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Case report

Frantz Tumor: Case Report

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Abstract

Solid pseudopapillary neoplasm (Frantz tumor) is a rare pathological entity, with a higher incidence in young women, and is typically classified as a tumor with low malignant potential. Clinical manifestations are nonspecific and depend on the anatomical location involved. A case is presented involving a 27-year-old female patient with a 10-month history of diffuse abdominal pain and a 5-kg weight loss over the preceding six months. Endoscopic ultrasound with biopsy revealed a 45 × 45 mm solid-cystic mass. A Whipple pancreatoduodenectomy was performed, with postoperative follow-up at eight days showing no complications. Histopathological analysis confirmed a solid pseudopapillary tumor with negative resection margins. This neoplasm accounts for approximately 0.2% to 2.7% of pancreatic tumors, and more than 50% of cases are asymptomatic, often discovered incidentally on imaging studies. Treatment is based on complete surgical resection, with documented five-year survival rates exceeding 90%. These tumors are exceedingly rare and are associated with an excellent long-term prognosis following complete surgical excision.

Keywords

Neoplasm, benign neoplasm, pancreatic neoplasms, pancreatoduodenectomy.

INTRODUCTION

Pancreatic tumors are classified as exocrine or endocrine and may be either malignant or benign. Benign tumors are rare and are generally diagnosed only once they reach a considerable size, as they produce obstructive symptoms affecting the posterior wall of the stomach or compromise the endocrine pancreas. Clinical manifestations are nonspecific and depend on the anatomical region involved; however, these tumors do not produce metabolic alterations or elevation of biomarkers[1,2].

Frantz tumor is a solid pseudopapillary exocrine pancreatic neoplasm with low incidence, accounting for less than 1% of pancreatic tumors. It occurs more frequently in young female patients (mean age: 28 years; 89%), in whom early diagnosis and complete surgical resection are associated with a 5-year survival rate exceeding 95%[1]. It is considered to have benign behavior due to its low metastatic potential (approximately 15%), with lymph node involvement and recurrence being uncommon[1,3]. Given the low incidence of this condition, the following case is reported.

CASE PRESENTATION

A 27-year-old female patient with no significant past medical history, originally from Samaná, Caldas, a home-

maker, presented with a 10-month history of symptoms. She reported diffuse abdominal pain predominantly in the upper abdomen, associated with a 5 kg weight loss over the previous six months and early satiety. Physical examination revealed a palpable mass in the right hypochondrium. The patient was initially evaluated in the outpatient setting, where an abdominal ultrasound was performed. The report described a "rounded mass adjacent to the head of the pancreas, hypoechoic, with well-defined margins and minimal Doppler signal, measuring 5.1 × 4.7 × 4.6 cm, with an estimated volume of 59 mL." Based on these findings, evaluation by general surgery was requested. The surgical team recommended endoscopic ultrasound with biopsy. Endoscopic ultrasound demonstrated the following findings: "A solid-cystic mass measuring 45 × 45 mm, with poorly defined hyperechoic septa and 20 mm cystic components, located in the head and neck of the pancreas. The lesion wall is well defined, hyperechoic, and rounded, with hypervascular areas, without calcifications or perilesional lymphadenopathy. The lesion compresses the mesenteric vessels but does not invade them. The celiac trunk, portal vein, and splenomesenteric confluence are normal. No free fluid is observed in the abdominal cavity" (**Figure 1A** and **B**). Biopsy was performed during the same procedure (**Figure 1A**).

Preoperative triphasic computed tomography (CT) was requested to exclude portomesenteric involvement. Imaging demonstrated a large 45 × 45 mm mass without involvement of the mesenteric vein (**Figure 2**).

A Whipple pancreatoduodenectomy was performed. Intraoperatively, a large tumor of the pancreatic head was identified, intensely vascularized, with multiple large efferent veins draining into the portal vein and superior mesenteric vein. The procedure was completed without complications (**Figure 3**). The patient remained in the intensive care unit (ICU) for three days, followed by two additional days of hospitalization. She was discharged and evaluated at follow-up eight days later without complications. Pathology confirmed the presence of a solid pseudopapillary tumor with negative surgical margins (**Figure 4**).

