AMELOBLASTOMA OF THE JAW BONES: CLINICAL STUDY AND CASE REPORT

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BACKGROUND
Ameloblastoma (the old English word “amel” means enamel, and the Greek word “blastos” means germ) is the second most common benign odontogenic tumor of the jaw bones after odontoma (1-3). Its origin is the epithelium of ectodermal origin, which means they are tumors arising from the cells around the tooth root, or in close approximation, derived from the ectoderm (4).

Ameloblastoma is a benign but locally aggressive tumor with a high tendency to recur (2, 5, 6). Patients after ameloblastoma treatment need a life-long follow-up (2).

CASE DESCRIPTION
We present an 82-year-old female patient diagnosed with ameloblastoma and treated by us. She was not suffering from any serious diseases besides primary hypertension, which was treated with a regular intake of tablets of Nebivolol 5 mg.

The patient was referred for further paraclinical imaging tests – dental panoramic radiography (OPG) and cone beam computed tomography (CBCT). The location and borders of the lesion were determined - it was circumferentially attached to the root of 44 tooth, well-outlined linea albuginea was present, the diameter of the lesion was approximately 26 mm (Figure 1).

Bone curettage was the treatment plan - the surgical intervention in this volume was chosen as a consequence of the refusal of the patient of a partial mandibulectomy and according to her age – 82.

The histopathological examination of the curettage revealed a locally infiltrative tumor process engaging the submitted bone and fibrous tissue. The final pathological diagnosis was conventional ameloblastoma with a predominant canthomatous pattern.

On the control panoramic rentgenography one year after the operation, no pathological changes in the field of the operative site were found.

Keywords: benign odontogenic tumor, jaw tumor, maxillofacial surgery, oral surgery, pathology,
Fig. 1. Preoperative OPG of the presented female patient.

Our plan of treatment included the following: enucleation of the lesion with bone curettage as surgical treatment, histological verification and follow-up of the patient.

Bone curettage was the treatment plan - the surgical intervention in this volume was chosen as a consequence of the refusal of the patient of a partial mandibulectomy and, according to her age – 82 (the quality of life would significantly decrease).

The surgical intervention has been performed under meticulous antiseptic of the operative field with the solution of Braunol and local infiltrative anesthesia with Ubistesin 4%. A trapezoidal mucoperiosteal flap on lower-nutrient baså was raised on the vestibular side of the mandible in the region of teeth 42-46. Pathologically changed bone was resected. Tooth 44 was extracted. Tumor formation with dimensions about 26 mm in diameter was excised, and both with the resected bone were fixated in a 10% solution of Formalin and sent for further histological examination. Bone curettage of the present osteotomy defect of the mandible was performed, and then collagen cones were placed inside the defect. Hemostasis was applied. The flap was mobilized, adapted and sutured with 4/0 non-resorbable polifilament (Polyamide).

The post-operative medication therapy included antibiotics (Augmentin 3x1g per os) and analgetics (Nimezulide 2x0,1 g per os). A solution of chlorhexidine 0,1% for antisepsis was applied. The wound has been treated by the patient with Eludril - locally (once per day) for two weeks.

The post-operative period was smooth, without significant oedema, pain and other complaints.

Sutures were removed on the tenth post-operative day.

During the checkup, the wound was without any bleeding, suppuration or signs of infection, no symptoms were detected, and no complaints from the patient were declared.

On the control panoramic rentgenography one year after the operation, no pathological changes in the field of the operative site were found (Figure 2).

Fig. 2. Post-operative OPG of the presented female patient.

The histopathological examination of the curettage revealed a locally infiltrative tumor process engaging the submitted bone and fibrous tissue. The tumor (Figure 3) had a cystic-nested appearance, with a majority of nests showing an outer layer of palisading basaloid cells with reverse polarity and subnuclear vacuoles. The cells in the centers of the nests exhibited either myxoid features or showed squamous differentiation (Figure 4). There were some foci of peripheral hyaline deposition and scattered osteoclast type giant cells. The final pathological diagnosis was conventional ameloblastoma with a predominant acanthomatous pattern.

Fig. 3. Tumor comprising of a nest of odontogenic epithelium embedded in fibrous connective tissue; tumor nests show peripheral palisading, some have classical central stellate-like reticulum centers, others have cystic and squamous change. H&E, x10.
Ameloblastoma is a very rare tumor, presenting around 1% of the tumors of the oral cavity and between 9 and 11% of the odontogenic tumors (1-5). It occurs more often in the lower jaw than in the maxilla (1, 2). Around 80% of them occur in the mandible and 20% in the maxilla (1, 5). The most common location is in the posterior part of the lower jaw (mostly the molar region) compared to the anterior area (1, 2, 5-7). Ameloblastoma is a slow-growing tumor (1, 2, 4-7).

It can rarely develop into malignancy (2). According to the World Health Organization (WHO), there are two malignant types of ameloblastomas - ameloblastic carcinoma and malignant ameloblastoma (8). About 2% of people diagnosed with ameloblastoma have malignant tumors (9).

