

Solid pseudopapillary neoplasm of the pancreas. A clinicopathologic study of 25 cases from Pakistan and review of Literature



Nasir Ud Din, MBBS, FCPS*, Huma Arshad, MBBS, FCPS, Zubair Ahmad, MBBS, FCPS

Department of Pathology and Microbiology, Aga Khan University Hospital, Stadium Road Karachi, Karachi, Pakistan

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ABSTRACT

Our aim was to describe the clinicopathologic features of cases of solid pseudopapillary neoplasm (SPN) of the pancreas diagnosed in our section. We retrieved hematoxylin and eosin and immunohistochemistry slides of SPN of pancreas diagnosed between 2002 and 2014 and reviewed slides. A total of 25 cases were included in the study. All were females. Age ranged from 12 to 45 years with mean age of 24 years. Tail of pancreas was the commonest location (40%). Tumor size ranged from 2.5 to 20 cm (mean tumor size, 9.5 cm). Distal pancreatectomy and pancreaticoduodenectomy were the commonest types of surgical resections performed. Most cases were confined to the pancreas. However, 2 cases extended beyond the pancreas, and an additional case metastasized to the omentum and liver. Pseudopapillary architecture and hyaline globules were the commonest histologic features, seen in 100% and 84% of cases, respectively. Vimentin, cluster of differentiation 10, cluster of differentiation 56, and neuron-specific enolase were the most useful immunohistochemical stains. Of those cases, in which follow-up was available, almost all (except 1) patients were alive and well even several years after resection. In conclusion, SPN is a rare tumor in our practice. As in international studies, our study also showed predilection for young females and excellent prognosis after surgical resection.

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1. Introduction

Solid pseudopapillary neoplasm (SPN) of the pancreas is a rare pancreatic tumor, which comprises approximately 1% to 3% of all pancreatic tumors and occurs predominantly in young females [1–3]. Approximately 90% occur in females, and mean age is 28 years. It behaves mostly in a benign manner, and complete surgical resection is curative in the large majority (85%–95%) of patients. However, recurrence may occur in some cases, and a small percentage (5%–15%) of patients may develop subsequent metastases most commonly to peritoneum and liver [2,4,5]. In the past 2 decades, there has been increasing recognition of this lesion leading to a steady increase in the number of diagnosed cases. The exact pathogenesis is still not fully understood.

A recent study has postulated that SPNs may arise from centroacinar cells located between pancreatic acini and ducts [6]. Almost all SPNs show nuclear staining for β catenin (demonstrated by immunohistochemistry [IHC]) and point mutations in exon 3 of catenin (cadherin-associated protein), beta 1, the gene encoding β catenin, suggesting that the int/wingless family signalling pathway may play an important role in their tumorigenesis [7,8]. Because SPN can usually be cured by complete resection even if the mass is large and because it can in some cases invade the surrounding tissues and even metastasize, it is important that it be recognized early and definitive surgical excision be performed [9]. According to Lee et al

[10], long-term survival can be achieved (even with metastatic tumors) by aggressive treatment, and they advocate not just complete resection but even metastatectomy (in tumors with metastases). Solid pseudopapillary neoplasm is more likely to recur (and metastasize) in males [11], younger patients, and patients with larger tumors [10,12].

No previous case series on SPN has been published from Pakistan. The aim of this study is to describe the various histologic features seen in our cases together with the epidemiologic data, exact site in pancreas, tumor size, etc as well as follow-up in cases, where it was available.

2. Material and methods

Twenty-two patients with SPN of pancreas whose pancreatectomy specimens were received in the Section of Histopathology, Aga Khan University Hospital, Karachi, and an additional 3 cases in which only blocks were received for second opinion during the period 2002 to 2013 were included in the study. The hematoxylin and eosin as well as IHC slides of all cases were retrieved and reviewed by the authors. Histopathologic features including presence or absence of papillary architecture, myxoid stroma, cystic change, hemorrhage, necrosis, hyaline globules, clear cells, eosinophilic cytoplasm, grooves, mitoses, calcification, cholesterol clefts, fibrosis, etc were noted. Immunohistochemistry reactivity was also noted.

Clinical data such as age, sex, tumor location in pancreas, tumor size (in centimeters), lymph node status and type of surgery, stage, and follow-up were recorded.

Statistical analysis was performed using SPSS 19.0 version (Singapore).

* Corresponding author. Tel.: +92 3054456656.

E-mail addresses: nd176@yahoo.com (N. Ud Din), huma.arshad@aku.edu (H. Arshad), zubair.ahmad@aku.edu (Z. Ahmad).

