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Original Article

Classification proposed of malignant intraosseous odontogenic tumors (MIOT)

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ABSTRACT

Malignant intraosseous odontogenic tumors (MIOT) of the jaws are very rare. The diagnosis is difficult. Clinical, paraclinical and histological diagnostic criteria, strict are well established. But the International Union Against Cancer (UICC) does not provide TNM classification that will allow harmonization of the treatment. Indeed, despite their location, they cannot be classified as primary tumors of the oral cavity because of their localization in the bone marrow, making them systematically classified as T4.

We propose a classification taking into account the clinical and radiological data.

1. Introduction

According to the World Health Organization (WHO) classification published in 2005 [1], odontogenic tumors constitute a group of heterogeneous diseases that range from hamartomatous or non-neoplastic tissue proliferations to benign neoplasms to malignant tumors with metastatic potential. These tumors are derived from epithelial, ectomesenchymal and/or mesenchymal elements of the tooth-forming apparatus. The majority of odontogenic tumors arise de novo, without an apparent causative factor. They are divided into benign and malignant tumors [1]. When these tumors are within the maxillofacial skeleton they are intraosseous or centrally located. The tumors are extraosseous or peripherally located when they occur in the soft tissues overlying tooth-bearing areas. The large majority of odontogenic tumors occur intraosseously. Malignant odontogenic tumors of the jaws are very rare with unknown etiology and classified in odontogenic carcinomas and odontogenic sarcomas [1].

In the new WHO classification, odontogenic carcinomas [1] include metastasizing (malignant) ameloblastomas, ameloblastic carcinomas (primary type, intraosseous secondary type, peripheral secondary type) and primary intraosseous squamous cell carcinomas (type solide, derived from keratocystic odontogenic tumor, derived from odontogenic cysts), clear cell odontogenic carcinomas and ghost odontogenic cell carcinomas. Odontogenic sarcomas include ameloblastic fibrosarcomas and ameloblastic fibrodentino- and fibro-odontosarcoma [1].

2. Discussion

2.1. Malignant intraosseous odontogenic tumors

Malignant intraosseous odontogenic tumors (MIOT) arise centrally in the jaws, with no communication with the upper aerodigestive tract mucosa. MIOT can provide from a solid tumor that invades bone marrow spaces and induces osseous resorption, or a cancer arising from the lining of an odontogenic cyst, or a cancer in association with a benign epithelial odontogenic tumors.

Criteria to define a MIOT are those used to describe primary intraosseous odontogenic carcinomas [2]: (1) Histological evidence of a malignant tumor with an intraosseous starting point, (2) absence of ulcer formation on the overlying oral mucosa except due to trauma or tooth extractions, (3) absence of a distant primary tumor at the time of diagnosis and at least 6 months during the follow-up period. In case of death before 6 months an autopsy eliminates a distant tumor.

To eliminate the possibility of distant primary tumor, chest radiographs, TEP-scan, and endoscopy of the gastrointestinal system and upper respiratory tract should be performed during the diagnostic phase and follow-up period [2,3]. But when the tumor destroys the cortex and merges with the surface mucosa, it may be difficult to distinguish between a MIOT and a cancer arising from the oral mucosa or a primary tumor of the maxillary sinus.

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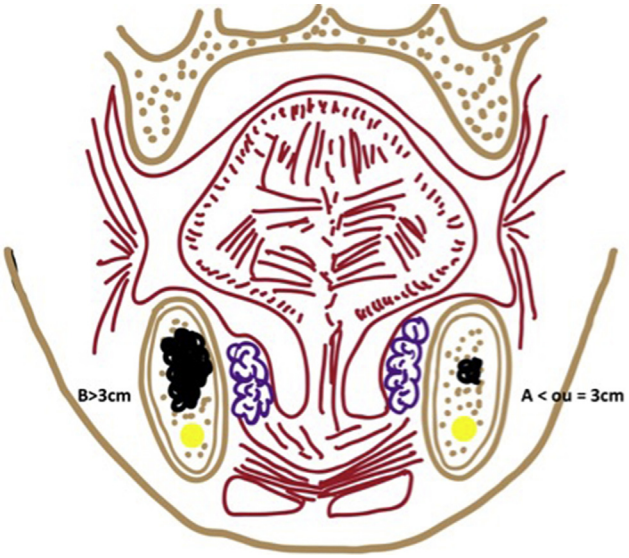

Fig. 1. Stage T1: strictly intraosseous tumor. This tumor will be classified T1A if its largest diameter is less than or equal to 3 cm and T1B if its largest diameter is greater than 3 cm.

