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Twenty-year follow-up after curettage treatment for ameloblastoma – a case report and literature review

Dwudziestoletni okres obserwacji po leczeniu chirurgicznym usunięcia szkliwiaka jednokomorowego – opis przypadku i przegląd literatury

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HASŁA INDEKSOWE:
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Summary

Ameloblastoma is the most common odontogenic tumour and generally arises inside the bone. It is locally invasive, typically localized at the angle of the mandible. The nature of this lesion is benign. It is usually an asymptomatic, slow-growing, and painless tumour. Treatment depends on the type and histopathological subtype of the lesion. According to the WHO classification from 2022, five basic types of ameloblastoma are distinguished: unicystic type ameloblastoma, extraosseous/peripheral type ameloblastoma, conventional ameloblastoma (in classification from 2005 solid/multicystic type), adenoid ameloblastoma and metastasizing ameloblastoma. The most common type is the conventional one, followed by the unicystic one. Treatment is primarily surgical. We present a rare case of a patient with unicystic

Streszczenie

Szkliwiak jest najczęstszym nowotworem zębopochodnym i zazwyczaj rozwija się wewnątrz kości. Jest zmianą o charakterze miejscowo złośliwym, najczęściej zlokalizowaną w kącie żuchwy. Jest to guz, którego wzrost jest powolny i bezobjawowy. Leczenie zależy od rodzaju i podtypu histopatologicznego zmiany. Zgodnie z klasyfikacją WHO z 2022 roku wyróżnia się 5 podstawowych typów szkliwiaka: typ jednokomorowy, typ pozakostny/obwodowy, konwencjonalny (wg klasyfikacji z 2005 r. lity/wielokomorowy), gruczołowy i przerzutowy. Najpowszechniejszym typem jest typ konwencjonalny, a następnie typ jednokomorowy. Leczenie jest głównie chirurgiczne.

W niniejszej pracy przedstawiono przypadek 20-letniego okresu obserwacji pacjenta ze szkliwiakiem jednokomorowym, który został przypadkowo zdiagnozowany podczas chirurgicznego

ameloblastoma as a chance finding during the surgical removal of the lower third molar and a 20-year follow-up. This case highlights the importance of diagnostic methods and difficulties encountered in the treatment of a patient with ameloblastoma.

usunięcia trzeciego zęba trzonowego w żuchwie. Przypadek podkreśla znaczenie metod diagnostycznych i trudności w leczeniu pacjenta ze szkliwiakiem jednokomorowym.

Introduction

Ameloblastoma is a benign odontogenic tumour of epithelial origin. It was described in 1827 by Cusack, designated as an adamantinoma in 1885 by the French physician Louis-Charles Malassez, and renamed as ameloblastoma in 1930 by Ivey and Churchill.[1]

Ameloblastoma is the most common odontogenic tumour and generally arises inside the bone. It is locally invasive, typically localized at the angle of the mandible (80%). It is usually diagnosed in the 3rd and 4th decades of life; no gender predominance is noted.[2]

The exact aetiology of ameloblastoma is unknown. It may arise from the cell rest of the enamel (dental) organ, either a remnant of the dental lamina, epithelium of lining of the odontogenic cyst, or disturbances of developing enamel organ.[3] Nowadays, according to the WHO classification from 2022, five basic types of ameloblastoma are distinguished:

1) ameloblastoma, unicystic type,
2) ameloblastoma, extraosseous/peripheral type,
3) ameloblastoma, conventional (in classification from 2005 solid/multicystic type),
4) adenoid ameloblastoma, and
5) metastasizing ameloblastoma.[4]

It is usually an asymptomatic, slow-growing, and painless tumour. Wide surgical excision with adequate safe margins is the treatment of choice.[5]

The study aimed to describe a long-term observation case of ameloblastoma located in the angle of the mandible after curettage treatment.

