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Pseudopapillary and Solid Tumors of the Pancreas: Case Report and Review of the Literature

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Abstract **Case Report**

The pseudopapillary and solid tumor of the pancreas (TPPSP) is characterized by a malignant potential attenuated with a low risk of local extension and rare metastatic evolution. We report an observation of TPPSP, in a 62-year-old man, revealed by chronic epigastralgia. The diagnosis of cystic tumor was made on the abdominal CT scan. Surgical treatment consisted of a left splenopancreatectomy. The histological study confirmed the diagnosis of TPPSP.

Keywords: Pseudopapillary, diagnosis of TPPSP, epigastralgia, splenopancreatectomy

I. INTRODUCTION

Pseudopapillary solid tumor of the pancreas is a rare tumor accounting for 2% of exocrine tumors of the pancreas. It occurs more frequently in adolescents and young women, and rarely in children. It is unique not only in its macroscopic appearance, but also in its histology. Described for the first time by Frantz in 1959, it has been given several names, but the WHO adopted pseudopapillary as its name in 1996. Adjuvant treatment is not indicated. The observation of a new case of TPPSP provides an opportunity to clarify the clinical and therapeutic features of this tumor.

II. CASE REPORT

A 62-year-old man was admitted for chronic epigastralgia without other associated signs, evolving for 4 months in a context of apyrexia and preservation of general condition.

The patient was admitted to the emergency department, where clinical examination revealed painful palpation of the left hypochondrium and epigastrium. Abdominal CT revealed a voluminous mass of the posterior epiploon, 10x8cm; well limited, solid cystic in favour of a mesenchymal tumour, penetrated by a voluminous branch of the splenic artery with respect for the planes of cleavage with the pancreas, stomach, spleen and angle of Treitz. The pancreas was of normal size and there was no adenopathy. Surgical exploration revealed a mass of 15x10cm polylobed and cystic back cavity of the omentum dependent on the tail of the pancreas and adherent to the mesocolon. The mass was resected, including part of the pancreatic tail. Anatomoclinical and immunohistochemical studies were in favor of a 14 cm solid pseudopapillary neoplasm of the pancreas. A postoperative CT scan showed a cystic lesion formation in the posterior cavity of the omentum, measuring 4.47x8.23x9.3 cm, hypodense and air-bubbled with discrete peripheral enhancement and at the level of the partitions, posteriorly filling the compartment of the caudal part of the pancreas. A collection in the left colonic angle measuring 2.3x5.4 cm with discrete enhancement in the neighbouring ACG, in contact with the first collection on the right and the left colonic angle on the left of the peri-lesional infiltration and mesenteric fat in the epigastrium, right parietocolic gutter and subcutaneous median abdominal fat infiltration of post-surgical reactive appearance. A few millimetric coelio mesenteric and latero aortic lymph nodes were noted, with a discrete pelvic peritoneal effusion. An exploratory puncture was performed with no sign of malignancy, and the patient is currently under surveillance with no sign of recurrence after 2 months.

