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Curative distal pancreatectomy in patients with acinar cell carcinoma of pancreas diagnosed by endoscopic aspiration via esophago-jejunostomy: A successful case report

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ABSTRACT

INTRODUCTION: This is a case report on the advances in preoperative endoscopic-guided fine-needle-aspiration (FNA) diagnosis for pancreatic carcinoma to achieve a curative operation even in patients who have a history of total gastrectomy.
CASE PRESENTATION: A 65-year-old man, who underwent total gastrectomy for gastric cancer 13 years ago, had discomfort in the left lateral abdomen. A 3-cm hypovascular mass accompanying a large distal pseudocyst in the pancreatic tail was observed on computed tomography. Endoscopic ultrasonography via elevation of the jejunal loop on esophago-jejunostomy also revealed similar lesions, and FNA for the proximal-side hypoechoic mass was successful. The cytological diagnosis with immunohistochemistry was acinar cell carcinoma of the pancreas. Distal pancreatectomy with splenectomy was successfully performed. Histology of the resected specimen also showed the acinar cell carcinoma, similar with pre-operative cytology, which involved the splenic vein and had extra-pancreatic extension but no lymph node metastasis. The tumor stage was IIA by the 2009 UICC classification. He had no tumor relapse on imaging follow-up until 12 months after the operation.
DISCUSSION: There have been marked technical advancements in endoscopic ultrasonography-guided diagnosis, including FNA, even in patients with prior digestive tract surgery. However, the risk of complication is still a concern. Accurate histological diagnosis is useful in the field of pancreatic surgery, especially in cases of rare or small malignant lesions.
CONCLUSION: Curative pancreatectomy was possible in a case of acinar cell carcinoma, a rare pancreatic malignancy, which was diagnosed by preoperative endoscopic FNA diagnosis via esophago-jejunostomy after previous total gastrectomy.

1. Introduction

Acinar cell carcinoma of the pancreas (ACC) is relatively rare malignancy representing approximately 1% in the pancreatic malignancies [1,2], and majority occur in the pancreatic head [3]. ACC was often found as a large pancreatic mass lesion with symtomps and the preoperatively accurate diagnosis by only abdominal imaging examinations might be difficult [4]. Complete pancreatectomy is required to obtain possibility for longer survival and ACC has been usually diagnosed by the resected specimen [5]. On the other hand, the multiple primary malignancies are often found since the cancer-survivors by effective treatments has been increased nowadays [6] and the prior abdominal surgery may affect the difficulty of preoperative diagnosis and operation itself by adhesion or complicated intestinal reconstructions. We herein report a case undergoing curative pancreatectomy for ACC who had a prior total gastrectomy and could be histologically diagnosed by the preoperative endoscopic examinations.

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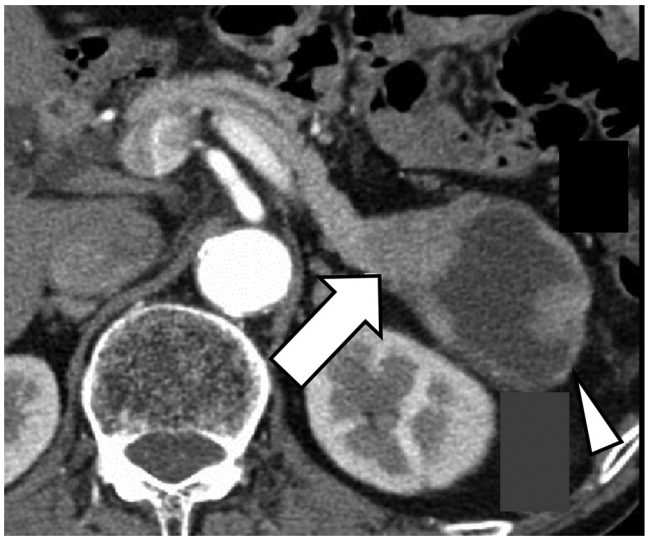

Fig. 1. Arterial-portal phase of the contrast CT. (a) White arrow showed a hypovascular irregular mass of pancreas; white arrowhead showed cystic lesion adjacent to solid mass revealed hematoma at the pancreatic tail.

