

Pancreatic metastasis from undifferentiated pleomorphic sarcoma. An extremely rare entity

Metástasis pancreática de sarcoma pleomórfico indiferenciado. Una entidad extremadamente rara

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Abstract

Undifferentiated pleomorphic sarcoma (UPS) is a very aggressive malignant tumor that originates in bone or soft tissues, being its most frequent location in the extremities. We present the case of a 71-year-old man who, two years of follow-up after a lower right limb UPS excision surgery, presented a single metastasis of sarcomatoid origin in the pancreas that was treated by cephalic pancreaticoduodenectomy. Sarcomatoid metastases to the pancreas are extremely rare. Resection of the metastasis is the only therapeutic alternative that has reported benefit in terms of the survival of these patients.

Keywords: Soft tissue sarcoma. Metastasis. Pancreatoduodenectomy. Pancreas.

Resumen

El sarcoma pleomórfico indiferenciado (SPI) es un tumor maligno muy agresivo que se origina en el hueso o tejidos blandos, siendo su localización más frecuente en las extremidades. Presentamos el caso de un varón de 71 años que a los dos años de seguimiento tras cirugía de exéresis de un SPI presenta una metástasis única de origen sarcomatoide a nivel del páncreas que se trató mediante duodenopancreatectomía cefálica. Las metástasis sarcomatoides a nivel de páncreas son extremadamente raras. La cirugía de exéresis de la metástasis es la única alternativa terapéutica que ha reportado beneficio en cuanto la supervivencia de estos pacientes.

Palabras clave: Sarcoma de partes blandas. Metástasis. Duodenopancreatectomía. Páncreas.

Introduction

Undifferentiated pleomorphic sarcoma (UPS), also known as malignant fibrous histiocytoma, is the most common soft tissue sarcoma in adults. Its approximate incidence is 1 for every 100 000 individuals¹.

It is more common in men, between the 6th and 7th decades of life, and is most frequently located in

the lower extremities¹. Recurrence is predominantly local. The lung is the most common site of distant metastasis, being pancreatic metastases extremely rare¹.

Only 2% of tumors located in the pancreatic gland are metastases, and their most common presentation is in the context of a disease already disseminated to other organs^{2,3}. The cancers most frequently causing

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metastases, generally solitary and localized at pancreatic level, are renal, followed by colorectal, melanoma, breast cancer, lung carcinoma, and sarcoma^{2,3}.

The benefit of pancreatic resection in these patients will be determined by the primary origin and its histopathological characteristics².

Up to 50% are asymptomatic and diagnosed during follow-up of the primary tumor². Morphology and radiological characteristics vary based on their hypothesis. Positron emission tomography may be useful to rule out metastases at other levels².

Case report

This is the case of a 71-year-old man who underwent wide musculoskeletal excision for a UPS located in the upper third of his right thigh. Adjuvant treatment was administered with radiotherapy (66 Gy) and chemotherapy (ifosfamide and doxorubicin). Pathological anatomy of the surgical specimen revealed a 13 cm x 11 cm x 6.8 cm UPS with free surgical margins. Due to its degree of differentiation, mitotic count, and tumor necrosis, it was categorized as grade 3 (stage IIIb) (AJCC, 8th edition)⁴.

Two years after surgery, a computed tomography of the thorax, abdomen, and pelvis revealed the presence of a 50 mm x 47 mm heterogeneous mass at the duodenopancreatic junction without dilatation of the bile duct or Wirsung's duct (Fig. 1). Percutaneous biopsy suggested a sarcomatoid origin of the mass, prompting a cephalic duodenopancreatectomy (Figs. 2 and 3). Histopathological diagnosis confirmed the presence of a 5 cm x 4.5 cm x 4.2 cm UPS metastasis originating in the pancreatic uncinate process, with tumor-free margins and negative lymphadenopathies (Fig. 4). The patient had a good and uneventful postoperative recovery. He remains asymptomatic and without signs of recurrence at the 1-year follow-up.

Discussion

Pancreatic metastases of sarcomatoid origin are extremely rare². Most reports on this topic consists of case series and isolated case reports. After the World Health Organization's reclassification from 2002⁵ only 3 cases of single pancreatic metastases from UPS have been reported^{1,3,6}. Radical resection surgery is the only potentially curative treatment, and is associated with increased disease-free intervals^{1,2}. There is

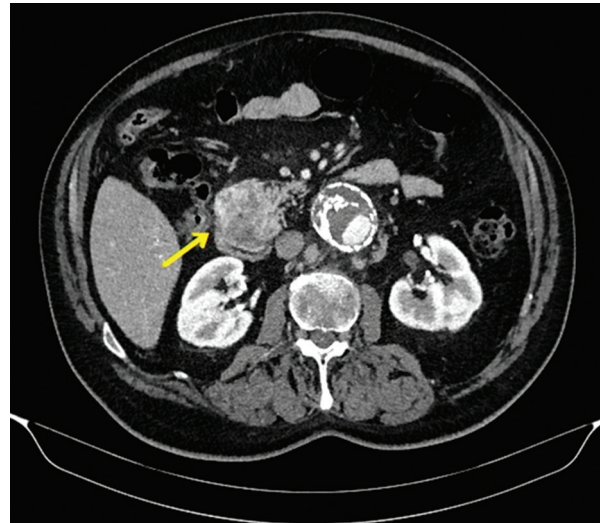


Figure 1. Axial view of abdominal computed tomography: solid and heterogeneous mass of 50 mm x 47 mm at the duodenopancreatic junction.