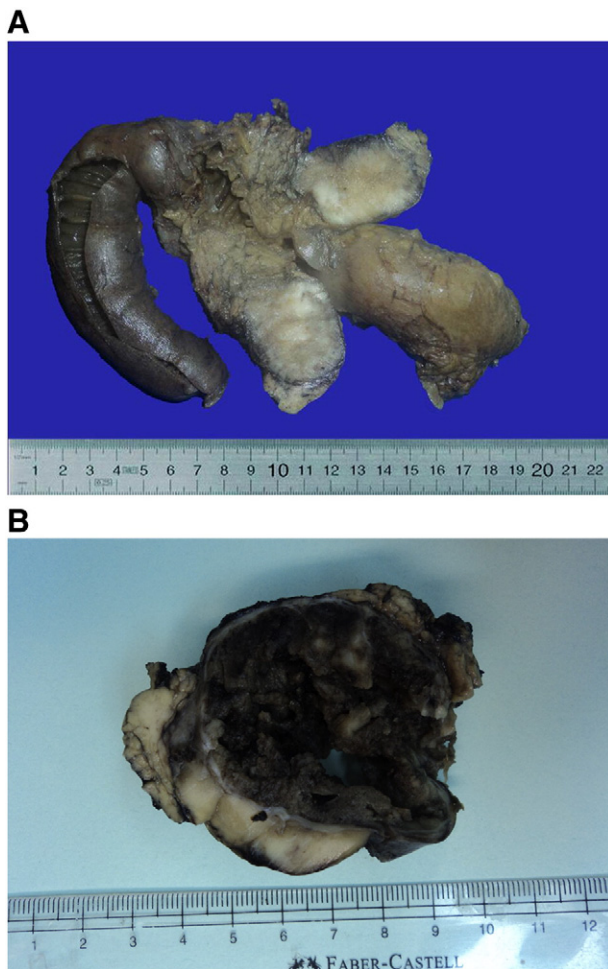


Fig. 1. Whipple resection specimen showing a circumscribed tumor in the head of pancreas with solid yellow white cut surface (A). An encapsulated tumor in the body of pancreas with hemorrhagic cut surface and cystic spaces (B).

3. Results

A total of 25 cases were included in the study. Age of the patients ranged from 12 to 45 years. Mean and median age were 24 and 25 years, respectively. All 25 patients were females. Tumors were located in the head of pancreas in 7 cases (28%), body in 4 cases (16%), and tail of the pancreas in 10 cases (40%). In 4 cases (16%), exact location could not be determined (as only blocks were received in 3 cases, whereas the tumor had extensively involved the pancreas and had tightly adhered to the duodenum and omentum resulting in distortion in 1 case). Clinical information was available in very few cases; however,

abdominal (especially epigastric) pain was the most common clinical presentation. Tumor size ranged from 2.5 to 20 cm in the largest dimension with a mean size of 9.5 cm. As mentioned above, only blocks were received in 3 cases. However, surgical resection specimens were received in the remaining 22 cases. These 22 cases included pancreaticoduodenectomy (Whipple resection) in 9 cases (40%), distal pancreatectomy in 8 cases (36.4%), distal pancreatectomy with splenectomy in 1 case (4.5%), partial pancreatectomy in 2 cases (9.1%), partial pancreatectomy with splenectomy in 1 case (4.5%), and partial pancreatectomy with splenectomy and adrenalectomy in 1 case (4.5%). Most of our cases were grossly circumscribed and encapsulated, and cut surface showed solid, cystic, hemorrhagic, and necrotic areas (Fig. 1A and B). The tumor was confined to the pancreas with all surgical margins negative in 19 (86.4%) of 22 cases. Tumor extended beyond the pancreas in 2 cases (9.1%); whereas in 1 (4.5%) of 21 cases, it riddled the omentum and liver with multiple metastatic nodules. Lymph nodes were recovered in 7 (31.8%) of 22 cases and were negative in 5 cases. Average number of lymph nodes recovered in these 6 cases was 4 per case. Only 1 case (the one with metastasis to liver) showed positive paraaortic lymph nodes.

Follow-up was available in only 11 of 25 cases. Of these 11 patients, 10 were alive and well without any apparent evidence of recurrence. The remaining patient who had developed omental and liver metastases on initial follow-up was later lost to follow-up. The details of these patients are given in Table 1.

Of the 22 cases (in which resection specimens were received), 18 (81.8%) were pT2 (tumor limited to the pancreas but >2 cm in greatest dimension), 3 (13.6%) were pT3 (tumor extending beyond the pancreas but without involvement of the celiac axis or superior mesenteric artery), and 1 (4.5%) was pT1 (tumor limited to the pancreas, ≤2 cm in greatest dimension).

Vascular and/or perineural invasion was seen in only 1 case (which metastasized to liver).

The histologic features are summarized in Table 2. Immunohistochemistry was performed in 22 of 25 cases. The antibodies used and IHC results are summarized in Table 3.

4. Discussion

In our series, tail was the commonest site for the tumor (40%), followed by head (28%) and body (16%). In a 2010 study from China, head was the commonest site (39.8%) followed by the tail (24.1%) [13]. However, a study from Korea by Lee et al [10] reported 80.9% of SPNs to be located in the body or tail of the pancreas.

Distal pancreatectomy and pancreaticoduodenectomy were the most common surgical procedures in our series. Studies by Yu et al [13] and Salvia et al [14] report similar findings.

A majority of our cases reached a considerable size yet were encapsulated and remained confined to the pancreas.

Table 1

Patients in whom follow-up was available (n = 11)

Sr no.	Age at resection	Year in which resection was performed	Pathologic stage	Current status
1	21	2014	pT2N0	Alive and well 3 months after resection
2	25	2014	pT2Nx	Alive and well 3 months after resection
3	18	2014	pT2Nx	Alive and well 9 months after resection
4	21	2013	pT3Nx	Alive and well 15 months after resection
5	17	2010	pT2Nx	Alive and well 4 years after resection
6	37	2008	pT3N0	Alive and well 6 years after resection
7	13	2008	pT2Nx	Alive and well 6 years after resection
8	23	2007	pT2Nx	Alive and well 7 years after resection
9	19	2005	pT2Nx	Alive and well 9 years after resection
10	18	2005	pT2Nx	Alive and well 9 years after resection
11	44	2004	pT3,N1,Mx	Metastatic disease on initial follow-up. Lost to follow-up later

Abbreviation: Sr, Serial.

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