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Fine-Needle Aspiration Biopsy of Ameloblastic Carcinoma of the Mandible: A Case Report

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The use of fine-needle aspiration biopsy (FNAB) in the diagnosis of odontogenic tumors seems to have attracted little attention. The presence of a firm preoperative diagnosis helps preventing suboptimal surgery, contributing to avoid recurrence of these tumors. A case of ameloblastic carcinoma of the mandible diagnosed by FNAB is presented in this report, illustrating its effectiveness for preoperative diagnosis of odontogenic tumors. A 74-year-old female presented with a painless swelling in the right mandibular angle. A panoramic radiograph revealed a radiolucent lesion in the body of the mandible. Cytological smears from FNAB in the area revealed basaloid cells with a palisade arrangement and presence of stellate-shaped cells. These cytological features lead to the diagnosis of ameloblastoma. However, when there are atypical cells and atypical mitoses, as in the present case, diagnosis of ameloblastic carcinoma may be established. The patient underwent chemotherapy, showing remission of the lesion after treatment. FNAB is a minimally invasive, safe, fast and inexpensive method for diagnosing benign and malignant ameloblastomas, which ensures that patients have a proper treatment without the need of performing an incisional biopsy, especially in neoplastic cases.

Key Words: fine-needle aspiration biopsy, cytology, ameloblastoma, odontogenic tumor.

INTRODUCTION

Fine-needle aspiration biopsy (FNAB) has gained wide acceptance in the medical field for being a practical, safe and accurate technique. However, this technique has been less used among dental or oral and maxillofacial surgeons in the diagnosis of the oral soft tissue and bone or salivary gland lesions (1,2).

Radiolucent lesions of the mandible represent a variety of metabolic, inflammatory and neoplastic disorders that may be quite variable in appearance and require different treatment approaches (2,3). FNAB is an accurate method for the presurgical diagnosis of these oral lesions, ensuring that patients have an adequate treatment.

A case of ameloblastic carcinoma of the mandible diagnosed by FNAB is presented, illustrating its effectiveness for preoperative diagnosis of oral tumors.

CASE REPORT

A 74 year-old-female patient with chronic renal insufficiency and autoimmune hemolytic anemia presented a painless swelling in the right mandible angle.

The panoramic radiograph revealed a well-delimited, unilocular radiolucent lesion on the body of the mandible (Fig. 1). The clinical-radiological differential diagnosis was ameloblastoma, brown tumor

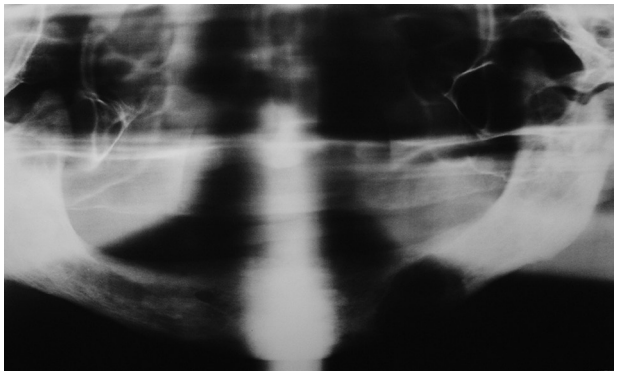

Figure 1. Panoramic radiograph showing radiolucent lesion of the mandible (arrow).

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and metastatic carcinoma.

FNAB of the lesion was proposed to obtain a presurgical diagnosis that could assist the surgical plan. FNAB was performed with a 22-gauge needle and 10-mL syringe connected to a cytoaspirator. Subsequently, alcohol-fixed cytological smears were obtained and stained with hematoxylin and eosin, and a blood clot that was fixed in formol, embedded in paraffin (cell block) and was cut into histological sections and stained with hematoxylin and eosin.

Microscopic examination revealed highly cellular smears (Fig. 2A) with cells in a palisade arrangement. Cellblock revealed a dual population of basaloid and stellate- shaped cells (Fig. 2B) as well as atypical cells and mitotic figures (Fig. 2C). The diagnosis revealed a malignant odontogenic neoplasia consistent with ameloblastic carcinoma. The patient showed remission of the lesion after chemotherapy.

DISCUSSION

Ameloblastomas are the most frequent odontogenic tumors, with great clinical significance, accounting for one percent of tumors and cysts of the mandible, and an estimated incidence of 0.3 cases *per* million people *per* year (3). Ameloblastomas are epithelial-derived odontogenic tumors (from cell remnants of the enamel organ, epithelial lining of an odontogenic cyst or from basal layer cells of the oral mucosa) (4).

These tumors commonly occur in the third to fifth decades of life with no preference based on gender and ethnicity, accounting for eighty percent of the cases in the mandible (4).