Case report

In 2003, a 26-year-old patient presented at the Department of Oral Surgery, Medical University of Gdańsk. He complained of spontaneous pain of one week duration in the area of the lower left third molar and pain on biting. The patient did not report systemic diseases or use of any medication. On the extraoral examination, swelling and enlarged submandibular lymph nodes were noted. On the intraoral examination, the swelling and redness of the alveolar process' mucosa in the partly impacted lower third molar area were observed. On the orthopantomograph (OPG; Fig. 1), there was a visible lesion well demarcated from the bone in the left body and ramus of the mandible surrounding the lower left third molar crown. The clinical diagnosis was a difficult eruption of the lower left third molar and a coincidence with a follicular cyst (dentigerous cyst). Before the surgery, the patient took 300 mg of Clindamycin every 8h.

The partly impacted tooth was extracted under local anaesthesia, and the cyst associated with the tooth was removed. During the extraction, the operator struggled with the problem of massive bleeding that was finally arrested. Unfortunately, one root apex was left in the socket because of the difficulties during the surgery. Biological material was collected during the operation for histopathology

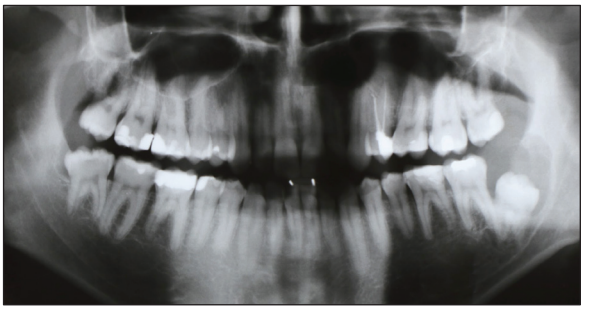

Fig. 1. Initial orthopantomography (December 2003) – large radiolucent lesion localized near tooth 38.

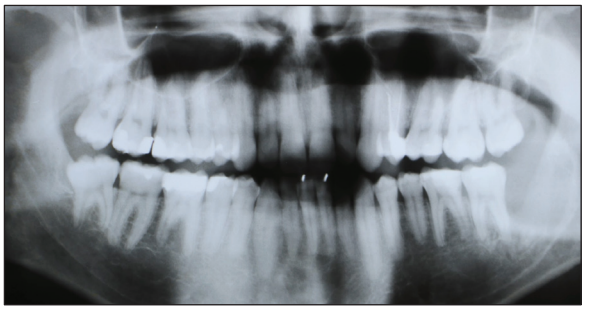

Fig. 2. Post-operative orthopantomography (January 2004) – mesial apex of tooth 38 and radiolucent lesion were visible.

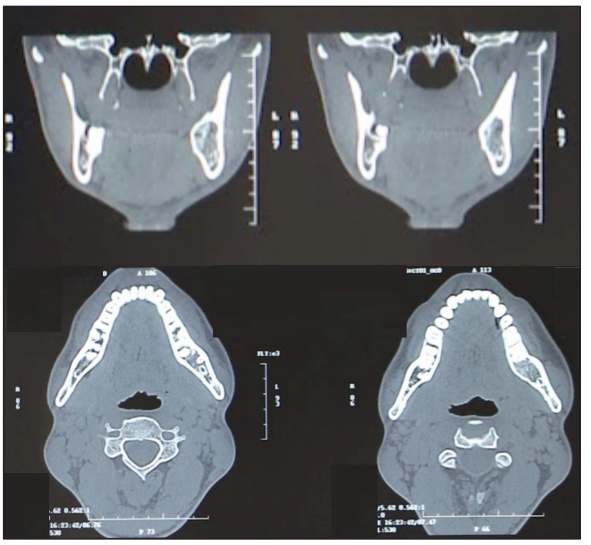

Fig. 3. Computed tomography (October 2004) – mesial apex of tooth 38 and radiolucent lesion were visible; A. coronal view; B. axial view.

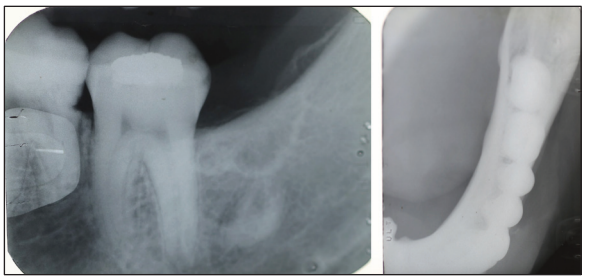

Fig. 4. Preoperative radiography (December 2004) – mesial apex of tooth 38 was visible.

examination. The result of the test was: ameloblastoma unicysticum (UA).

