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Case report

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A B S T R A C T

Ameloblastoma is a benign, slow growing but locally aggressive tumor that is clinically manifested as swelling in a jaws[1]. Prone to relapse (30%)[2] even 30 years after inadequate primary operation[3]. The most important cytological features of this tumor are small bazaloid cells in clusters, and single spindle and stellate shaped cells. We report on a case of 79-year-old patient, who was hospitalized due to tumor formation in the buccal region. FNA was performed and liquid material that contained only fagocites was collected. The conclusion was – cyst, while biopsy finding – adenoma baseocellulare, pointed to the salivary gland tumor. Patient refused the proposed surgical treatment. Four years later, the patient was urgently hospitalized due heavy bleeding from the tumor in the same region. It affected the crest portion of the upper jaw and a section of hard palate, and was bleeding on palpation.In second FNA we found phagocytes and a few small clusters of basaloid cells with palisade arrangement at the edges. Because of uniform and benign cytomorphological features it was concluded that it was a cystic tumor. On biopsy pattern the diagnose of ameloblastoma was determinated. The patient underwent surgery, however due to postoperative complications he died. Preoperative diagnosis is usually set on the basis of clinical and often nonspecific radiological findings. As it is very important to get the correct diagnose before planning an adequate surgical procedure, we would like to point out the potential value and disadvantages of FNA cytology in the diagnosis of ameloblastoma.

Key words: *ameloblastoma, cytology, tumors of the jaw*

Introduction

Ameloblastoma is a benign, slow growing but locally aggressive tumor that is clinically manifested as swelling in a jaws[1]. Prone to relapse (30%)[2] even 30 years after inadequate primary operation[3].

Because of its aggressive clinical picture characterized by locally invasive and destructive clinical behavior and benign histological features it represents a clinical – histopathological paradoks[4].

Although this is the most common odontogenictumor, it makes up less than 1% of all tumor lesions in oral cavity[1,5].

Etiology of ameloblastoma is still not fully explored, but it is considered that the appearance of tumor is associated with abnormalities in the control of genes responsible for the development of teeth[6]. The literature commonly cites trauma or cystic lesions. It is considered that ameloblastoma is developed on the basis of follicular or other odontogenic cysts, where the wall of cystic epithelium is fall to ameloblastic transformation[7].

It occurs characteristically between the third and fifth decade of life, but it has been described in children also. More than 80% of ameloblastoma is occurred in the mandible, only a smaller number of cases in maxilla. The most common location in maxillary bone is within the molar region, then in the part of the maxillary sinus and bottom of nasal cavity[7].

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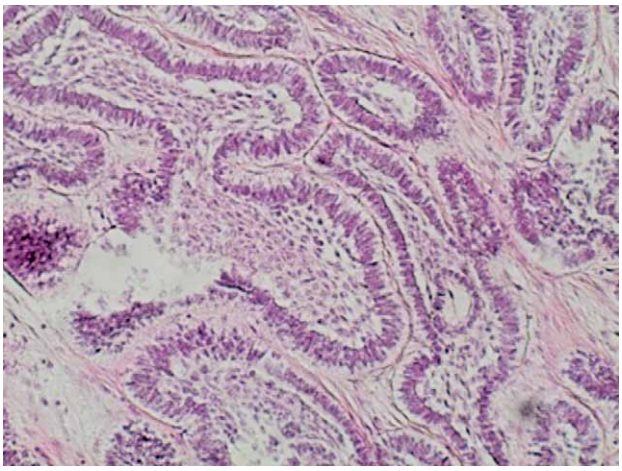

Fig. 1. Cell block section showing anastomosing clusters of stellate and spindle shaped cells with basaloid cells in palisade arrangement at the periphery of the clusters embedded in connective tissue stroma (Hematoxilyn and Eosin x 100 stain).

Radiologicaly ameloblastomas are differently presented. Some are difficult to distinguish from follicular cysts and keratocyst, for they are unilocular and associated with the crown of the tooth. Others, however, are multilocular and resemble large odontogenic keratocysts[8].