Three clinical patterns of ameloblastoma have been described in the literature based on clinical and radiological findings and prognostic factors (2). Multicystic or intraosseous is the most common and clinically important ameloblastoma variant due to its focal invasion feature with a trend towards recurrence and metastasis. Similar to the present case, the second variant is the unicystic ameloblastoma presented as a unilocular well-defined radiolucent area that affects mainly young patients. The third variant is the extraosseous ameloblastoma indicated by the presence of a pedunculated mass protruding from the gingiva (3).

Clinical and radiological findings of ameloblastoma can simulate several odontogenic cysts and tumors. Many non-odontogenic tumors and tumor-like lesions as central giant cell granuloma, eosinophilic granuloma, aneurismatic bone cyst, brown tumor and metastatic tumors must be considered in the differential diagnosis (5). Considering the age and clinical history of our patient and the radiological findings, differential diagnosis included ameloblastoma, brown tumor and metastatic carcinoma.

Five patterns are histologically distinguished: follicular, plexiform, basal cell, acanthomatous, and granular cell type. The plexiform and follicular variants are the most common ones (4).

Since ameloblastomas require complete excision with adequate margins to minimize recurrence, the preoperative diagnosis is important to determining the surgical technique. Recurrence may result from an inadequacy or failure of the primary surgical procedure and occur in more than 50 percent of the cases within the first 5 years after surgery (5).

There are two morphological ways of establishing the biological nature of the lesions preoperatively: histological and cytological investigation along with clinical and radiological findings.

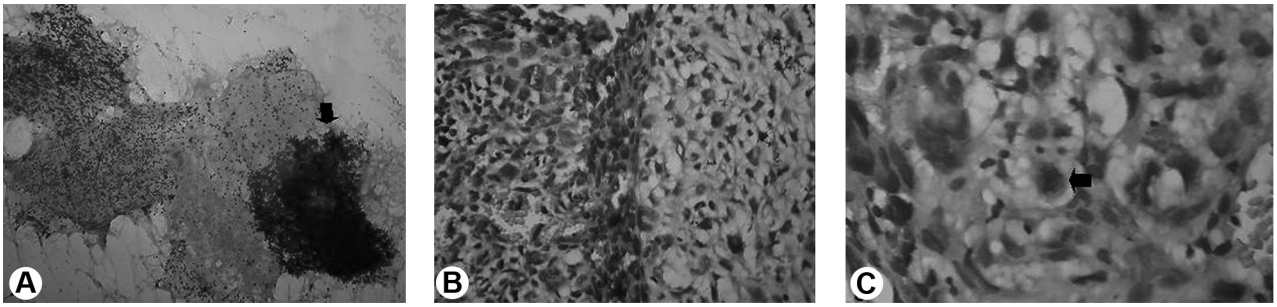

Figure 2. Panel of photomicrographs for microscopic examination. A = Highly cellular and hemorrhagic smear, with clusters of cells (arrow). Hematoxylin-eosin, ×100 magnification. B = Dual population of cells: basaloid type on the left and stellate-shaped cells on the right side. Cellblock, Hematoxylin-eosin, ×250 magnification. C = Detail showing atypical cells with large and pleomorphic nuclei and atypical mitotic figure (arrow). Cellblock, Hematoxylin-eosin, ×400 magnification.

FNAB accuracy rate for oral cavity lesions represents 80 to 94.5% (1,5,6), and only few cases had insufficient material for the cytological diagnosis (5). Because this is a minimal-invasive technique that does not require the preparing of the patient previously, it is safe enough for pregnant women, children or high-risk patients (5). As the patient of this case has hemolytic anemia, FNAB is well indicated because it causes minimal blood loss during the procedure.

Ramzy et al. (2) were the first to describe the findings of ameloblastoma diagnosed by FNAB, characterized by the presence of basaloid, squamous, stellate-shaped or spindle cells in the aspirates. Radhika et al. (7) included to that description the presence of epithelial cells in a palisade arrangement, and Günhan et al. (6) observed the presence of spherical keratinized bodies in squamous cells. In the present case, the cytological features were basaloid cells, peripheral palisade pattern and stellate-shaped cells consistent with the diagnosis of ameloblastoma.

The description of several cases of ameloblastoma diagnosed from FNAB results (1-3,5-12) reveals that this tumor has sufficiently distinctive cytological features to make this diagnosis by FNAB possible.

Even though ameloblastomas are well studied and documented (over 3,600 cases of ameloblastomas have been described in the literature), little is known about their malignant features, as fewer than 60 cases of ameloblastic carcinoma have been reported (13).