DISCUSSION

Pancreatic tumors account for approximately 7% of deaths caused by neoplasms. These tumors are classified into exocrine (95%) and endocrine (5%) neoplasms. Ductal adenocarcinoma (85%) is the most frequent exocrine tumor, whereas solid pseudopapillary tumor, also known as Frantz tumor, represents approximately 0.2%–2.7% of the remaining cases[4]. This entity belongs to the group of pancreatic cystic tumors, which may be classified as unilocular or multilocular, neoplastic or non-neoplastic, and composed of epithelial or mesenchymal tissue[5]. This tumor is defined as an uncommon pancreatic lesion characterized by slow growth and low malignant potential, with a reported mortality of approximately 2%. It is typically composed of solid areas, pseudocysts, and pseudopapillary structures interspersed with regions of necrosis and hemorrhage[6].

The tumor is named after V. K. Frantz, who first described it in 1959 in the *Atlas of Tumor Pathology*, based on a patient who underwent pancreatoduodenectomy and died during the procedure[3,7]. At that time, it was defined as a papillary

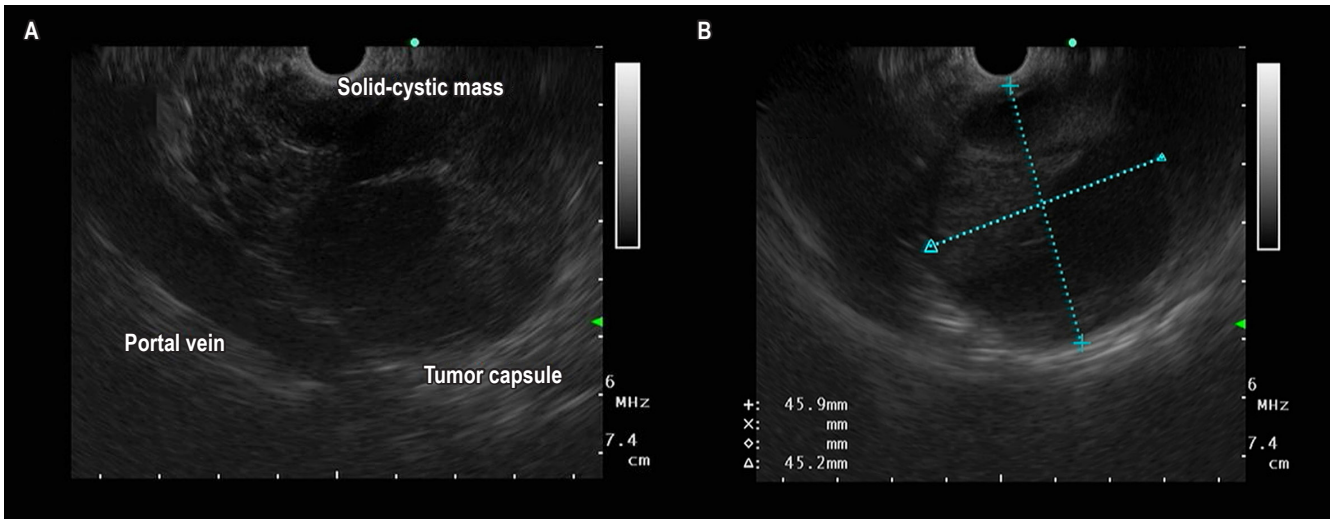

Figure 1. Pancreatic endoscopic ultrasound showing a solid-cystic mass in the head of the pancreas, well defined with a thick capsule. Images property of the authors.