Histopathologicaly tumor is composed of proliferative odontogenic epithelium, consisted of two types of tumor cells: oval, spindle and stellate-shaped cells, and ameloblast-like columnar cells that show peripheral palisade arrangement and have basal located hiperkromatic nuclei (Figure 2-A, 2-B).Tumor appears in several variants, often with follicular or plexiform base that lies in the fibrous stroma. Follicular variant is characterized by islands of stellate-like cells that form a central cystic spaces. Plexiform variant is characterized by anastomozing cords of stellate-like cells with two or three layers of ameloblast-like cell peripherally[1]. Malignant, or metastazing ameloblastoma is extremely rare and constitutes about 1% of ameloblastoma. Its histopathologic features are indistinguishable from benign variants, but it gives local metastases in the cervical lymph nodes, or distant metastases, most commonly in the lungs, where it probably gets by aspiration[6,9,11].

Cytologic aspirates are composed of two characteristic types of cells: bazaloid epithelial cells in clusters and single spindle or stellate shaped cells. In addition to these cells, squamous epithelium, mature lymphatic cells and stromal fragmens can be found occasionaly[1, 3]. Basal cells have dark deeply chromatic nuclei and scanty, light cytoplasm, and spindle-shaped or stellate-shaped cells contain spindle or oval nuclei with loose chromatin structure and small peripherally located nucleoli. Clusters of two characteristic cells are found in two forms: clusters of spindled and stellate shaped cells with peripherally located bazaloid cells that show palisade arrangement at the edges of cluster, or tumor cells can be arranged in the form of linear streams[1]. In aspirates that contain liquid content, phagocytes are often found, and in the absence

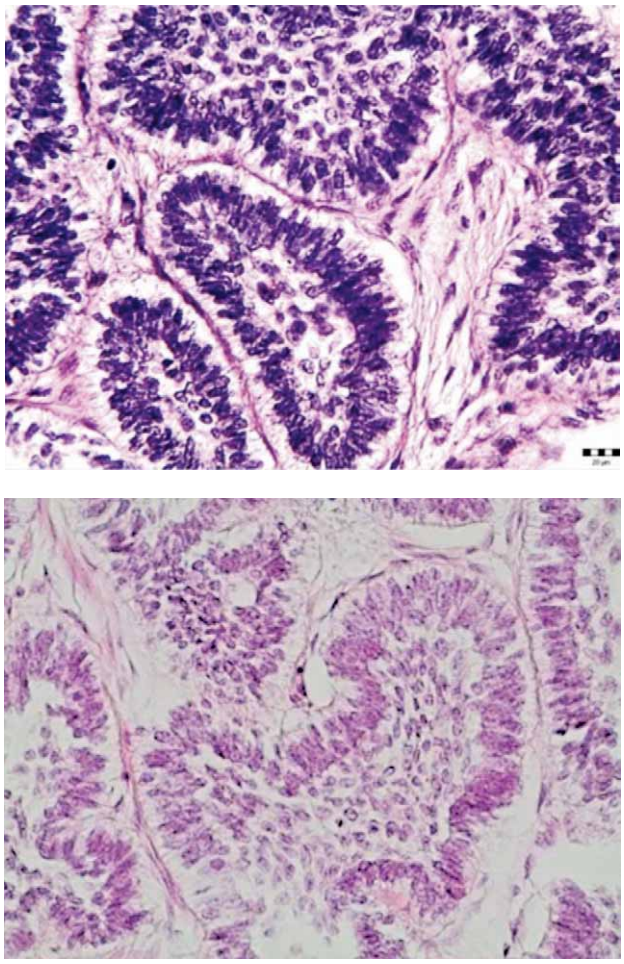

Fig. 2-A,2-B. Tissue section showing basal cells with basal located nuclei in palisade arrangement, and spindle shaped cells with loose chromatin structure and small peripherally located nucleoli (Hematoxylin and Eosin x 400 stain).

of epithelial elements they can mislead cytologist to the diagnose of an ordinary cyst.

Preoperative diagnosis is usually set on the basis of clinical and often nonspecific radiological findings. As it is very important to get the correct diagnosis before planning an adequate surgical procedure, we would like to point out the potential value and disadvantages of fine-needle aspiration cytology in the diagnosis of this rare tumor.

Case Report

79-year-old patient, who is a diabetic, hypertensive patient with cardiomyopathy, was hospitalized at the Department of Maxillofacial Surgery, Osijek University Hospital Center in 2007 due to tumor formation of the buccal region of size 4x4 cm. FNA cytology was obtained and 0.2 mL of liquid material that contained only phagocites was collected, while biopsy finding – adenoma baseocellulare, pointed to the salivary gland tumor. Patient refused the proposed surgical treatment.