2.2. Clinical features

The incidence of MIOT is unknown. Males and females are affected and the sex ratio varies depending on the tumor [1]: sex equality (ameloblastic carcinoma primary type), male predominance (primary intraosseous squamous cell carcinomas solid type and derived from odontogenic cysts, ghost cell odontogenic carcinoma, ameloblastic sarcoma, ameloblastic fibrodentino, and fibro-odonto sarcomas) and female predominance (clear cell odontogenic carcinoma) [1].

Some MIOT arise preferentially in older patients (ameloblastic carcinomas, primary intraosseous squamous cell carcinomas derived from keratocystic odontogenic tumor, clear cell odontogenic carcinomas) and others are predominant in young patients (primary intraosseous squamous cell carcinomas solid type, primary intraosseous squamous cell carcinoma derived from odontogenic cysts). And others have a wide age range (ghost cell odontogenic carcinomas, odontogenic sarcomas, ameloblastic fibrodentino, and fibro-odontosarcomas) [1].

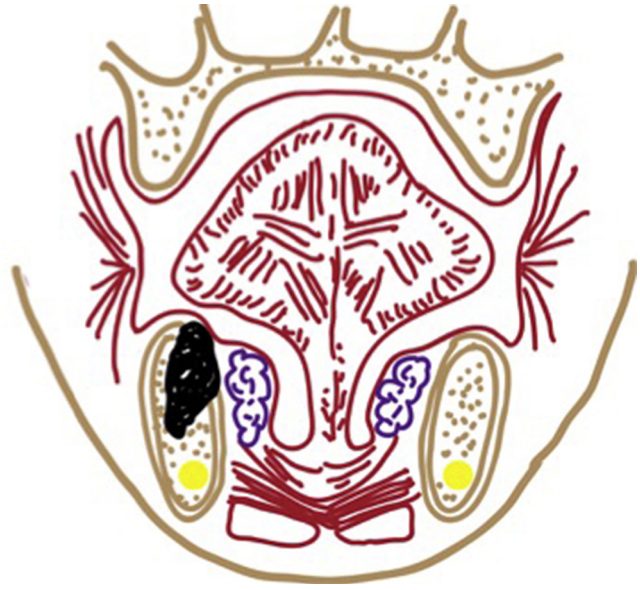

Fig. 2. Stage T2: tumor has exceeded at least one cortical without invasion of adjacent soft tissues. This tumor will be classified T2A if its largest diameter less than or equal to 3 cm and T2B if its largest diameter is greater than 3 cm.

Fig. 3. Stage T3: Tumor invading adjacent soft tissues, whatever the size.

Usual clinical features of MIOT are nonspecific (painful jaw swelling with rapidly developing or not) [2,4,5]. But some symptoms are very worrying: bleeding, ulceration of the oral mucosa, mobility of teeth, lower lip paraesthesia or anesthesia, involvement of neck lymph nodes and distant metastases. In several cases, loosening of teeth, and non-healing extraction sockets aided to the right diagnosis.

However most cases are asymptomatic and are discovered incidentally during the course of routine dental radiographs [1,4].

Imaging features are nonspecific: radiographic appearance will vary from radiolucent to radiopaque. Intraoral dental radiographs and panoramic radiography are usually the first means for identifying the presence of an intraosseous lesion [1,6]. But some aspects may lead to question the diagnosis: bony cortical destruction with ill-defined borders or a fracture, surrounding soft-tissue or surrounding cavity (maxillary sinus, nasal or orbital cavity) invasion [5].

CT-scan and MRI are mandatory to evaluate tumor extension (local, regional, metastases).

MIOT are more often found in the body and posterior parts of the mandible, exceptionally in the maxilla (ghost cell odontogenic carcinomas). Maxillary locations are most frequently observed in the anterior segment [1–4].

The diagnosis is histological and immunohistochemical. It specifies the type of cancer.

2.3. Prognosis and treatment modalities

The prognosis of MIOT is poor [1,2,6]. The location in the bone marrow is an important factor of metastatic migration. Recurrences are common. MIOT spread both regionally and distantly and may even cause tumor related death.

Due to the small number of cases of MIOT there is no consensus on specific treatment protocols or guidelines for these tumors. But the poor prognosis justifies an aggressive approach [2,3,6]. Surgical resection with tumor free-margins is the therapy of choice. This surgery associated to with postoperative radiotherapy gives the best results mostly in tumors eroding or exceeding cortical bone [4]. In order to limit the functional and esthetic sequelae reconstruction is often necessary.

In case of tumor chemosensitivity, chemotherapy may be used before surgery to reduce tumor size and limit the width of the surgical resection, hence limiting sequelae. But often chemothera-