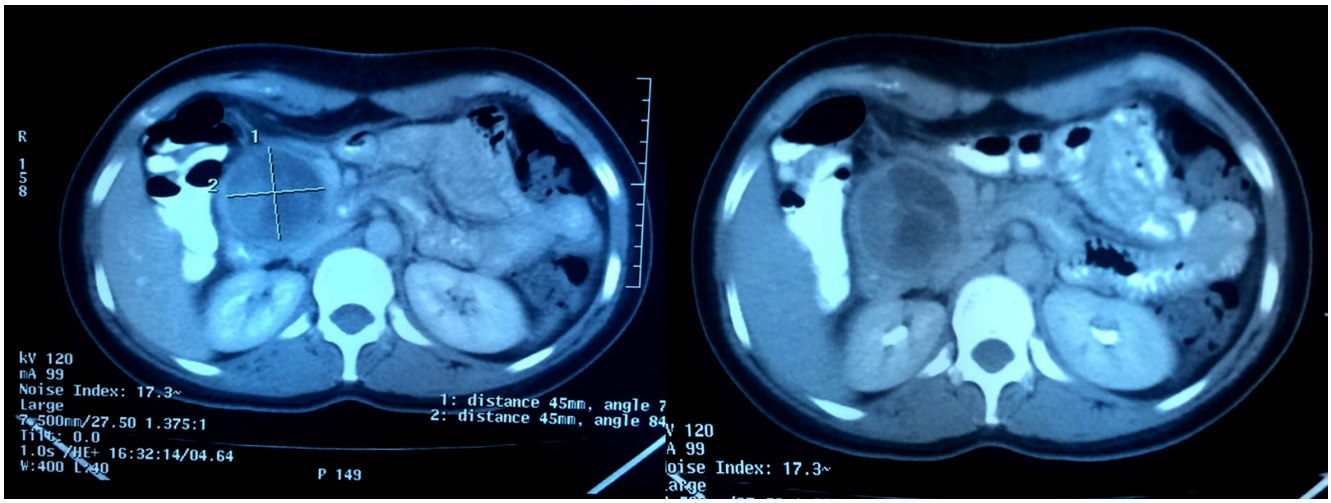

Figure 2. Triphasic abdominal computed tomography: heterogeneous mass involving the head of the pancreas, with a cystic component measuring 5 × 5.5 cm and internal areas of enhancement persisting in the equilibrium phase. Images property of the authors.

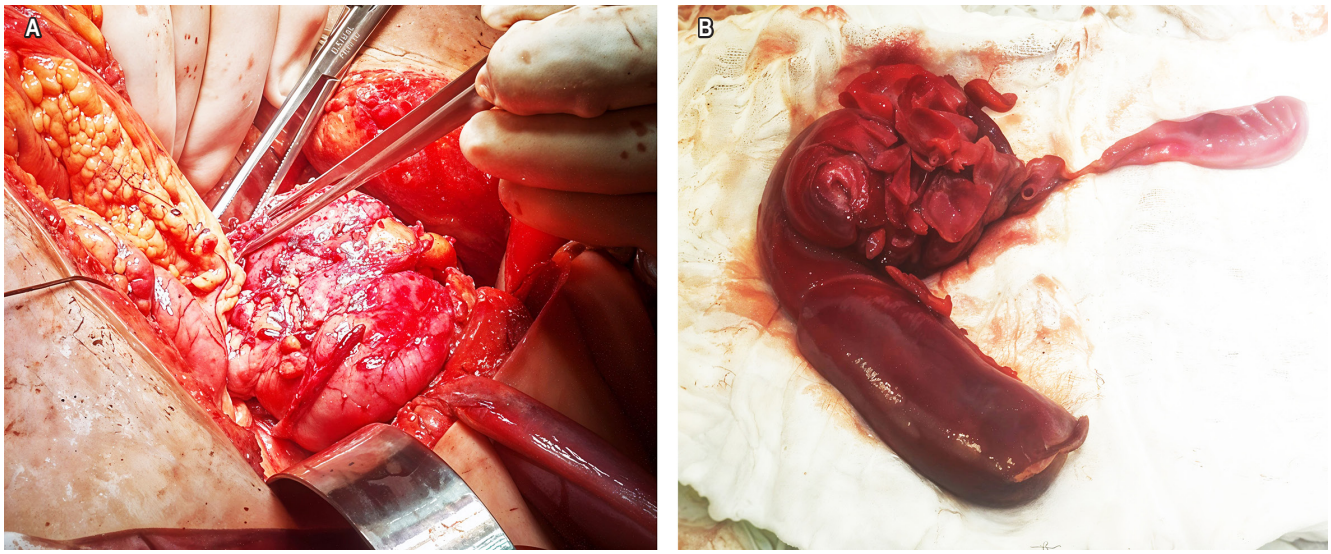

Figure 3. A. Whipple pancreatoduodenectomy. **B.** Surgical specimen of solid pseudopapillary tumor with negative margins. Images property of the authors.