Ameloblastomas rarely exhibit a malignant behavior with development of metastasis. The term ameloblastic carcinoma was introduced by Shafer in 1983 (14) and is an extremely rare odontogenic tumor (15,16). The incidence of malignancy has been estimated in 1% of the cases. Waldron (4) suggested that the term malignant ameloblastoma must be applied to tumors with histological features of ameloblastoma in the primary and metastatic tumors. This has been usually applied to tumors with malignant cellular features (increase in the nucleus/cytoplasm ratio, nuclear hyperchromatism and atypical mitosis) present in the primary tumor, in the recurrence or in the metastasis (4,13,14).

This carcinoma occurs in a wide range of age groups, but the mean age is 30.1 years, as in ameloblastomas (14). The most common site of occurrence is the posterior portion of the mandible (14,17,18). It is very rare in the maxillary region (13,14,18). There is no apparent sex predilection, but some authors have described predominance in males (13). The most common sign described has been swelling, although others included associated pain, rapid growth, trismus and dysphonia (13,14,17,18). The patient of this case presented a painless swelling in the right mandible angle, the most common sign and site of ameloblastic carcinoma. Her age was different from most cases described, but there are cases of ameloblastic carcinoma in patients over 70 years of age (15).

There are only two cases of ameloblastic carcinoma diagnosed by FNAB in the literature (15,19), which have the same cytological features of the present case. Sharma et al. (20) reported a case of malignant ameloblastoma that had malignant features, as atypical mitosis and vascular invasion, only at histology, showing that the cytological diagnosis of malignancy can be difficult. In the present case, a diagnosis of ameloblastic carcinoma was thereby rendered based on the presence of atypical cells and atypical mitotic figures associated with typical features of ameloblastoma.

Adequate sampling is very important to accurate diagnosis, and is especially important when attempting to distinguish ameloblastoma from ameloblastic carcinoma (21) and ameloblastic carcinoma from carcinomas in the jaw metastasizing from lung, breast and the gastrointestinal tract (14). Paraffin-embedded blood clots obtained by FNAB (cellblock) previously fixed in formalin or alcohol have allowed an assessment of the specimen without artifacts that can be present in cytological smears, such as cellular overlap. This provides a better evaluation of the atypical cells and the histological pattern of the lesion. In the present case, stellate-shaped cells and atypical cells could be better observed in cellblock, vital features in establishing the final diagnosis.

Cellblock specimen can still be used in performing complementary tests that may assist diagnosis, as immunohistochemistry and molecular biology. Therefore, not only smears, but also blood clots and liquid proceeding from FNAB have to undergo a pathological examination.

The radiographic features of ameloblastic carcinoma are similar to ameloblastomas. In most cases one can observe a radiolucent intraosseous lesion (16), as in the case described here.

The lung is the most common area of distant metastasis (16,17), but metastasis to the skull (16) and lymph nodes (21) have also been described. In the present case, the patient had not metastasis at the diagnosis.

FNAB can be an effective technique not only

for the presurgical diagnosis of odontogenic tumors, but also to assess recurrence and metastases (22,23), and to assist the diagnosis mainly in non-surgical cases.

Controversy still exists regarding the treatment of ameloblastic carcinoma. Some authors have suggested surgery plus radiotherapy, while other doubt the effectiveness of this association (13). There are scarce reports on chemotherapy regimens for ameloblastic carcinoma (13). In this case, surgery was not done because the patient had chronic renal insufficiency and autoimmune hemolytic anemia. Chemotherapy alone had good results, with remission of the lesion.

FNAB is a minimally invasive, safe, fast and inexpensive method for diagnosing benign and malignant ameloblastomas, which ensures that patients have a proper treatment without the need of performing an incisional biopsy, especially in neoplastic cases.

RESUMO

A punção aspirativa por agulha fina (PAAF) para o diagnóstico dos tumores odontogênicos tem atraído pouca atenção. A presença de um diagnóstico pré-operatório ajuda a prevenir tratamentos subótimos e evita recidivas no caso destes tumores. Um caso de PAAF de carcinoma ameloblástico é apresentado, ressaltando a eficácia deste método para o diagnóstico pré-operatório dos tumores odontogênicos. Paciente feminina, de 74 anos, com abaulamento indolor na região mandibular à direita. O RX panorâmico mostrou lesão radiolúscida no corpo da mandíbula. Realizada PAAF da área, cujos esfregaços citológicos mostraram arranjo em paliçada de células basalóides e presença de células de aspecto estrelar. Tais características citológicas levam ao diagnóstico de ameloblastoma, porém na presença de atipias celulares e de mitoses atípicas, como no presente caso, o diagnóstico deve ser de carcinoma ameloblástico. Após o diagnóstico a paciente foi submetida à quimioterapia. A PAAF é um método minimamente invasivo, seguro, rápido e barato para o diagnóstico de ameloblastomas benignos e malignos, que permite que os pacientes tenham um tratamento apropriado sem a necessidade de biópsia incisional, especialmente em casos neoplásicos.

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