CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 41 (2017) 71–75

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Malignant fibrous histiocytoma in the right portion of the mandible with metastasis in pancreas

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ARTICLE INFO

Article history:
Received 18 August 2017
Received in revised form
26 September 2017
Accepted 27 September 2017
Available online 12 October 2017

Keywords:
Malignant fibrous histiocytoma
Metastasis
Pancreas
Gastric
Tumor
Case report

ABSTRACT

BACKGROUND: Malignant fibrous histiocytoma is a sarcoma of uncertain origin that can be found both in soft tissues and in bones. It is currently called undifferentiated pleomorphic sarcoma not otherwise specified and it represents a final common pathway in several tumors that are subject to the progress of dedifferentiation.

Local recurrence of the tumor in the same location where it was originated occurs in 20–30% of the total number of soft tissue sarcomas. It is less frequent in limbs and more likely to recur in retroperitoneal sarcomas and in head and neck.

Most tumors grow during the first two years after treatment.

Most sarcoma metastases, especially those in the head and neck (including malignant fibrous histiocytoma) present as a lung disease (90%). Extrapulmonary diseases are not frequent and can occur in lymph nodes (10%), bones (8%) and in the liver (1%).

CASE REPORT: 61 years old woman with history of malignant fibrous histiocytoma in the right portion of the mandible, resected four years ago. Subsequent checkups did not show any disease. The patient reports a constant abdominal pain not diagnosed, related to a pancreatic nodule.

CONCLUSION: Malignant fibrous histiocytoma is a potentially curable disease. The most important part of the treatment is complete surgical removal, usually followed by adjuvant radiation therapy. Chemotherapy is a treatment usually used only on those patients with higher recurrence risk or on those showingrecurrence or who have already showed recurrence.

1. Background

Malignant fibrous histiocytoma (MFH) is a sarcoma of uncertain origin found both in soft tissue and in bones [1–3]. It can appear as primary bone injury or as a result of a preexistent condition such as fibrous dysplasia or Paget disease [2,4].

MFH is the most common soft tissue sarcoma found in adults mostly aged from 20 to 70 years old [4,5]. Even though the MFH is considered the most common soft tissue sarcoma, it can also appear as a primary bone tumor [2]. Both fibrosarcoma and primary bone MFH differ from osteosarcoma in that they lack traces of malignant osteoid material [6], which can affect any bone, but in most cases, it affects the femur, followed by the tibia and the humerus [4,7].

Most patients suffer pain, swelling or a pathologic fracture [2,4,5]. In terms of radiology, tumors are not clearly defined lytic lesions and they can have a considerably soft tissue mass.

MFHs are more frequent in lower extremities and retroperitoneum and less frequent in the head and neck (3–10%) [2]. The nasal cavity and the paranasal sinuses are mostly affected (54%), and consequently, MFHs can affect the maxillary alveolar bone [2] and lead to laryngeal, maxillary, and mandibular sinus injuries. These are the most severe cases [2].

The incidence of MFH in the mandible is not frequent and incidence risk is only 3% of all MFHs in bones [2].

There are five different histologic subtypes: (1) storiform/pleomorphic, (2) myxoid, (3) giant cells, (4) inflammatory and (5) angiomatoid. The most frequent subtype is storiform/pleomorphic. Angiomatoid and myxoid present the most favorable prognosis of them all, whereas MFH with giant cells is the most harmful of all subtypes. The latter is considered a high-grade lesion in terms of local growth, recurrence and distant metastasis [2].

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https://doi.org/10.1016/j.ijscr.2017.09.026

Due to the variable range of histological differentiation, MFHs show no regular images in computerized tomography (CT) and magnetic resonance (MRI). In most cases, the tumor is observed as a round bulk with no clear sides and it always pervades contiguous bones and soft tissues. Density signal intensity is diverse. Necrosis can be observed in hyperintense areas as well as calcification and ossification (in 5–20% of these areas) [4,5].

Currently, there is no reported evidence of malignant fibrous histiocytoma having metastasized in pancreas and stomach.

The present work is reported in line with the SCARE criteria [8].

2. Case report

2.1. Personal history

61-year-old female patient, with no pathological history of drug use, illegal drugs, tobacco, alcohol abuse or genetic alterations, with a history of right lower jaw malignant fibrohistiocytoma, completely removed 4 years ago.

2.2. Pathology

2.2.1. Macroscopic properties

Portion of right side mandibulectomy. A 2 × 2 cm tumor formation with unclear edges can be observed.