2. Case presentation

A 65-year-old man had a discomfort on the left lateral abdomen and the pancreatic mass lesion was suspected. In the past history, he underwent total gastrectomy for gastric cancer and the esophagojejunostomy reconstruction was performed 13 year ago and had no tumor relapse so far. He also underwent right mastectomy for male breast cancer one year ago and, therefore, this patient had triple cancer but no history of environmental episodes in occupation. Family history did not reveal history of pancreatic cancer. He was referred to the our department for further examination and surgical treatment.

Laboratory data showed a hyperglycemia otherwise normal. Tumor markers were limited in normal range as; carcinoembryonic antigen was 5.9 ng/mL, CA19-9 12.6 U/mL, DUPAN2 170 U/mL, SPAN-1 35 U/mL and squamous cell carcinoma antigen 1.1 ng/mL

A physical examination showed no abnormalities but only the operative scar. Abdominal computed tomography (CT) using contrast media revealed a 3cm-in-size of hypovascular mass lesion accompanied with a 6.5cm-in-size of pseudocyst at the distal side in the pancreatic tail (Fig. 1). Endoscopic ultrasonography (EUS) also showed the pancreatic lesions, which were detected via elevated jejunal loop of esophago-jejunostomy. The 3cm-in-size of irregular hypoechoic mass lesion was observed in the pancreas tail and the pancreatic cancer was suspected and the 6.5 cm-in-size of cystic mass lesion adjacent to solid mass was also detected (Fig. 2). As it seemed to be closed to jejunal loop, the fine needle aspiration (FNA) was attempted for hypoechoic solid mass lesion at the proximal-side (Fig. 2). The enough specimen could be obtained and cytological diagnosis showed acinar cell carcinoma of the pancreas, which showed a plenty of cells with tubular proliferation of eosinophilic or basophilic cuboidal tumor cells with increased oval nuclei with high nuclear/cytoplasm (N/C) ratio, hyper-chromatin condensation and scattered mitosis (Fig. 3a and b). Immunohistocheistry examination revealed the positive expression of BCL10 and trypsin at cytoplasmic membrane (Fig. 3c and d), and negative expression of synaptophysin and chromogranin A (as markers for neuroendocrine neoplasm). By these results, the solid mass was diagnosed as the acinar cell carcinomas (ACC) of pancreas. Extension of pancreas cancer, node metastasis and distant metastasis were not remarkable by CT and magnetic resonance (MR). The positron-emission tomography (PET) showed high accumulation of 18-fluorodeoxy-glucose (FDG) at the solid mass lesion (Fig. 4), otherwise no accumulation at cystic pancreatic mass and other lesions systemically.

The curative operation was supposed to be possible and the distal pancreatectomy was scheduled. Under the upper median incision along the operative scar, we dissected the postoperative severe adhesion and the pancreatic body and tail could be isolated from the mesentery of jejunal loop without injury of intestine and its vessels. After ligation of splenic artery, the pancreas body was transected at the site of superior mesenteric vein. Blood loss was 480 mL and operating time was 329 min. The distal pancreatectomy with splenectomy and surrounding node dissections were successfully accomplished without any trouble during operation. However, hypovolemic shock with anemia was observed at day 1 and the blood coagulation test revealed the suspicious thrombosis at day 7. The general conditions and blood test results gradually improved until day 20. He was transferred to the follow-up hospital.