Four years later, the patient was urgently hospitalized due heavy bleeding from the tumor. In the intraoral buccal region egzofitic formation of size 6x6 cm was found.

It affected the crest portion of the upper jaw and a section of hard palate, and was bleeding on palpation. On native and postkontrastic CT the described formation appeared as an irregular tumor mass, which sticked out the subcutaneous tissue, destroyed the bone structure of the lateral wall of the right maxillary sinus, filled the same, and eroded his nasal walls and minimal the roof and the bottom of the orbit. Local lymph nodes were negative.

FNA cytology was obtained and 2.5 mL of thick yellow fluid was collected. Sediments and smears, stained by May-Grünwald Giemsa and Papanicolaou, contained numerous phagocytes and only an occasional small clusters of low basaloid cells, some naked nuclei of pale chromatin structure with sometimes prominent nucleoli and abundant soft, slimy, fibrous substance in the background, which suggested the diagnose of cyst (Figure 3).

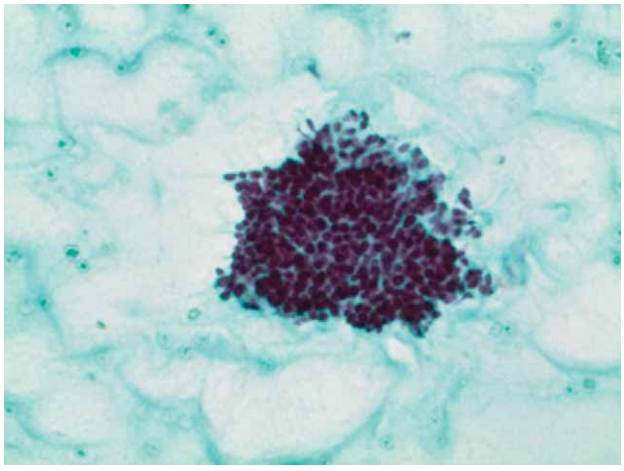

Fig. 3. Smear showing small clusterof low basaloid cells, with dark deeply chromatic nuclei that show palisade arrangement at the edges of clusters. Abundant soft, slimy, fibrous substance with phagocytes inthe background. (Papanicolaeou x 400).

On biopsy pattern the diagnose of ameloblastoma was determinated.

The patient underwent surgery where total resection of the maxilla, hard palate, buccal mucosa, and segmental resection of the mandible, with tracheotomy and reconstruction of the defect was done. Due to postoperative complications the patient died.

Discussion

Diagnostic valuable cytologic samples should contain some of the specific cellular elements, primarily small basaloid cells in clusters, and stelate and spindle-shaped cells, possibly squamous epithelial cells, fagocytes and spherical bodys[10].

In our patient in both aspirates cystic fluid was evacuated, and phagocytes were the only, or dominant type of cells. The first aspirate, for lack of specific epithelial elements, had only suggested that it was a cystic formation and ultimately it was inadequate for diagnosis. In the second aspirate we found phagocytes and a few small clusters of basaloid cells with palisade arrangement at the edges.

Because of uniform and benign cytomorphological features it was concluded that it was a cystic tumor, without considering of the possibility that it was ameloblastoma.

Several authors correctly recognized the tumor by citology speciments. In sediments of aspirated liquid Bisht et al. described bazaloid spindle shaped or round shaped cells, often in pseudopapilar arrangement[3]. Due to better cellularity it is easier to set up a diagnose on the imprint of biopsied tumor. Bokun et al. described hypercellular samples in which they described two types of tumor cells in clusters: the cells similar to ameloblasts that showed palisade arrangement at the edge while other types of cells are squamous cells of polygonal shape with a clearly defined cytoplasm in which vacuoles were visible, with regular nuclear chromatin structure. At the background of spacement amorphous protein material was found, in which were embedded naked nuclei and spindle-shaped and star-like cells with regular, oval nuclei[10] (Figure 4).