tumor of the pancreas that could be benign or malignant. Subsequently, according to evolving classifications of pancreatic neoplasms, its nomenclature has changed multiple times, including solid and cystic tumor, papillary cystic tumor, papillary-cystic epithelial neoplasm, solid and papillary epithelial neoplasm, until the current designation recommended by the World Health Organization (WHO, 1996): solid pseudopapillary tumor[8].

According to anatomical distribution, this tumor occurs in the following pancreatic segments: head and neck (34%–43%), tail (31%–40%), body (14.8%–25%), and uncinate process (0.43%)[4,9,10]. It occurs more frequently in adolescent and young adult women, with a mean age of 23.9 years. Its predominance in this age group suggests a possible hormonal influence in its development. This hypothesis is supported primarily by immunohistochemical findings (positivity for progesterone and estrogen receptors) and by the close embryologic relationship between pancreatic and genital tissues during early organogenesis[11-13]. However, cases have also been described in men, with a markedly lower frequency, with a male-to-female ratio of approximately 1:10 and a prevalence ranging from 3.9% to 6.6%. Several studies have demonstrated no association between age, sex, or tumor location and degree of malignancy[4].

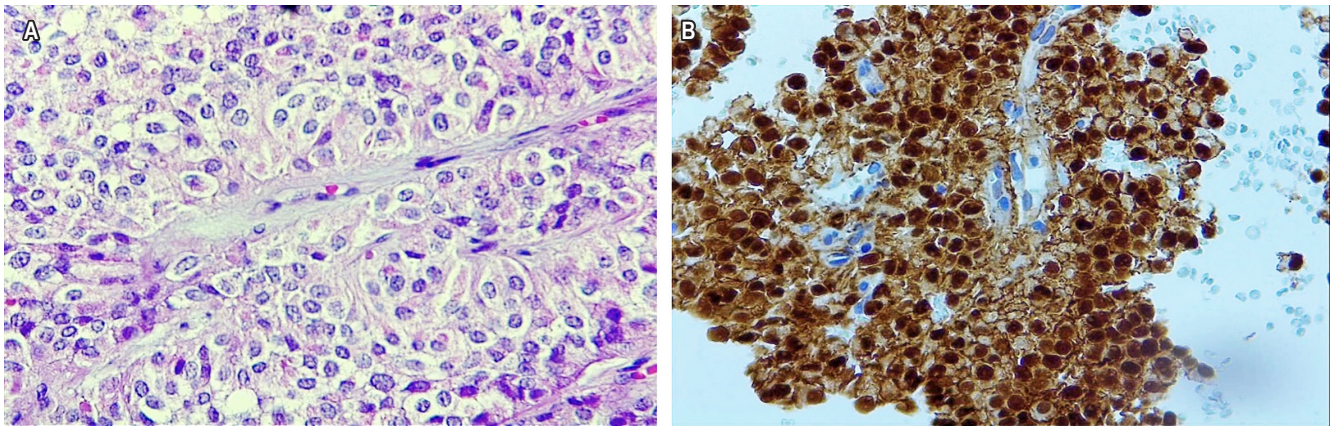

Figure 4. A. Hematoxylin and eosin staining demonstrating a neoplasm composed of monomorphic groups of polygonal cells with abundant cytoplasm and rounded nuclei with well-defined borders and fine chromatin, without mitotic figures. The scant stroma contains delicate vascular stalks. **B.** Immunohistochemistry demonstrating tumor cells with nuclear and membranous immunoreactivity for β-catenin; this staining pattern is characteristic of solid pseudopapillary tumor of the pancreas. Images property of the authors.