The pathological diagnosis of resected specimen showed a 2.7 cm-in-size of nodular mass lesion and adjacent cystic lesion (Fig. 5) and the cystic lesion was just pseudocyst. The solid mass also revealed ACC as well as specimen by FNA, and the tumor component was observed in the main pancreatic duct and necrosis was not focally. Atypical epithelial cells with hyperchromatic nuclei, notable nucleoid and amphophilic to eosinophilic granular cytoplasm proliferate in acinar pattern. Partly, nest-like and trabecular patterns were noted. (Fig. 6a and b). Immunohistochemitry showed cancer cells with positive BCL-10, beta-catenin, trypsin, CK7 but negative for synaptophysin, CK8 and CK20. Cytoplasm showed the Periodic acid-Schiff stain (PAS)-positive granules. Acinar cell cancer was considered, eventually. The cancer extended to the surface of pancreatic parenchyma and adjacent splenic vein but no node

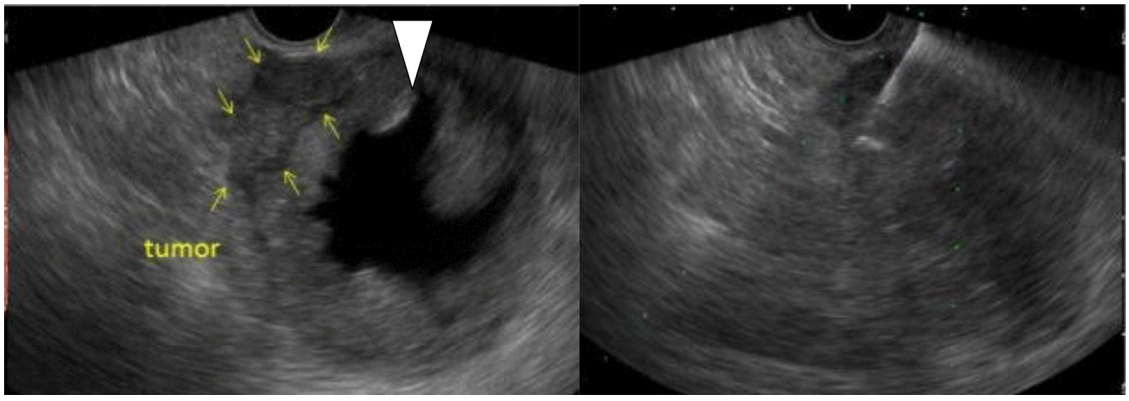

Fig. 2. EUS guided FNA. Arrow was 3cm-in-size of solid mass and arrow head was cystic lesion with thick wall.