2.2.2. Microscopic properties

High-grade pleomorphic sarcoma (pleomorphic malignant fibrous histiocytoma). Clean margin.

Right submental node, jugular level 2 and facial without neoplastic cells.

Extension studies with chest CT were normal.

Ongoing adjuvant radiation therapy, no adjuvant chemotherapy according to the latest results from the European Organization for Research and Treatment of Cancer (EORTC) showing good tolerance and response to the indicated therapy.

Subsequently, craniofacial, neck, thorax and abdominalclinical and tomographic follow-up was performed, showing no pathology.

Three years after maxillary MHF resection, the patient consults for continuousepigastric pain of moderate intensitythat evolved for a few days, without any other accompanying symptom.

The physical examination showed no alteration of the abdominal semiology. Laboratory and CT control is requested.

2.3. Laboratorytests

Erythrocytes 4.1 × 10”12/L; hemoglobin 134 g/l; hematocrit 0.41; leukocytes 5.3 × 10”9/L (0.76 segmentedneutrophils. 0.18 lymphocytes. 0.6 monocytes); urea 3.2 mmol/L; creatinine 92 mol/L; blood sugar 9.7 mmol/L; ionogram within regular values.

2.4. Additional procedure

Abdominal CT: partially necrotic 3 × 2 × 7 cm nodule with hypodense center in the tail section of the pancreas.

Normal CA 19.9 and CEA.

CT IMAGES

- Abdominal CT: pancreas, nodule image with periferal contrast.Hypodense center

In view of the impossibility of an accurate etiological diagnosis, and in accordance with pathological antecedents, the general surgery department, together with the multidisciplinary committee, and the patient’s consent, decides on surgical procedure.

2.5. Treatment

Diagnostic laparoscopy was performed showing negative results for peritoneal implants and hepatic metastasis. A 4 cm pancreatic tumor was found, possibly harmful for splenic vessels.

A laparotomy and a cavity evaluation were performed. Besides the tumor in the tail section of the pancreas, a 3 cm tumor in the body section of the stomach was also found. Splenectomy, distal pancreatectomy and partial gastrectomy with Roux-en-Y anastomosis were performed.

Samples are sent for anatomical pathology study.

Sample images

- Nodule in the body section of the pancreas.
- Lesion in the body section of the stomach.

2.5.1. Clinical progress

Patient undergoes post-surgical treatment in the critical care unit showing positive clinical evolution. On the tenth day of admission, due to positive evolution, hospital discharge, with indications, and scheduled medical check-ups are indicated.

Two years after surgery, the patient continues under scheduled check-ups, showing no oncological disease, and with high risk of relapse, without a standardized adjuvant treatment.

2.5.2. Histological report

Stomach and pancreas: high-grade pleomorphic sarcoma (pleomorphic malignant fibrous histiocytoma). COMPATIBLE WITH LOWER MAXILAR PRIMARY INJURY

2.6. Clean margin

Immunohistochemistry techniques with monoclonal antibody through the streptavidin-biotin-peroxidase method showing DAB chromogen on the slide showed the following: Negative pankeratine and CD45; positive lysozyme, vimentin, S-100 (only in giant cells) and CD68.

SLIDE IMAGES FOR PATHOLOGICAL ANALYSIS

- Immunohistochemistry of malignant fibrous histiocytoma.
- Histology of malignant fibrous histiocytoma.

3. Discussion

According to the World Health Organization, bone MFH is a malignant neoplasia composed of fibroblasts and polimorphic cells with a mostly storiform pattern [6]. Mandibular involvement resulting from MFH is not frequent (less than 30 cases reported) and is mainly located in the back of the mandible.

Soft tissues in extremities and in the retroperitoneum are usually the most affected [7]. A 70% of the cases start as primary tumors and 30% derive from preexistent conditions (such as radiation therapy on the affected area) [2,5].

The best choice is surgical resection with wide margins related to adjuvant chemotherapy [4–6]. Intralesional or narrow margin resection is related to high rates of recurrence and metastasis. Chemotherapy without surgical resection is not effective [2,5,7].

Local recurrence after exeresis is 16–52% and it depends on the size, spreading and positive microscopic margins [2]. Most recurrence cases occur during the first 2 years after treatment [7], and most cases with metastatic dissemination in sarcomas, particularly in the head and neck, are present as lung diseases (90%). Metastasis in extrapulmonary locations are not frequent and can occur in lymph nodes (10%), bones (8%) and in the liver (1%) [1].

Regarding histologic diagnose, with highly undifferentiated cells, it requiered a wide immunohistochemistry panel to deter-