Ameloblastomas affect mostly people between 20 and 40 years of age, they can be seen rarely in children younger than ten years of age (10). The ratio of affection between men and women is from 1:1 to 1:2, according to the different studies (10, 11). The black population is more affected compared to white people (10, 12).

In 2017 WHO updated the classification of ameloblastoma, dividing the entity into three groups: conventional (multicystic), unicystic and peripheral ameloblastoma. The conventional variant is the most common one, comprising around 85% of all cases of ameloblastoma. It occurs mainly in the 3rd and 4th decades of life but is virtually present in all age groups. Conventional ameloblastoma can exhibit the following main histological patterns: follicular, plexiform, achanthomatous, granular cell, basal cell and desmoplastic. Usually, single patterns predominate in a given lesion, but sometimes two or more patterns may coexist. It is currently accepted that the histological pattern plays no role in the clinical behavior of the tumor and its prognosis (14).

In spite of the great histological versatility of conventional ameloblastoma, there are five histological traits that are suggestive of the diagnosis of the entity and play a crucial role in its distinction from other jaw lesions:

- the basal layer of cells showing elongated hyperchromatic nuclei with peripheral palisading and reverse polarity;
- subnuclear vacuoles;
- suprabasal cells with a loose arrangement, recapturing stellate reticulum formation seen in normal odontogenesis;
- lack of dentin or enamel formation.

The unicystic ameloblastoma can be further divided into three histopathological patterns: luminal, intraluminal and mural. The luminal and intraluminal patterns have a lower rate of recurrence and are good candidates for more conservative surgical treatment, while the luminal variant requires a more aggressive surgical approach, similar to the conventional ameloblastoma (15).

Peripheral ameloblastoma is the least common variant, accounting for only 1% of cases. It primarily affects patients with a mean age of 52 years and occurs most frequently in the gingiva of the mandible. It rarely presents recurrences, even when treated conservatively (16).

The histological distinction of benign ameloblastoma from malignant ameloblastoma is impossible as they show identical microscopical changes. The diagnosis can be achieved only on clinical grounds, as the malignant variant presents with metastatic spread, while the benign one can show only local invasion.

Ameloblastic carcinoma is generally more aggressive and has a worse prognosis in late disease. It has certain features of benign ameloblastoma, such as reverse polarization, peripheral palisading, and stellate reticulum-like cells. It also has features of malignancy common to many cancers, such as a high nuclear to cytoplasmic ratio, increased mitoses with atypical forms, cytological atypia, and necrosis (2).

It is usually lately diagnosed because of its poor symptoms or accidentally discovered on an X-ray (1). It is asymptomatic until patients notice swelling, expansion of the mandible or maxilla or facial asymmetry and deformity (2, 13). In some patients, teeth loss and bite changes can be one of the first symptoms (2).

The diagnosis of the tumor requires panoramic X-ray (most often orthopantomography), computed tomography (CT) imaging and a biopsy (4).

The most common differential diagnosis of ameloblastoma includes ossifying fibroma, osteomyelitis, giant cell tumor, cystic fibrous dysplasia, myeloma and sarcoma (2, 4).

Ameloblastoma spreads locally through the surrounding bone and can invade the soft tissues if there is enough time to do this (2, 17). This is why the best way of treatment is the en block surgical resection (4, 17). Excavation, curettage, cryotherapy and marsupialization are not suitable ways of treatment for this kind of tumors (17). The surgical excision with the normal bone surrounding the tumor must include a margin of a minimum of 5 mm, and if it is possible, it should be even 10 to 15 mm (2). In every case of surgical resection, some elements like age, sex, general and health status of the patient and size, localization and a histopathological variant of the tumor...
should be considered (5).

Because of the advanced age of our patient, a more conservative surgical approach was chosen to avoid post-surgical complications. In the literature, there is limited data regarding the recommended surgical behavior for ameloblastoma in the elderly population, as this tumor is extremely rare in the age group of 80+ years old).

In the early post-surgical period, gastroenterologist and dietitians can be involved because of eating issues. It takes weeks to months for the oral tissues to heal, and in this period, enteral and parenteral feeding should be considered.

Voice therapists, speech and language pathologists are needed in post-operative care after some major surgical excisions.

Because some head surgeries can result in big facial changes, plastic surgeons and medical doctors of aesthetic medicine can also be involved in post-operative care.

Radio- and chemotherapy are not used for the treatment of ameloblastoma.

The development of targeted therapy in ameloblastoma treatment is in progress, and good outcomes are expected (4).

According to different studies, the recurrence of the tumor is up to 70%, and incomplete resection is the most common reason for this (2, 5, 17).

Patients after ameloblastoma treatment need a lifelong follow-up (2, 18).

**CONCLUSION:**

The standard treatment of ameloblastoma is radical resection with a margin above 5 mm. In our case, it was not performed due to the age of the patient and her refusal – bone curettage was accepted as surgical treatment. No recurrence were observed - clinically and radiologically, after a one-year follow-up.

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