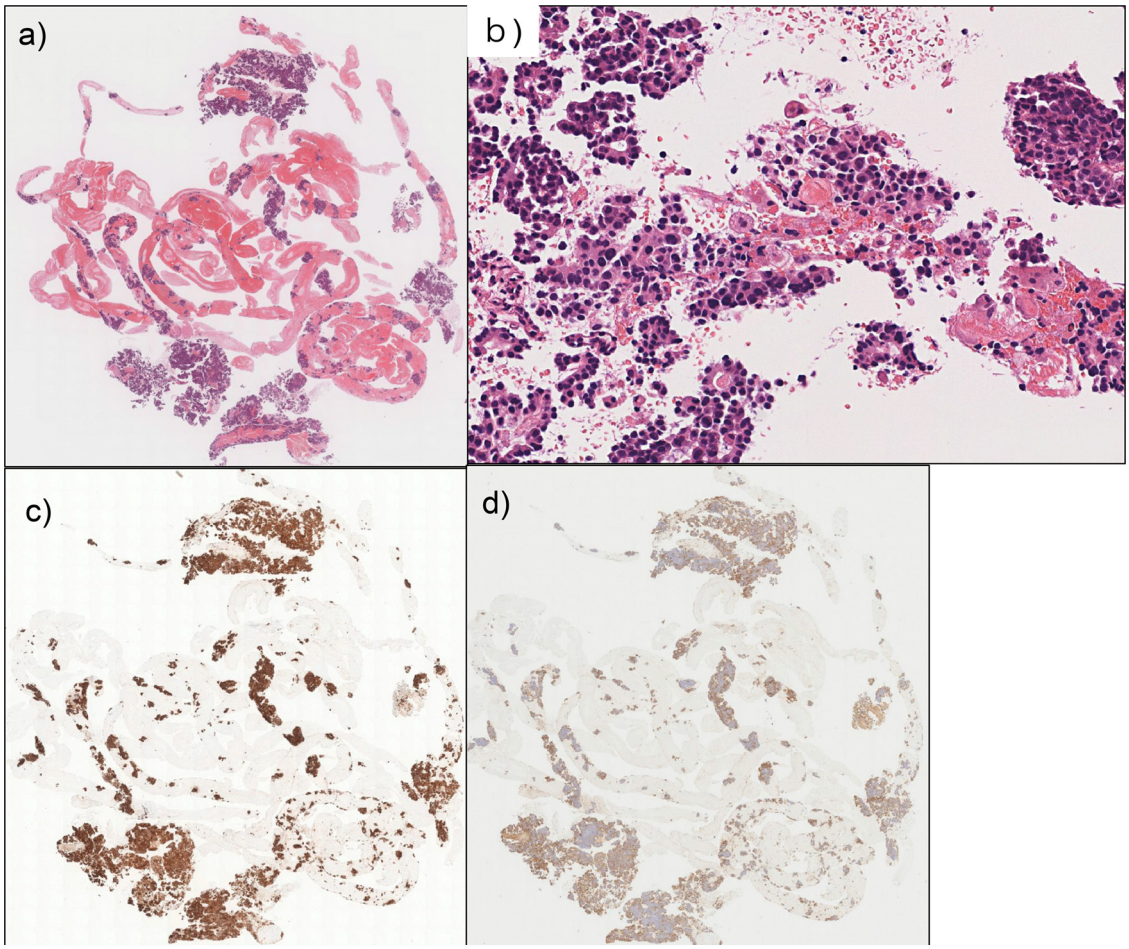

Fig. 3. Histological findings of specimens by EUS-FNA. (a) Hematoxylin-Eosin stain of weak expansion image, (b) Strong expansion; acinar, diffuse, or tubular proliferation of eosinophilic or basophilic cuboidal tumor cells with increased N/C ratio, (c) Immunohistochemitry. Positive stain of BCL-10, (d) Positive stain of trypsin.

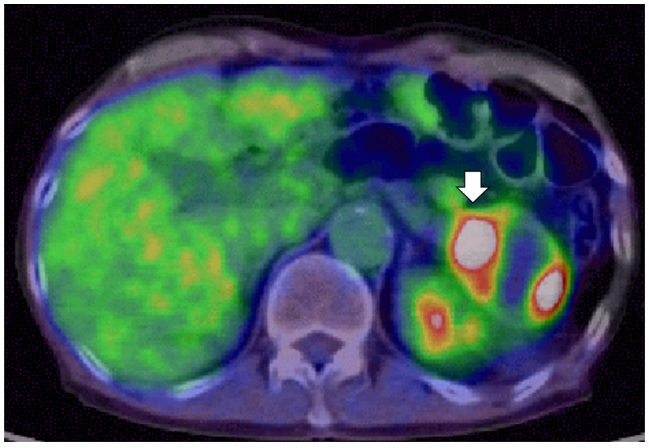

Fig. 4. At 1 h after administration of 18-FDG PET. Arrow showed an accumulation according to a solid mass lesion but no significant accumulation was observed at other lesions.

metastasis were observed. Therefore, The final diagnosis showed T3N0M0, Stage IIA by the UICC classification 2009 [12]. R0 operation was achieved.

The patient did not undergo the postoperative adjuvant chemotherapy because of patient's request and postoperative coarse was uneventful. He had no tumor relapse by the imaging follow-up until 12 months after operation.

3. Discussion

Characteristics of ACC has been well understood by some systemic review worldwide and the effectiveness of preoperative diagnosis and surgical treatments were clarified at this stage [7–9]. ACC patients may have non-specific symptoms or laboratory data [10], and the present case had a discomfort at the site of mass lesion. This symptom would be due to adjacent pseudocyst of the pancreatic duct dilatation causing the ductal obstruction by ACC. Although the radiological imaging exam sometimes show the characteristics of ACC [11], the accurate differential diagnosis with pancreatic ductal adenocarcinoma is still difficult. In the present case, the latest CT, MR or PET only showed the malignant mass lesion. However, these modalities were useful to follow-up after the prior total gastrectomy for gastric cancer and the second mastectomy for breast cancer. In the field of pancreatic malignancies, the multiple primary cancer was found in the second author's previous reports and the patient prognosis was better than the solitary pancreatic cancer patients [12,13]. This reason was supposed to be related to the more tendency of earlier stage of cancer and we are not sure whether this was biological or genetic characteristics or not in the multiple cancers. Another speculation was the careful follow-up by imaging modalities after the first surgery described above. In the present case, the careful follow-up for 13 years after gastrectomy has been continued and the present ACC might be found at the earlier stage just after a mild symptom of discomfort.

