Case report

Ameloblastoma of the Lower Jaw: A Case Report

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SUMMARY

Ameloblastoma, a semi-benign odontogenic tumor, is divided into four subtypes based on the clinical presentation, radiological manifestations and histological characteristics. Radiologically, it manifests itself in the form of uni- or multicystic intrabony expansive lesions, with destruction of the medullary zone of the existing bone, resorption and possible perforation of the buccal and lingual cortex.

The patient, a 62-year-old woman, came to the Clinic for Maxillofacial Surgery in Niš with an anamnestic present painless tumefaction in the anterior segment of the mandible present for the past three years. In the course of an intraoral examination in the region of symphysis and parasymphysis of the lower jaw on the left side, an oval-shaped tumor was spotted, 5x3 cm in size; palpation showed that it was hard, painless and motionless. Multislice computed tomography (MSCT) of the oro - and hypopharynx and neck showed an extensive tumor formation of the soft tissue density in the anterior segments of the lower jaw, with the signs of destruction of bone tissue of the buccal and lingual cortex of the lower jaw.

The frequency of recurrence depends on the histological type of ameloblastoma and the degree of radicalism of the performed surgical intervention. Ameloblastoma in most cases is definitely diagnosed in the advanced stage of the disease due to the absence of symptoms and very slow tumor progression.

Key words: ameloblastoma, patohistological subtypes, surgical treatment

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INTRODUCTION

According to the classification published by the World Health Organisation (WHO) in 2005, ameloblastoma is defined as a benign, however, locally very aggressive tumor with a very high rate of relapse; it is composed of odontogenic epithelium surrounded by the fibrous stroma (1).

It is divided into four subtypes based on the clinical presentation, radiological manifestations, and histological characteristics.

Solid/multicystic ameloblastoma (SMA) is characterized by the intramural emergence of multiple cystic spaces. It accounts for 86% of all described ameloblastomas (2). This form is characterized by extremely aggressive behavior in relation to the surrounding anatomical structures and frequent recurrence in the absence of radical treatment. Division into follicular, plexiform, acanthomatous, basaloid and granulomatous form was done based on the dominance of cell types (3).

Peripheral (PA) ameloblastoma is characterized by the existence of infiltration of the surrounding soft tissue structures in most cases without the engagement of the surrounding bone (4).

Desmoplastic (DA) ameloblastoma represents the most aggressive form of ameloblastoma characterized by extreme stromal collagenation or desmoplasia.

Unicystic (UA) ameloblastoma clinically and radiologically resembles odontogenic cyst with possible intramural or intraluminal propagation of the process (5).

Radiologically, it manifests in the form of uni- or multicystic intrabony expansive lesions, with destruction of the medullary zone of the existing bone, resorption and possible perforation of the buccal and lingual cortex.

Malignant ameloblastoma is characterized by the appearance of regional or distant metastases contrary to benign histological characteristics that are similar to the benign form of ameloblastomas. Regional and distant metastases are encountered in 2% of cases (6).

Ameloblastic carcinoma is by definition a primary odontogenic malignant tumor with histological characteristics of a benign ameloblastoma followed by cellular atypia. The primary type can appear de novo, whereas the secondary type develops through malignant transformation of intrabony or peripheral ameloblastoma. Dissemination of the processes into the regional lymph nodes and distant organs is found in 3% of cases (7).

CASE REPORT

The patient, a 62-year-old woman, came to the Clinic for Maxillofacial Surgery in Niš with anamnestically present painless tumefaction in the anterior segment of the mandible present for the past three years. She did not link the growth and spontaneous enlargement of the change to subjective symptoms. The patient anamnestically also reported the existence of long-term treatment of diabetes mellitus type II and chronic arterial hypertension.

During an intraoral examination in the region of symphysis and parasymphysis of the lower jaw on the left side, an oval-shaped tumor was spotted, 5x3 cm in size; palpation showed that it was hard, painless and motionless (Figure 1). The described change was covered with an intact mucous membrane. Findings of the neck were normal, without enlarged lymph nodes.

Figure 1. Tumor in the region of symphysis and parasymphysis of the lower jaw on the left side.

Orthopantomogram (OPG) shows the existence of an expansive radiolucent unicystic formation in the segment of symphysis and parasymphysis of the mandible (Figure 2).

Figure 2. Orthopantomogram shows the existence of an expansive radiolucent unicystic formation in the segment of symphysis and parasymphysis of the mandible.

Multislice computed tomography (MSCT) of the oro- and hyopharynx and neck showed an extensive tumor formation of the soft tissue density in the anterior segments of the lower jaw, with the signs of destruction of bone tissue of the buccal and lingual cortex of the lower jaw.

The process of tumor expansion towards the floor of the mouth without signs of infiltration was present (Figure 3).

Figure 3. Multislice computed tomography of the oro-and hyopharynx and neck showed an extensive tumor formation of the soft tissue density in the anterior segments of the lower jaw, with the signs of destruction of bone tissue of the buccal and lingual cortex of the lower jaw.

Given the spectrum of pathologic entities that are included in the differential diagnosis of the described tumor formation, an excisional biopsy was performed. Histopathology showed multicystic ameloblastoma of the mandible – a follicular variant. Neoplastic odontogenic epithelium in a palisade arrangement surrounded the stellate reticulum. Squamous metaplasia was focally present. On the periphery, the remains of the bone tissue were visible (Figure 4).