Although this tumor is generally benign, approximately 15% of patients may develop metastases or malignant transformation. Metastatic spread most commonly involves the liver, portal vein, spleen, lymph nodes, omentum, duodenum, colon, lung, and retroperitoneum[1,4,8,14].

Clinically, more than 50% of cases are asymptomatic, and the lesion is discovered incidentally on imaging studies. Symptoms typically occur when the tumor is located in the pancreatic head or has grown sufficiently to compress adjacent structures. Manifestations are generally vague and nonspecific. The most common symptoms include poorly localized abdominal pain, nausea, vomiting, weight loss, anorexia, and a palpable epigastric mass or sensation of gastric fullness. In some cases, progression to gastric outlet obstruction may occur. Less frequently, intestinal obstruction, anemia, jaundice (rare, associated with involvement of the pancreatic head or tail), pancreatitis, and even hemoperitoneum secondary to traumatic rupture may be observed[4,11,15].

These tumors are generally large (8–10 cm), rounded, and typically well circumscribed, with a solid, solid-cystic, or purely cystic appearance[16]. Microscopically, they are characterized by polygonal cells with abundant eosinophilic cytoplasm that do not coalesce, surrounded by small-caliber blood vessels. Degenerative changes and intracystic hemorrhage are frequently observed. Pathognomonic histological findings include papillary groups of neoplastic cells with vascular cores surrounded by mucinous stroma and an additional layer of monomorphic cells. Histological degeneration is characterized by cellular vacuolization, formation of pseudopapillae, hemorrhage, collections of foamy macrophages, cholesterol clefts and crystals, foreign-body giant cell reaction, and extensive fibrosis with focal calcifications[5,15].

Furthermore, current literature reports that tumor cells consistently demonstrate positivity for β-catenin and vimentin staining, combined with absence of E-cadherin expression. These markers are highly useful histological indicators for the diagnosis of this tumor, whereas only approximately half of cases exhibit positivity for CD10, CD56, and alpha-1 antitrypsin. Based on these findings, multiple theories have been proposed regarding the tumor's origin and associated cell lineage. Evidence suggests that it arises from pluripotent pancreatic cells and that its development is influenced by genetic and hormonal factors, particularly estrogenic and progesterone-related pathways, as demonstrated by the expression of these receptors in tumor tissue[4,11].

Diagnosis is primarily based on imaging studies, especially cross-sectional imaging modalities. Ultrasound may reveal large, solid, rounded, oblong, or multilobulated masses, typically encapsulated or heterogeneous, with both echogenic tissue components and anechoic cystic components, as well as peripheral vascularity[4,17]. Additionally, peripheral calcifications and posterior acoustic enhancement may be observed, and a mass effect on adjacent structures may be present[18]. Computed tomography (CT) is considered the diagnostic modality of choice because it allows differentiation among various types of cystic neoplasms based on location and intralesional pattern (unilocular, oligocystic, polycystic, mixed, or solid-cystic), presence of calcifications, communication with the main pancreatic duct or side branches, septations, and mural nodules[4,19]. However, CT

has limitations in visualizing and characterizing hemorrhagic and necrotic lesions, which is relevant when establishing differential diagnoses. Magnetic resonance imaging (MRI) represents an alternative imaging modality with high sensitivity and specificity. It allows improved identification of hemorrhage and facilitates assessment of resectability and surgical planning. Nevertheless, MRI is not always readily available and is associated with higher cost[6,20-22].

In these tumors, significant impairment of hepatic or pancreatic function is typically not observed, and laboratory findings are usually within normal ranges. Measurement of carcinoembryonic antigen (CEA) and carbohydrate antigen 19-9 (CA 19-9) is considered essential in patients presenting with these clinical features. Negative results help exclude alternative etiologies and support further diagnostic evaluation for this specific tumor[4].

The differential diagnosis includes any solid or cystic pancreatic disease entity, such as mucinous cystic tumor, serous microcystic adenoma, cystic adenoma, sarcoma, islet cell tumor, cystadenocarcinoma, acinar cell carcinoma, inflammatory pseudocyst, mucin-secreting tumor, angiolymphoma, pancreatoblastoma, and vascular hemangioma[8,20,23]. The first four entities typically occur in older patients and do not demonstrate sex predominance. Pancreatoblastoma is generally observed in younger individuals of both sexes. Radiologically, a characteristic sunburst pattern of linear calcification is typically observed in microcystic adenoma[8].

