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Case report

The Importance of Thorough Preoperative Diagnostics of Maxillary Ameloblastoma: Report of Three Cases

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A B S T R A C T

Ameloblastoma, especially maxillary, is a rare benign neoplasm of odontogenic origin. Diagnosis of significant number of lesions is usually established postoperatively, because ameloblastoma, especially the unicystic form, mimics wide range of more frequent jaw lesions. From January 1993 to December 2005, three cases of the maxillary ameloblastoma were surgically treated at our Department. The authors present clinical, radiological and pathohistological features of the ameloblastomas in this rare localization with special attention to need of accurate preoperative diagnostics.

Key words: *ameloblastoma, maxilla, odontogenic tumors, diagnosis*

Introduction

Intraosseous amelobastoma is the most common aggressive odontogenic tumor, but still comparatively rare, comprising only 1% of tumors and cysts arising in the jaws[1]. They are divided into three types: intraosseous solid or multicystic, intraosseous unicystic and peripheral extraosseous form. Furthermore, upon the pathohistological characteristics, solid ameloblastomas are subdivided into plexiform, acanthomatous, follicular, granular cell, desmoplastic and basoloid subgroups[2,3]. Eighty *per* cent of ameloblastomas arise in the mandible, especially in their posterior parts, while approximately 20% of the lesions arise in the maxilla[4]. Half of maxillary ameloblastomas occur in molar area, involving the maxillary sinus in 15% of cases[4,5]. Contrary to their mandibular counterparts, maxillary ameloblastomas erode thin compact bone much faster, invading the maxillary sinus, nasal cavity and neighboring structures; orbit, infratemporal fossa, pterygopalatine fossa, buccal soft tissues etc. Due to above mentioned reasons maxillary ameloblastomas require surgical treatment as radical as cancers. If not, the recurrences occur in 90–100% of cases and can be fatal[2,6]. Proper surgical treatment should include at least 10–15 mm of healthy bone[7]. Due to its rare occurrence and clinically unspecificity, the preoperative diagnosis of maxillary ameloblastoma represents a leading problem. In this article we present own diagnostic errors and difficulties and try to give advices for its avoidance.

Report of Cases

From January 1993 to December 2005, three patients with maxillary ameloblastoma have been treated at our Department. In all three, the diagnosis was established postoperatively. Also, in all, only routine laboratory tests and conventional plain radiographs were performed.

Case 1

Twenty six year old female patient was admitted at our Department in December 1995 due to firm, slowly growing and painless tumor in the left upper oral vestibule. Duration of the symptoms was five years. Intraoral inspection and palpation revealed firm, well circumscri-

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bed, submucous lesion, 40mm in diameter, arising in the region of canine and first premolar. The over lining mucosa was normal. Although the tumorous appearance was evident, due to poor dental status, the odontogenic cyst was also taken in consideration. Radiographic image revealed very unclear radiolucency in above mentioned area (because of that reason the radiogram is not presented). Intraoperativelly, ameloblastous mixed appearance (solid and cystic) was found. Complete resection of the tumor, including safe bone margins was done. The operation was diagnostic and therapeutic in the same time. Pathohistological report described ameloblastoma of typical follicular growth pattern. Eighty eight months after the initial treatment the patient was without of any clinical or radiographical signs of recurrence.

Case 2

A female patient, 69 year old, was referred in June 2001 due to swelling of the right upper oral vestibule which has been lasted six months, without any subjective complaints. Clinically, the tumor was palpable in the region of canine fossa, as well as in a whole right upper oral vestibule, including a region of maxillary tuberosity. Panoramic view revealed an oval tumor of inhomogeneous density in above mentioned region (Figure 1). Tumor was approached intraoraly and partial maxillectomy was done. Pathohystologically, the ameloblastous appearance with areas of a stromal sqamous metaplasia was found. Differential diagnostically, the adamantinous type of a craniopharyngeoma has been also taken in consideration. To date, the patient is clinically and radiographically free of disease.

Case 3

Fifty seven year old male patient was referred in December 2002. due to surgical management of clinically and radiographically typical cystic formation extending to the left maxillary sinus, present at least 1, 5 years (Figure 2). Clinically, the lack of the maxillary bone in the area of the upper left wisdom tooth was obvious. Also, the cystic formation was palpable submucously in premolar and molar area. The preoperative diagnoses were odontogenic cyst extending to maxillary sinus, mucocelle or retentional maxillary sinus cyst. Caldwell-Luc procedure and cyst evacuation was performed. Pathohistological report revealed that, in fact, the whole lesion was multicystic, and the basal cell growth pattern of ameloblastoma was found. To date no recurrence, clinically or radiologically, was noted during follow up.