Figure 4. Hematoxylin- eosin staining A. x10 ; B. x20 ; C. x20 ; D. x40.

Upon receiving the histopathological findings, under endotracheal anesthesia, a marginal resection of the lower jaw was performed in the area of symphysis and parasymphysis, extending 1.5 cm into the healthy tissue, while preserving the base of the lower jaw. Extraction of teeth 35, 41 and 42 was performed. Postoperative defect was reconstructed by using the local mucosal flaps. Two years after the surgical intervention and regular quarterly check-ups, we noted the absence of the recurrence of the tumor process. The patient refused a proposed surgical intervention in terms of reconstruction of the resected part of the lower jaw.

DISCUSSION

Ameloblastoma is a benign odontogenic tumor that is considered to originate from the dental lamina remnants, stratified squamous epithelium of odontogenic cysts or epithelium of the enamel organ. Novel theories about the origin of ameloblastoma favor the role of the absence of differentiation of the epithelium of the enamel organ in hard dental tissues in relation to others (8).

It accounts for 1% of all intraoral tumors and 10% of odontogenic tumors (9). Around 80% of ameloblastomas are localized in the region of the angle and branch of the mandible (10). The tumor is most common in the third and fourth decade of life, especially in men (11).

Unique biological behavior of ameloblastoma, a high incidence of relapse and possible malignant alteration indicate that the radical resection of jaws, extending macroscopically 1.5 to 2 cm into the healthy tissue, is a surgical method of choice in the treatment of ameloblastoma (12). Regarding the expansiveness of the tumor process, marginal or segmental resection of the jaw is indicated.

The frequency of recurrence depends on the histological type of ameloblastoma and the degree of radicalism of the performed surgical intervention. A follicular form of ameloblastoma exhibits a significantly higher recurrence than a plexiform, while the solid type of ameloblastoma also exhibits a significantly higher incidence of recurrence than all the other subtypes of ameloblastoma (13).

Recurrence of the tumor process after resection of jaws occurs in 15% of cases, while after conservative treatments that involve tumor enucleation and curettage it occurs in the range from 75% to 90% (14).

UA subtype represents the only form of ameloblastoma, which can be successfully treated with conservative methods to some extent. This claim derives from the histological evidence that the fibrous membrane that surrounds the odontogenic epithelium is significantly firmer and more compact compared to other forms of ameloblastoma (15).

The occurrence of secondary deposits is encountered in 2% to 5 % of cases. In 80% of cases, secondary deposits are present in the lungs (16). By frequency, it is followed by regional lymph nodes of the neck , pleura , diaphragm, parotid glands and liver (17).

The mechanism of metastasis of malignant ameloblastoma in the lungs has not been explained in detail yet. There are three ways of tumors

metastasizing: hematogenous, lymphatic, and aspiration. Since most of secondary deposits are localized in the lungs peripherally, it is considered that the lymphatic or hematogenous dissemination process mode is more frequent than aspiration (18).

CONCLUSION

Ameloblastoma in most cases is definitely diagnosed in the advanced stage of the disease due to the absence of symptoms and very slow tumor progression.

An extremely wide range of pathologic entities that make up the differential diagnosis of intrabony lesions, which carry a variety of therapeutic approaches, represent an absolute indication for biopsy.

Adequately performed resection of a jaw with a possible postoperative defect reconstruction and long-term follow-up of a patient is a successful method of treatment of ameloblastoma.

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Ameloblastom donje vilice: prikaz slučaja

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SAŽETAK

Ameloblastom, semi-benigni odontogeni tumor, podeljen je na četiri podtipa na osnovu kliničke prezentacije, radioloških manifestacija i histoloških karakteristika. Radiološki se manifestuje u vidu uni- ili multicističnih ekspanzivnih intra koštanih lezija sa prisutnom destrukcijom medularne zone postojeće kosti, prisutnom resorpcijom i mogućom perforacijom bukalnog i lingvalnog korteksa.

Bolesnica stara 62 godine dolazi na Kliniku za maksilofacijalnu hirurgiju u Nišu sa anamnestički prisutnim bezbolnim tumefaktom u prednjem segmentu donje vilice u prethodne tri godine. Intraoralnim pregledom se u simfiznom i parasimfiznom predelu donje vilice sa leve strane uočava tumefakt ovalnog oblika, veličine 5x3 cm, palpatorno tvrd, bezbolan i nepokretan. Na MSCT-u oro-hipofaringsa i vrata se u prednjim segmentima donje vilice uočava postojanje ekspanzivne tumorske formacije denziteta mekog tkiva sa znacima koštane destrukcije tela donje vilice, bukalnog i lingvalnog korteksa.

Učestalost recidiva zavisi od histološkog tipa ameloblastoma i stepena radikaliteta sprovedene hirurške intervencije. Definitivna dijagnoza ameloblastoma se u većini slučajeva postavlja u uznapredovaloj fazi bolesti zbog odsustva simptoma i veoma spore tumorske progresije.

Ključne reči: ameloblastom, biopsija, patohistološki podtipovi, hirurški tretman