Primary Squamous Cell Carcinoma of the Pancreas From a Large Cyst: A Case Report

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ABSTRACT: Pancreatic squamous cell cancer (PSCC) is a rare and aggressive form of pancreatic cancer that has a poor prognosis. The 5-year survival rate for PSCC is estimated to be approximately 10%, and the median overall survival time is 6 to 12 months. Treatment options for PSCC include surgery, chemotherapy, and radiation therapy, but the outcomes are usually not very favorable. The outcomes depend on the stage of the cancer and the patient's overall health and response to treatment. The optimal management remains early diagnosis and surgical resection. We present a rare case of PSCC with spleen invasion, which arises from a large cyst with eggshell calcification, the patient was treated by surgical resection of the tumor and adjuvant chemotherapy. This case report emphasizes the necessity of regular follow for pancreatic cyst.

KEYWORDS: Pancreas, squamous cell carcinoma, large cyst

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Background

Primary pancreatic squamous cell cancer (PSCC) is an extremely rare pathological subtype of pancreatic neoplasm, accounting for less than 1% of all pancreatic malignancies. It is considered an aggressive subtype with a worse prognosis.[1] The clinical presentation of PSCC varies, but it is believed to originate from the intrapancreatic ducts, glands, or pancreaticobiliary junction.[2] It is thought that the rarity of PSCC is due to the low rate of squamous metaplasia in the pancreatic ductal epithelium.[3]

Additionally, other factors such as immunosuppression and environmental carcinogens may contribute to the development of PSCC.[4] We report a rare PSCC arising from a large cyst as the main presentation.

Case Report

A 68-year-old Asian woman was referred from local clinics, she complained with left flank dull pain and mild tenderness about 1 month. She had been found to have a large cyst at the pancreatic tail for about 30 years, but she lost regular follow-up after several times of ultrasound examinations in the initial years. She denied having the habit of alcohol consumption or smoking, and previous medical history, including pancreatitis.

At the initial presentation, she was well looking and she denied anorexia and body weight loss in the recent months. The initial physical examination revealed nothing remarkable except minimal tenderness over the left flank. The laboratory tests disclosed hemoglobin of 12.5 g/dL, white blood cell count of 6.75×10^9/L, platelet of235 000/μL, D-Dimer of 1150 ng/mL, and serum creatinine of 0.9 mg/dL. Other laboratory biochemistries were unremarkable and within normal value. The chest plain film showed a calcified-rim mass in the left upper abdomen (Figure 1A). The abdominal ultrasound showed a heterogeneous mass at the spleen and a large cystic lesion with a calcified rim at the left abdomen. The contrast-enhanced abdominal computed tomography (CT) revealed a heterogeneously enhanced hypodense lesion measuring 9.5 cm × 8.1 cm × 5.9 cm at the spleen and a large cyst with eggshell calcification measuring 6.4 cm × 6.2 cm × 6.2 cm at pancreas tail, without metastatic lesion (Figure 1B).

After a discussion with the general surgeon, the patient underwent a laparoscopic partial pancreatectomy with splenectomy 1 week later. Laparoscopic finding show a mass at the pancreatic tail measuring 8.0 cm × 7.0 cm × 6.1 cm, involved in the spleen. The operation was successful, with no special challenges. Laparoscopic removal of a pancreas tail tumor invading the spleen, as shown in Figure 2, was performed without any difficulty. The active follow-up plan for this patient is to have a CT scan every 6 months. The pathological report disclosed that the tumor originated from the cystic wall and invaded peripancreatic soft tissues and the spleen. (Figure 3A and B) The tumor was composed of squamous cell carcinoma with keratinization. (Figure 3C) Moreover, the tumor's morphological appearances were considered moderately differentiated squamous cell carcinoma, confirmed with strong diffuse immunostaining with P40 and patchy positive of P16. (Figure 3D) The tumor had lymphovascular invasion and perineural invasion. However, no regional lymph node or resection margins were involved, and the closest margin from peri-pancreatic soft tissue was 2 mm. The postoperative tumor marker showed CEA 2.57 ng/mL,

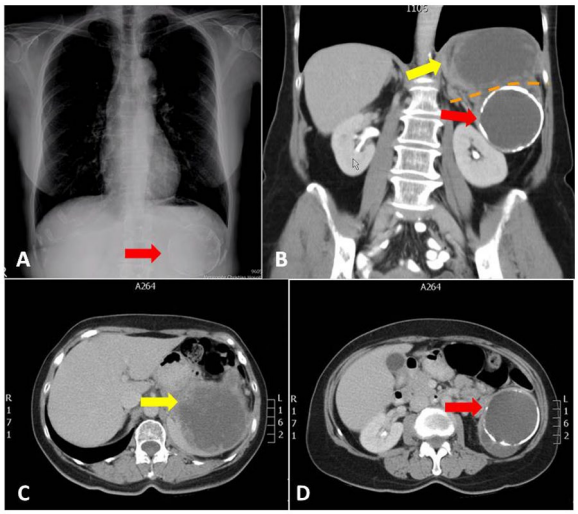

Figure 1. Chest x-ray (A) and computed tomography scans (B, C, and D) revealed an eggshell cystic lesion in the pancreas tail (yellow arrow) and a heterogeneous mass in the spleen (red arrow).