The treatment of choice is complete surgical tumor resection (R0), which generally involves pancreatoduodenectomy. The specific surgical approach depends on tumor size and anatomical location. When the lesion is located in the pancreatic head or neck, proximal pancreatectomy is recommended. Conversely, when the lesion is located in the body or tail, distal pancreatectomy with or without splenectomy is the preferred approach, depending on whether the tumor involves the tail or body of the pancreas[24]. Current evidence suggests that splenectomy should be avoided whenever possible, as spleen preservation is associated with fewer postoperative complications, including reduced risk of infection, shorter hospital stay, lower incidence of pancreatic fistula, and decreased morbidity and mortality[23].

Other less invasive techniques described in the literature include partial pancreatectomy and tumor enucleation. However, these approaches are less commonly used in clinical practice due to limited evidence and lower rates of complete tumor resection[4,7,25]. Enucleation has been studied in cystic or neuroendocrine lesions with favorable outcomes in some studies and has demonstrated satisfactory postoperative results, with pancreatic fistula representing the primary potential complication[7]. Nevertheless, other reports advise caution due to the potential risk of tumor dissemination and fistula formation, highlighting the need for further studies to establish its safety and efficacy[5]. Additionally, laparoscopic approaches have been explored; however, consensus has not been reached due to the limited number of reported cases and concerns regarding potential tumor dissemination[5].

Surgical management has demonstrated favorable clinical outcomes and is associated with a good prognosis. The literature reports 5-year survival rates exceeding 90%[26,27]. Although the incidence of recurrence is low, recurrence has been documented several years after treatment, particularly in patients presenting risk factors such as vascular or lymphatic invasion, synchronous metastasis, lymphatic involvement, tumors larger than 5 cm, positive surgical margins, and male sex[11,21,28-30].

Although predictors of malignancy remain controversial in the current literature, several authors have identified elevated Ki-67 index, positive surgical margins, presence of an irregular capsule, exophytic growth pattern, and large tumor size as statistically significant factors associated with increased malignant potential[29,31].

Adjuvant chemotherapy and radiotherapy are not currently standardized and evidence supporting their use remains limited. Nevertheless, these therapies are included in certain treatment protocols, particularly in cases with metastatic disease or when tumors become unresectable due to size or anatomical location[1-3,5,7,8]. Routine lymphadenectomy is not considered necessary, as lymph node metastasis is uncommon in Frantz tumor[5].

CONCLUSION

The relevance of the present case lies in the low incidence of this tumor type, which predominantly affects young female patients and demonstrates less aggressive biological behavior compared with other pancreatic lesions. Prognosis is favorable when surgical resection is performed according to oncologic standards (R0), achieving high survival rates (>90%) without the need for adjuvant therapy. Therefore, clinical suspicion in the aforementioned population without significant prior medical history is essential in order to achieve early diagnosis and timely treatment, thereby improving prognosis and reducing mortality.

Ethical considerations

Informed consent: This study was classified as a "no-risk" intervention according to Article 11 of Resolution 8430 of 1993 issued by the Colombian Ministry of Health, as it involved the use of secondary sources (review of medical records and arteriographic studies). No procedures

or interventions were performed on patients; therefore, informed consent was not required. Prior authorization was obtained from the institutional Ethics and Research Committee.

Conflicts of interest

The authors declare no conflicts of interest with any company or commercial entity.

Funding sources

The authors declare that the study was funded exclusively through their own resources. They also declare no financial relationships with pharmaceutical companies or other commercial entities.

Authors' contributions

Each author made substantial contributions to the literature search, manuscript review, adaptation, and writing of the article.

Ethical considerations

No experiments involving human subjects or animals were conducted for this research. Established protocols regarding patient data confidentiality in publications were strictly followed.

Use of artificial intelligence

No artificial intelligence tools were used.

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