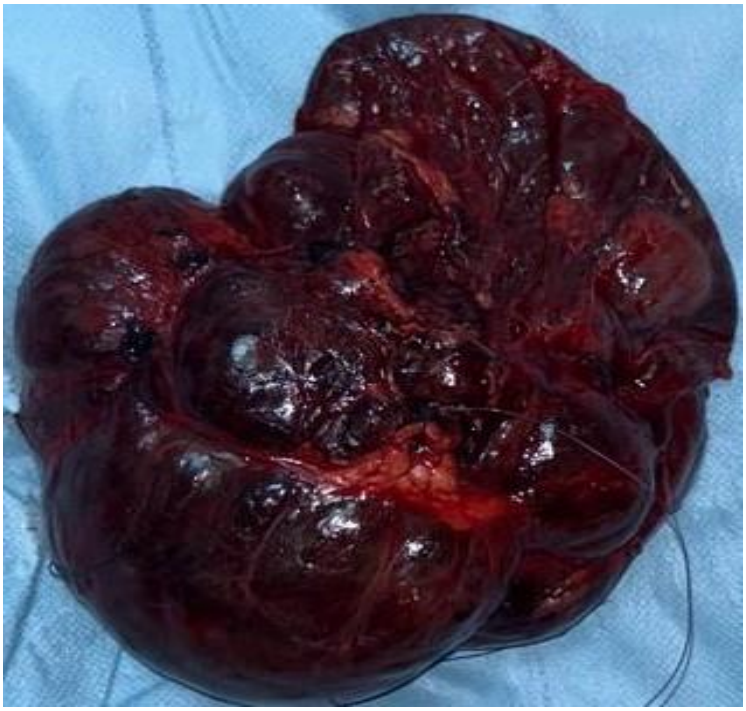

Image 1: Photograph of the surgical specimen

III. DISCUSSION

TPPSP are rare, accounting for less than 2% of exocrine pancreatic tumours and less than 5% of cystic pancreatic tumours [4, 5]. In the 718 cases of TPPSP reported in the literature, there is a clear female predominance, exceeding 90%, with an average age of 22 years [5]. These tumours are exceptionally seen in males and children [2, 4, 6]. The clinical signs of TPPSP are not specific [1, 7, 8]. The symptomatology depends on the location and size of the tumour [4,8]. It is often revealed by supra-umbilical abdominal pain or an abdominal mass [2, 3, 7], as was the case in our observation. Occasionally, as the tumour increases in size, it may cause compression of neighbouring digestive, biliary or vascular structures [2]. It may be revealed in the aftermath of abdominal trauma, or on the occasion of a complication such as rupture or intratumoral haemorrhage [3, 6, 9]. Ultrasound, CT and MRI can describe cystic, mixed and solid forms [2, 3, 8]. They usually show a well-limited, poorly vascularized mass that develops preferentially in the corporocaudal region of the pancreas (64% of cases) [2-6]. Ultrasound may show hypoechoic, homogeneous or heterogeneous images, depending on the size of the cystic areas [6, 10]. Abdominal CT shows a heterogeneous, hypodense lesion with partial peripheral enhancement [3, 10]. Ultrasound and abdominal CT scans in both our patients showed pancreatic lesions with dual cystic and solid components. Magnetic resonance imaging is the most effective examination. It shows hemorrhagic foci hyperintense on T1 and T2, surrounded by a capsule that is often hypointense on T2-weighted sequences [10]. This examination, carried out on a 14-year-old girl, was useful for orientation, as it showed a cystic lesion with solid contents. The differential diagnosis in adults is mainly neuroendocrine tumours and pseudocysts of the pancreas, and in children pancreatoblastoma [3]. TPPSP is diagnosed on pathological analysis of the resection specimen [3-5]. The macroscopic appearance is often characteristic: the tumor is usually well limited, rounded, encapsulated, partially or totally necrotic with hemorrhagic and cystic changes [1, 8]. It is often large, averaging 10 cm [4, 10]. The microscopic appearance is quite suggestive: a tumoral proliferation consisting of monomorphic polygonal or cubic cells in solid or pseudopapillary regions. The cytoplasm is sparse, poorly eosinophilic and often contains PAS granules that may correspond to alpha-1-antitrypsin [8]. The nucleus is rounded with finely dispersed chromatin. Mitosis and cytonuclear atypia are exceptional [1]. Immunohistochemical study confirms the diagnosis, eliminating tumors of another nature [3]. Tumor cells express vimentin, alpha-1-antitrypsin and NSE in 90% of cases [8]. Progesterone receptor positivity is remarkable. In our patients, tumour cells expressed vimentin and alpha-1-antitrypsin in both cases, and NSE in one.

TPPSP is treated exclusively surgically, ranging from a simple lumpectomy to partial or even total pancreatectomy, depending on the topography of the tumour [2-4, 6]. Exeresis should be as complete as possible, avoiding over-conservative resections which expose the patient to the risk of tumour recurrence [5]. Simple enucleation of the tumour has been proposed by some authors, in view of the low degree of malignancy, but this entails a higher risk of local recurrence, particularly in the case of capsular invasion [2]. Accessible metastases and invaded adjacent organs should be resected wherever possible. Whatever the surgical technique used, prolonged monitoring

(ultrasound and abdominal CT scan) should be systematically performed in search of locoregional or metastatic (mainly hepatic) recurrence. Zhang *et al*., [2] reported a case of local recurrence three years after surgical removal of a TPPSP. Cheng-Hong *et al*., [4] reported a case of liver metastasis 14 years after tumor removal. The prognosis of TPPSP is favourable, with prolonged mean survival, especially after controlled excision surgery [3-5, 7].

IV. CONCLUSION

TPPSP is a rare, slowly evolving anatomic and clinical entity with attenuated malignancy. It occurs preferentially in young women, rarely in children. The diagnosis, rarely made preoperatively, should be made in the presence of any pancreatic mass. The prognosis is excellent after complete surgical excision.

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