Diagnostic accuracy and technical improvement of EUS and EUS-FNA for digestive tract malignancies including the field of

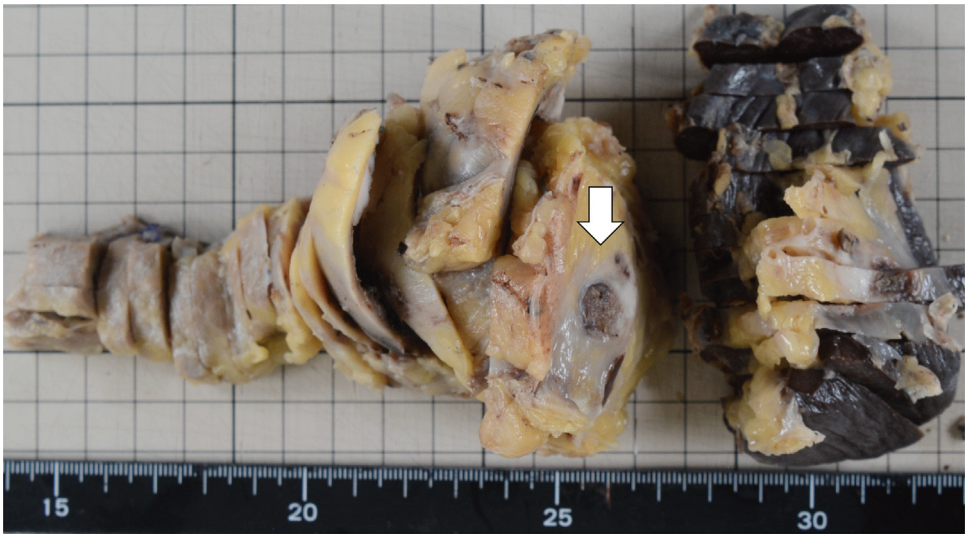

Fig. 5. Formalin-fixed resected specimen. Arrow showed a 3cm-in-size of solid mass lesion at the pancreatic tail.