After the surgery, an additional OPG (Fig. 2) and computed tomography (CT, Fig. 3) were made. The OPG and CT scan revealed the retained tooth apex and the post-resection bone loss involving the vestibular bone surface of the alveolar part of the mandible. Diagnostics were broadened, including ultrasound of the neck lymph nodes, chest X-rays, and abdominal ultrasound, to exclude distant pathologies.

The treatment continued one year later, in December 2004. Under local anaesthesia, the left apex was extracted, and the area of curettage was expanded. The aim of the procedure was to do it minimally invasively, only within the alveolar part of the mandible. Radiography before and after the surgery was performed (Fig. 4). During the procedure, the apex was removed (Fig. 5). The sample material was subjected to histopathological examination for the second time. Two fragments were collected: the first one was the resected soft tissue (22×10×10 mm), whereas the second one was the resected bone (12×8×8 mm). The histopathologic examination revealed ameloblastoma unicysticum (UA) with fibrous

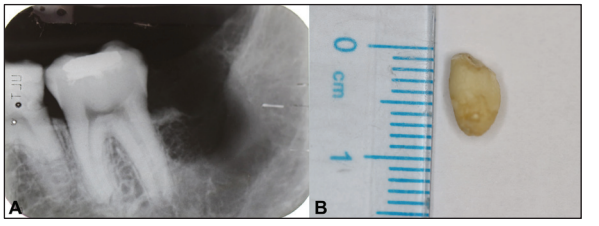

Fig. 5. A. Postoperative radiography (December 2004) and 5B. The mesial apex of tooth 38.

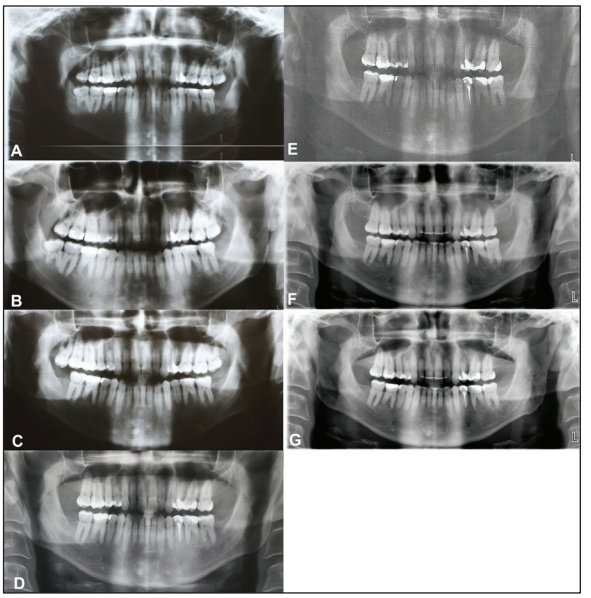

Fig. 6. Follow-up – OPG; A. March 2005; B. October 2005; C. November 2008; D. November 2014; E. November 2018 F. September 2020; G. November 2023.

elements. Additionally, a drain was introduced to the post-operative wound to prevent hematoma formation. Swelling appeared a few days after the surgery, but the patient did not experience any pain. The drain and the stitches were removed after one week.

Clinical and radiological controls took place in March 2005, October 2005, November 2008, November 2014, November 2018, December 2020, and November 2023 (Figure 6A-6G). The evidence of normal bone repair was noticeable. Signs of recurrence were not found.