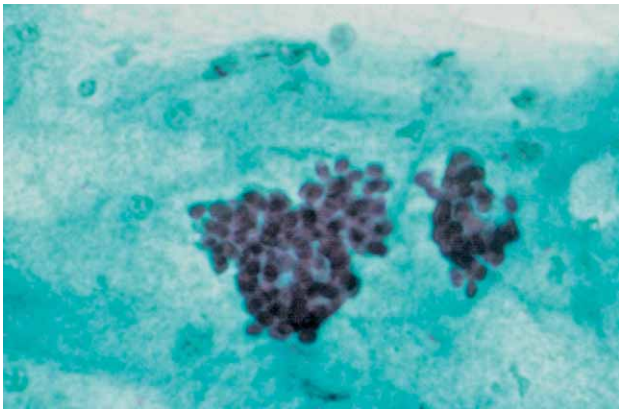

Fig. 4. Smear showing small cluster of low basaloid cells, and some naked nuclei with pale finely dispersed chromatin. Occasionally small nucleoli are seen.

Depending on the quality of the sample morphologicaly differential diagnosis of ameloblastoma includes small cell carcinoma, lymphoma, adenoid cystic carcinoma, poorly differentiated squamous carcinoma and ameloblastic lymphoma[1].

In the biopsy specimen we described anastomosing clusters of basaloid cells with a palisade arrangement at the periphery that are embedded in connective tissue stroma, (Figure 1) but those are not properly recognized as part of the jaw bone structure in the first sample, due to the anatomic localization, and small biopsy samples accompanied by incomplete clinical data, and were fig-

ured as tumor that origin in salivary gland. While the second sample was diagnosed as ameloblastoma.

Just an adequately sampled pattern, and properly concluded cytological and histopathological diagnosis is essential factor for planning the rate of surgery. Selection of appropriate surgical procedure will depend of the general health of the patient, the local extent of disease, anatomic localization of the tumor and the characteristics of the tumor itself.

As ameloblastoma is a locally aggressive tumor with a tendency to relapse, adequate surgical treatment is a decisive factor for prognosis. Conservative treatment including enukleation and curettage is associated with frequent relapses, while radical surgery significantly reduces the likelihood of recurrence.

Conclusion

Although ameloblastoma accounts for less than 1% of all tumors of the oral cavity, because of its locally destructive growth and its tendency to relapse, it is important to include it in the differential diagnosis of all cystic tumor formations that appear in the jaw, as prompt and exact preoperative diagnosis directly determines the success of treatment.

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POTENCIJALNA VRIJEDNOST I NEDOSTATCI CITOLOŠKE PUNCIJE U DIJAGNOSTICI AMELOBLASTOMA

SAŽETAK

Ameloblastom je benigni, sporastući, ali lokalno agresivan tumor koji se klinički manifestira kao oteklina u čeljusti[1]. Sklon je recidiviranju (oko 30%)[2] čak i 30 godina nakon neadekvatne primarne operacije[3]. Najznačajnija citološka značajka ovog tumora su male bazaloidne stanice u nakupinama, te pojedinačne vretenaste i zvjezdolike stanice. Prikazan je slučaj 79-godišnjeg pacijenta, koji je hospitaliziran zbog tumorske tvorbe bukalne regije.Citološkom punkcijom je dobiven tekući materijal koji je sadržavao samo fagocite te je zaključak citologa bio da se radi o cisti. Nalaz biopsije – adenoma baseocellulare, ukazivao je na tumor slinovnice. Predloženo operativno liječenje pacijent je odbio. Četiri godine kasnije bolesnik je hitno hospitaliziran zbog profuznog krvarenja iz tumora u istoj regiji. Tumor je zahvaćao greben gornje čeljusti te dio tvrdog nepca i na palpaciju je krvario. Pri ponovnoj punkciji tankom iglom u punktatu je uz fagocite dobivena po koja manja nakupina stanica bazaloidnog izgleda s palisadnim rasporedom na rubovima. Zbog monomorfije i benignih morfoloških karakteristika je zaključeno samo da se radi o cističnom tumoru. Na bioptičkom uzorku postavljena je dijagnoza ameloblastoma.Pacijent je podvrgnut radikalnom operativnom zahvatu, međutim uslijed postoperativnih komplikacija je preminuo. Prijeoperativna dijagnoza ameloblastoma se postavlja najčešće na temelju kliničkog i često nespecifičnog radiološkog nalaza. Kako je za planiranje adekvatnog operativnog zahvata važna ispravna dijagnoza, želimo ukazati na potencijalnu vrijednost i nedostatke citološke punkcije u dijagnozi ovog rijetkog tumora.