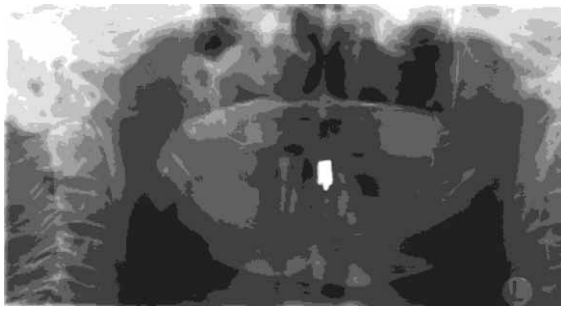

Fig. 1. Oval osseous tumor in the region of right maxillary tuber showing tipical soap bubble radiographic appearance.

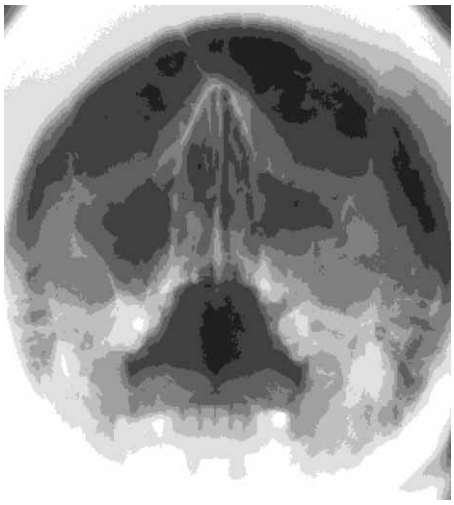

Fig. 2. Unicystic radiographic appearance of the ameloblastoma, extending to the maxillary sinus and destroying the anterior and lateral sinus walls which mimic radiographic appearance of the mucocelle.

Discussion

Solid/multicystic form of intraosseous ameloblastoma is the most frequent and comprises 86% of all ameloblastomas, while unicystic lessions occur in 13%. The rest of 1% of ameloblastomas are peripheral ameloblastomas[3].

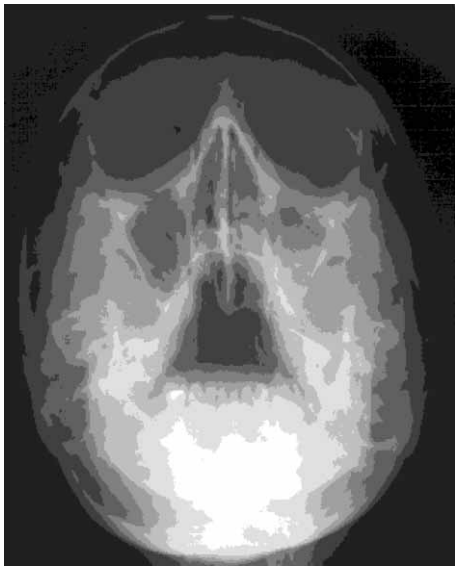

Fig. 3. Radilogical appearance of pathohistologically confirmed retentional cyst of the maxillary antrum followed with bony walls destruction, radiographically indistinguishable from radiographic appearance of Case 3 (Figure 2).