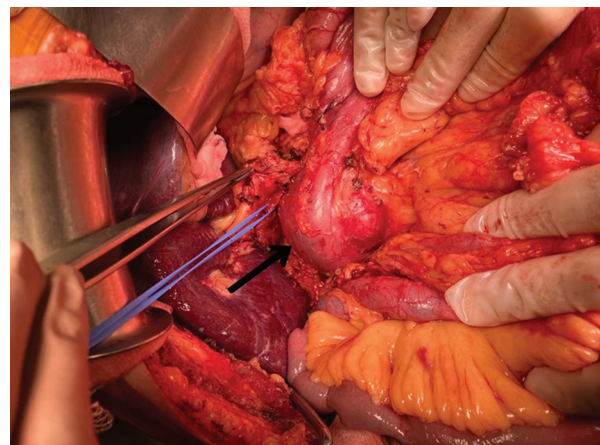


Figure 2. Intraoperative image of a single mass in the pancreatic head-body uncinate process and duodenal compression.

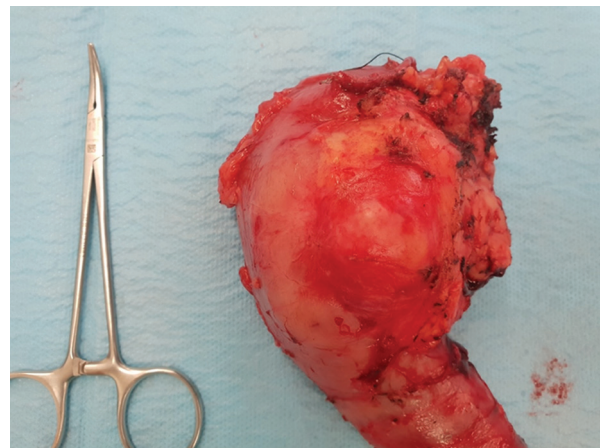


Figure 3. Surgical specimen after cephalic duodenopancreatectomy with pylorus preservation.

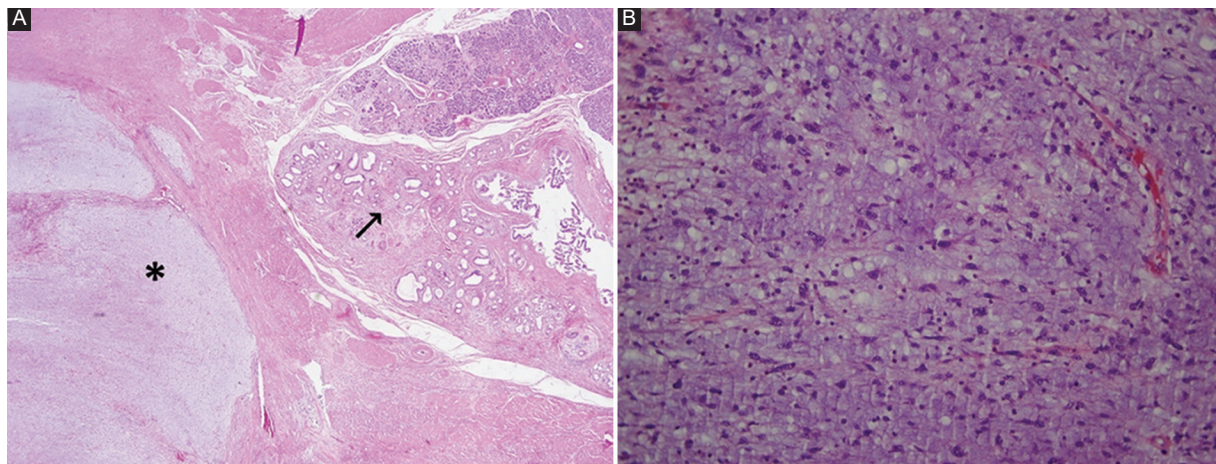


Figure 4. Pathological anatomy with hematoxylin-eosin staining: **A:** relationship of the tumor (asterisk) with healthy pancreatic tissue (arrow) (X2). **B:** spindle-shaped atypical tumor cells (X20).

no consensus on the benefit of adjuvant chemotherapy or radiotherapy regarding these tumors¹.

Atypical pancreatic resections or enucleations increase the risk of local recurrence and have a high rate of pancreatic fistulas, which is why standardized pancreatectomy is advised^{2,7,8}. Such surgeries should be performed in high-volume centers specialized in pancreatic surgery².

Patient prognosis depends on the aggressiveness of the primary tumor, with renal origin tumors having a better prognosis⁹. Local recurrence is a common thing.

The absence of prospective studies and meta-analyses on single pancreatic metastases of sarcomatoid origin prevents accurate knowledge on the prognosis and survival of these patients.

Conclusions

Published series on pancreatic metastasis resections are limited, but authors report a long disease-free interval in many patients. Therefore, although treatment should be individualized, surgery is advised whenever possible^{2,3,10}.

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Conflicts of interest

None declared.

Ethical disclosures

Protection of human and animal subjects. The authors declare that no experiments were performed on humans or animals for this study.

Confidentiality of data. The authors declare that they have followed the protocols of their work center on the publication of patient data.

Right to privacy and informed consent. The authors have obtained the written informed consent of the patients or subjects mentioned in the article. The corresponding author is in possession of this document.

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