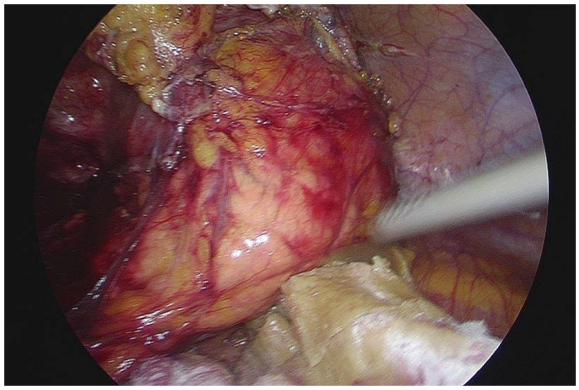

Figure 2. Laparoscopic removal of a pancreas tail tumor invading the spleen.

CA 19-9 85.27 U/mL, and no other occult potential primary site was visible. After the successful operation, the patient received adjuvant chemotherapy with Gemcitabine 1000 mg/m^2 i.v. plus Cisplatin 35 mg/m^2 i.v. on D1, D8, and D15 at 4-week interval for 6 cycles. No tumor recurrence was observed in contrast-enhanced computed tomography during the 1-year follow-up period until December 2022.

Discussion

The first case of PSCC was described by Lowry in 1949,[5] then sparse case series was reported, most in an advanced stage or metastasis.[6] Squamous cells are not present in the normal pancreas; hence the pathogenesis of PSCC remains controversial. It is thought to develop from a cyst that is present in the pancreas, although the exact mechanism is not yet understood. It is thought that the cyst may be caused by chronic inflammation, which then progresses to squamous cell carcinoma. In our case, the cyst may become malignant and develop into squamous cell carcinoma.

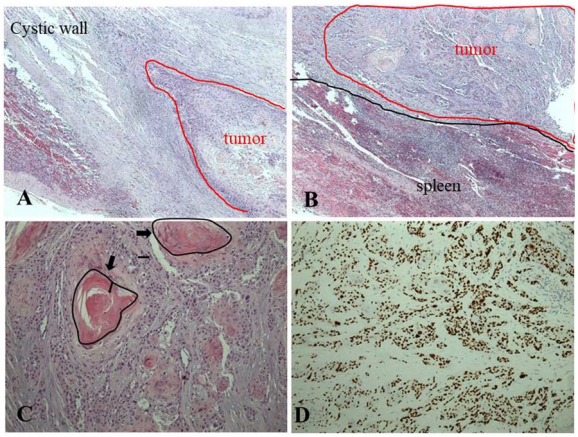

Figure 3. Pathological findings of the tumor. (A) Squamous cell carcinoma in the wall of the cystic lesion in the pancreas tail (hematoxylin and eosin, × 200); (B) Spleen invaded by the tumor (hematoxylin and eosin, × 200); (C) Squamous cell carcinoma with keratinization (black arrow) (hematoxylin and eosin, × 200); (D) Tumor cells are P40 positive (p40 immunohistochemistry, × 200).

The risk factors for pancreatic squamous cell carcinoma (PSCC) are not well-defined.[3] However, some factors have been associated with an increased risk of developing pancreatic SCC. These include smoking, obesity, a family history of pancreatic cancer, and a history of pancreatitis. Other factors such as diabetes mellitus and alcohol consumption may also increase the risk. Additionally, individuals with very fair skin, light-colored eyes, and red or blond hair are more likely to develop the disease.

The typical presentations of PSCC include epigastric pain (77.8%) and body weight loss (57.4%) in Ioannis's review.[5] The median overall survival of PSCC is lower than pancreatic adenocarcinoma (8.67 months vs 13.93 months).[7] The effective treatment of PSCC is curative resection in the early stage. There is no substantial evidence about postoperative adjuvant therapy, including chemotherapy or radiotherapy. However, 75%–80% of patients with PSCC were stage III and IV disease at diagnosis. Patients with advanced PSCC usually have an unsatisfied outcome with a median survival period of 3 to 4 months,[8] Unlike pancreatic adenocarcinoma, there are only limited case series and case reports about PSCC, so no optimal treatment guideline was established till date.

In Tella et al 's study,[6] the largest published retrospective study design, analyzed 515 cases of PSCC 2004-2015 from National Cancer Database in U.S. In stage I/II disease, surgical group had better outcome than non-surgical group, irrespective of chemotherapy or radiotherapy (21vs 5 months, $P < .01$). and postoperative adjuvant therapy show no significant difference in OS (21vs 24 months, $P = .21$).

In stage III disease, there was no significant OS between surgical group or other treatment modality group. In stage IV unresectable disease, better OS was noticed in patient who receive chemoradiation (4.9 months), compared to patient who not receive chemoradiation (1.5 month, $P < .01$).

Conclusion

PSCC is a rare neoplasm with a poor prognosis. It commonly presents with advanced or metastatic disease. Sometimes it may originate from the pancreatic cyst, as the presentation of our patient. To ensure that a pancreatic cyst is not growing or changing, regular follow-up imaging is recommended. Follow-up imaging can be done with CT or magnetic resonance imaging. This case report emphasizes the necessity of regular follow for pancreatic cyst. Regular follow of the pancreatic cyst is vital for early diagnosis, while surgical resection is the most effective treatment in resectable PSCC.

Author contributions

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