Discussion

Ameloblastoma is usually asymptomatic, so each patient should have an orthopantomographic radiograph taken at least every two years or before tooth extraction, especially for the mandibular third molars. Ameloblastoma can sometimes imitate follicular cyst, both on clinical and radiological examination. A similar situation was present in this case. Collecting dental follicles during the extraction of the impacted teeth and passing them on for the histopathological examination is essential. If the diagnosis of ameloblastoma is made, then the radical treatment is preferred – wide surgical excision with safe margins (minimum 1-1.5 cm). A typical radiological diagnostic tool is cone beam computed tomography (CBCT) or CT to evaluate the tumour's extent and the operative margins. Its purpose is not only to decide about the resection treatment but also to plan the following reconstruction. Conservative treatment can be insufficient for many variants of ameloblastoma.[6]

Ameloblastoma of the unicystic type (UA) is a rare type of ameloblastoma, accounting for about 6% of all ameloblastomas. It usually occurs at a younger age – 2nd decade of life and is associated with impacted teeth. UA is considered to be a less aggressive form of ameloblastoma that can usually be successfully removed with enucleation or curettage. Only more invasive types need a more extensive approach – radical, wide surgical excision. Each case involves complex issues to avoid subsequent recurrence. The classical treatment approach starts with an accurate radiological examination – CBCT, CT, and sometimes magnetic resonance imaging (MRI). Radiologically, the unicystic type of ameloblastoma is osteolytic, usually lucent, and frequently unicystic or multicystic shape with well-defined sclerotic margins at the angle of the mandible surrounding the impacted tooth. There are different classifications of unicystic ameloblastoma. Ackerman distinguishes three histological types: luminal UA (tumour conned to the luminal surface of the cyst), intraluminal/plexiform UA (nodular proliferation into lumen without infiltration of tumour cells into connective tissue wall), and mural UA (invasive islands of ameloblastomatous epithelium in the connective tissue wall not involving the entire epithelium).[7] Histologic

subgrouping by Philipsen and Reichart has also been proposed: subgroup 1 – luminal UA; subgroup 1.2 – luminal and intraluminal; subgroup 1.2.3 – luminal, intraluminal and intramural and subgroup 1.3 – luminal and intramural.[8]

Treatment depends on the histological variant and the diameter of UA. The procedure algorithm if the diameter is under 3 cm: CBCT/CT exposed semi-ocular jaw bone osteolysis, and the following classical treatment is surgical enucleation. When the histologically confirmed type is luminal, intraluminal or mural with slight proliferation into the tumour capsule, it allows at least 10-year clinical and radiological observation. A histologically confirmed mural variant with deep proliferation into the tumour capsule requires mandible marginal or part resection and at least 10 years of clinical and radiological observation. If the diameter is **over** 3 cm, CBCT/CT exposed semi-ocular jaw bone osteolysis and the following biopsy is required. Each of these conditions must be fulfilled: histologically confirmed type – luminal or intraluminal; localization – mandible and bundle bone preserved. Enucleation is the typical treatment with the addition of aggressive methods – after operative nitrogen or Carnoy liquid application. This approach allows at least 10 years of clinical and radiological observation.[4,9-13]

If at least one condition is fulfilled - mural variant, localization - maxilla or pathologically destroyed bundle bone - treatment requires mandible marginal or part resection and at least 10-year clinical and radiological observation.[9-13]

It is a tumour with a strong propensity of recurrence, especially when the ameloblastic focus penetrates the adjacent tissue from the cyst's wall. Luminal unicystic ameloblastomas are less aggressive and respond better to conservative treatment. In contrast, mural types frequently result in recurrence. Malignant transformations of ameloblastoma are rarely seen (about 1% of cases). Malignant ameloblastoma may arise *de novo* or transform into a pre-existing ameloblastoma. The most common sites to spread are the lungs.[3]

Exceptional for this case was the surgical saving approach – curettage. In 2004, there was no histopathological distinction of unicystic ameloblastoma, so the saving treatment was not typical for such cases. At that time, the treatment was based on mandible resection. Due to the limited margins, uniocularity, small size of the tumour, no infiltration, and young age of the patient, the decision was made to perform a non-invasive operation. After 20 years of observation, a great result was reported with bone healing and no tumour recurrence.

Conclusions

It is imperative to diagnose ameloblastomas following clinical, radiological, and histopathological examination. Since many different intraosseous tumours and cysts can imitate ameloblastomas, histopathology examination is crucial. Each ameloblastoma case treatment should be considered individually. Nowadays, treatment of unicystic ameloblastoma depends on histopathological variant and presents a few options. After 20 years of observation, UA curettage proved a successful form of conservative treatment.

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