, 2008.
- [2] S. Kler, S. Palaskar, V. P. Shetty, and A. Bhushan, "Intraosseous calcifying cystic odontogenic tumor.," *J. Oral Maxillofac. Pathol.*, vol. 13, no. 1, pp. 27–9, 2009.
- [3] A. P. de Lima, D. Kitakawa, J. D. Almeida, A. A. H. Brandão, and A. L. Anbinder,
 "Peripheral calcifying cystic odontogenic tumour of the maxillary gingiva.," *BMC Res. Notes*, vol. 5, p. 455, 2012.
- [4] A. Hanttash, A. Hanttash, A. T. Oncul, and U. A. Aktaş, "Calcifying Cystic Odontogenic Tumor Presenting as a Dentigerous Cyst: A Case Report with Literature Review," *Int. J. Oral Maxillofac. Pathol. Int. J. Oral Maxillofac. Pathol.*, vol. 66, no. 20151, pp. 11–1811, 2015.
- [5] C. Radheshyam, B. Alokenath, H. Kumar, and P. Abikshyeet, "Calcifying cystic odontogenic tumor associated with an odontome - A diverse lesion encountered," *Clin. Cosmet. Investig. Dent.*, vol. 7, pp. 91–95, 2015.
- [6] J. Chindasombatjaroen, S. Poomsawat, and K. Boonsiriseth, "Two unique cases of calcifying cystic odontogenic tumor in the maxillary posterior region," *Oral Surg. Oral Med. Oral Pathol. Oral Radiol.*, vol. 118, no. 4, pp. 497–504, 2014.
- [7] P. Khandelwal, A. Aditya, and A. Mhapuskar, "Bilateral calcifying cystic odontogenic tumour of mandible: A rare case report and review of literature," *J. Clin. Diagnostic Res.*, vol. 9, no. 11, p. ZD20-ZD22, 2015.
- [8] J. Sheikh, M. D. Cohen, N. Ramer, and A. Payami, "Ghost Cell Tumors," J. Oral Maxillofac. Surg., vol. 75, no. 4, pp. 750–758, 2017.
- [9] J. Chindasombatjaroen, S. Poomsawat, and B. Klongnoi, "Calcifying cystic odontogenic tumor associated with other lesions: Case report with cone-beam computed tomography findings," *Oral Surg. Oral Med. Oral Pathol. Oral Radiol.*, vol. 113, no. 3, pp. 414–420, 2012.
- [10] S. V.T., F. C.E., M. A.B., P. A.A., H. J.A., and D. J.A., "Conservative surgical treatment of an

aggressive calcifying cystic odontogenic maxillary tumor in the young permanent dentition," *Pediatric dentistry*, vol. 33, no. 3. pp. 261–265, 2011.

Z. Y. Zhu *et al.*, "Ghost cell odontogenic carcinoma arising from calcifying cystic odontogenic tumor: A case report," *Korean J. Pathol.*, vol. 46, no. 5, pp. 478–482, 2012.