Records addressing the problem of maxillary ameloblastoma are incoherent. Most reports are in forms of case report studies[8,9], some authors present their maxillary ameloblastomas in overall series including mandibular counterparts also[6], many articles are focused to some exclusive clinical or pathohistological variant etc.[4,10,11]. There are very few reports about maxillary ameloblastoma in whom the larger number of cases are presented[12]. Nastri et al. report of scarcely 250 cases evaluated in the world literature until 1993, while Reichart et al. report of 454 cases published until 1995[5,12]. Such lack of accurate data is the main reason why the true nature of this tumor is still unclear. Because the same denominator in most instances is a lack of preoperative diagnosis, it is obvious that accurate preoperative diagnosis of ameloblastoma is a top priority in the treatment plan. Larger lesion with significant bone destruction and extension to neighboring structures generally are easier to be diagnosed. Due to low suspicion index, the problem arises when we are facing small lesion, or lesion mimicking odontogenic cyst or cyst of maxillary sinus. For an example, Figure 3 shows pathologically proved cyst of the maxillary sinus in other patient, which is radiographically indistinguishable from radiographic appearance of Case 3 (Figure 2). This problem of diagnostics is reported by several authors[2,13]. Also, ameloblastomas are not of specific structure clinically, nor radiographically. Solid ameloblastomas are not necessary of radiologically multilocular or soap bubble structure, as well as the unicystic ameloblastomas are not always of unilocular appearance. Reichart et al. found 15% of radiologically unilocular appearances in 1234 ameloblastomas of the maxilla and mandible. Among them, only 102 lesions have been verified as unicystic forms pathohistologically[5]. Similarly, Li et al., in 20% of the pathohistologically confirmed unicystic ameloblastomas found radiological multilocularity[13]. Consequently, the misdiagnosis of the maxillary ameloblastoma is not unusual and leads, due to insufficient safe bone margins in surgical specimen, to unfavorable outcome. In our series, all three cases have been either misdiagnosed (Case 3), under diagnosed (Case 1) or unrecognized (Case 2). Thus, our series represents practically the whole spectrum of possible diagnostic errors which all finally resulted with absence of further diagnostic efforts: CT/MR scanning and preoperative biopsy. The reasons for that were too much everyday practice routine behavior in the case of anteriorely located, well circumscribed, evidently benign tumor, without extension to maxillary antrum (Case 1); typical misdiagnosing of the cystic formation of the maxillary antrum which resembled large odontogenic cyst, mucocelle or retentional maxillary sinus cyst (Case 3); and even ignoration and ununderstanding of evident clinical and radiographic findings of ameloblastoma in a typical localization (Case 2). Accurate CT delineation of ameloblastoma extent is of paramount importance, indicating adequate surgical approach which is a key of successful treatment, since recurrences mostly occur in unrecognized and underdiagnosed patients[2]. Li et al., reviewing the clinicopathological features in thirty three patients with unicystic maxillary ameloblastoma, found six recurrences and all six were misdiagnosed as odontogenic cysts preoperatively[13]. Ninety five *per* cent of recurrences are detected within 5 years of initial treatment[14], as well as half of recurrences are usually find within 1 year postoperatively[15]. Although all our patients are without any signs of disease, it does not mean to be cured due to possibility of late recurrences, ten or more years after the initial treatment[12]. Generally speaking, there is no place for enucleation in treatment of amelobastoma. The only exception of this rule is peripheral amelobastoma, a distinct type of tumor arising in gingiva, predominantly in the mandible. Peripheral ameloblastoma is thought to arise from either extraosseous remnants of dental lamina or the basal cell layer of surface epithelium[16]. Peripheral ameloblastoma is less common in maxilla, where the posterior tuberosity is the most common site of occurrence. Extremely rare it could be present in other sites of oral mucosa, except the gingiva[17,18]. Limited number of the reports regarding peripheral amelobastoma treatment proved the local excision, extraosseous equivalent of enucleation, as a sufficient treatment[19]. Other two types of ameloblastoma; solid and unicystic, require more aggressive resection, which usually includes 10–15 mm of healthy bone[7]. In the case of the mandible it usually means segmental bone resection, while in the case of the maxilla partial of even total maxillectomy should be performed. Otherwise, the reccurrence rate exceed 60–80 %[20]. This is especially true for maxillary amelobastomas where reccurrences exceed 90–100 % if not treated radically[2,6]. The reason for such aggressive surgical attitude lies in the fact that ameloblastic cells are usually find few millimeters beyond the radiographic and clinical margins[21]. Indeed, there is no reports in the recent literature regarding to the enucleation or curettage in treatment of solid or multicystic ameloblastoma[20]. The treatment of unicystic lesions is more controversial. Enucleation of such ameloblastoma could results with complete resection and free margins. One should always keep in mind that there are three pathohistological subtypes of unicystic ameloblastoma, depending on whether they have simple odontogenic cystic lining, a cystic lining which shows intraluminal plexiform proliferation, or a cystic lesion with invasion of supporting connective tissue or even a bone (intramural type of unicystic amelobastoma)[22]. Simple enucleation is obviously inadequate treatment for intramural type of unicystic ameloblastoma and it will surely reccurr. Unfortunately, the distinguishing of three types of unicystic amelobalstoma is possible only retrospectively, from pathohystological specimens[11]. Therefore, it is much safer to radically resect all unicystic amelobalstic lesions, since available diagnostic tools do not allow us to distinguish unfavorable intramural form. Some reports propose combination of enucleation and use of liquid nitrogen or tissue fixatives, such as Carnoy's solution, supposed to be effective in elimination of cell remmants[23–25]. Such treatment might be reserved for small, unicystic, completely intrabony lesions and exclusively in the mandible. Because of anatomical considerations there is no place for such treatment for maxillary ameloblastomas.

Conclusion

Our limited experience leads us to conclude that any maxillary osseous tumors or cysts, especially those suspicious to ameloblastoma, as well as cysts larger then 35 mm require CT or MR evaluation. Special attention should be paid to posteriorely located lesions. Also, preoperative biopsy of above mentioned lesions is mandatory. In cystic lesions, especially those associated with loss of sinus walls, fine needle aspiration biopsy has been proved as an accurate diagnostic method[26]. The above mentioned recommendations and diagnostic strategy could minimize unexpectable situations during a treatment, as well as unfavorable outcomes.

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ZNAČAJ TEMELJITE PREOPERATIVNE DIJAGNOSTIKE AMELOBLASTOMA GORNJE ČELJUSTI: PRIKAZ TRI SLUČAJA

SAŽETAK

Ameloblastom, posebice onaj smješten u gornjoj čeljusti, je rijedak tumor odontogenog porijekla. Budući da ameloblastom, a naročito njegov unicistični oblik, imitira puno učestalije lezije čeljusti, dijagnoza se u značajnom broju ameloblastoma gornje čeljusti postavi tek nakon operacije. Od siječnja 1993. do prosinca 2005., na našem Odjelu su kirurški liječena tri bolesnika s ameloblastomom gornje čeljusti. Autori iznose kliničke, radiološke i patohistološke značajke ameloblastoma na ovoj rijetkoj lokalizaciji, uz poseban osvrt na značaj temeljite preoperativne dijagnostike.