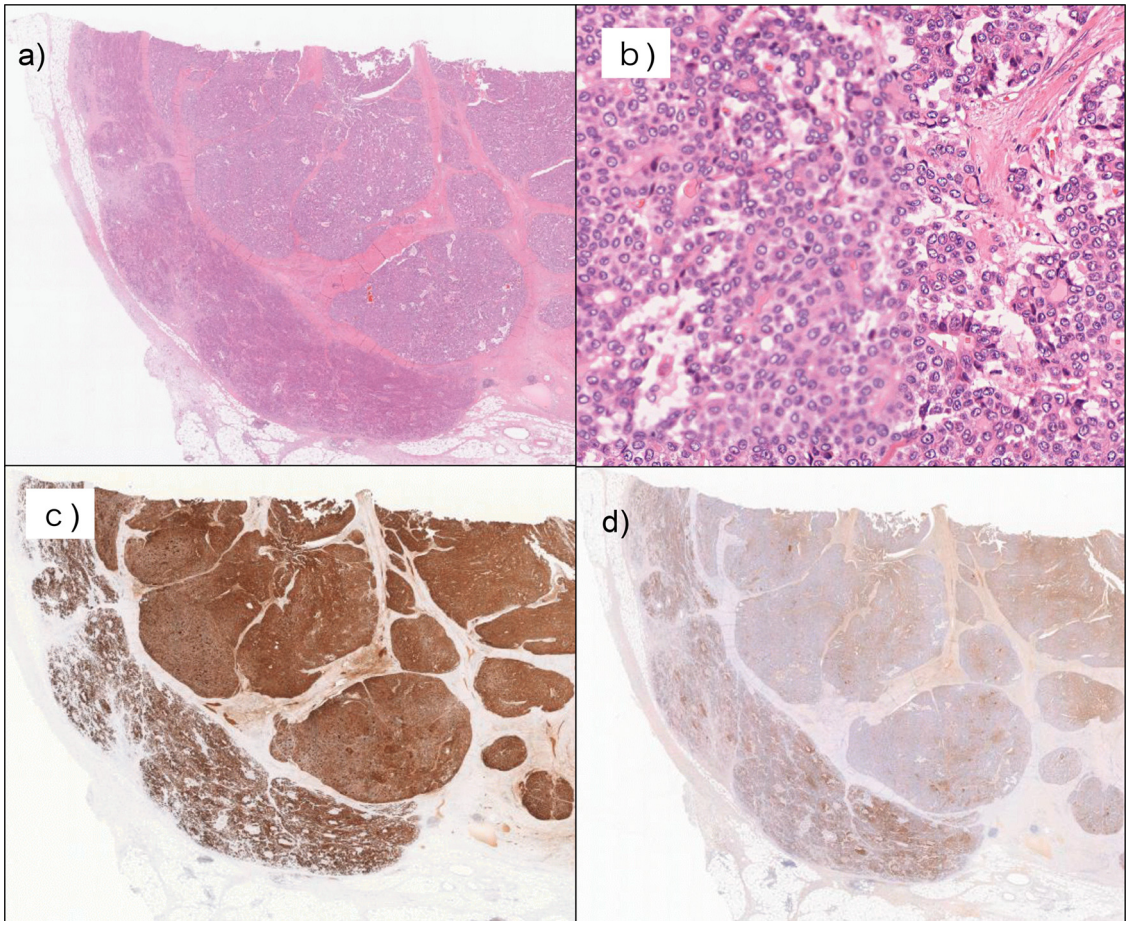

Fig. 6. Histological findings of resected specimens. (a) Hematoxylin-Eosin stain of weak expansion image, (b) Strong expansion; Atypical epithelial cells with hyperchromatic nuclei, notable nucleoid and amphophilic to eosinophilic granular cytoplasm proliferate in acinar pattern, (c) Immunohistochemitry. Positive stain of BCL-10, (d) Positive stain of trypsin as well as FNA specimen.

hepato-biliary-pancreas cancers has been well known in the recent years [14]. Histological diagnosis for ACC by EUS-FNA has been fully evaluated [15] and, on the other hand, a risk of tumor seeding of pancreatic cancer to the stomach, particularly in the body or tail of the pancreas [16]. Immunohistochemistry examinations using the aspiration biopsy is a useful adjunct diagnostic modality, especially the rare disease as the present ACC or neuroendocrine neoplasm [2,17]. In the present case, access route of FNA is the first hurdle to achieve safely because of the prior gastrectomy and limited route as esophago-jejunostomy. As the FNA via jejunal loop was supposed to be risky of perforation and seeding, we needed the accurate diagnosis for solid mass lesion to schedule

the sequentially curative operation. Our recent strategy in pancreatic malignancy is to obtain preoperatively histological diagnosis by FNA to avoid miss-diagnosis with benign diseases as mass-forming pancreatitis. Eventually, histological diagnosis of ACC was achieved and the patient course was uneventful up to operation. The interval between cytology and operation was a few weeks in this case. To our knowledge, EUS-FNA for pancreas malignancy, particularly ACC via jejunal loop has not been reported yet.

Curative pancreatectomy could be performed less than half of all ACC patients [3,7] and curability and tumor stage were influenced to patient prognosis although the tumor recurrence rate was still high [18]. In the present case, the problematic issue regarding operation was influences of prior operation and reconstruction procedure and we attempted to avoid injury of jejunal loop and its mesenteric vascularization. By careful dissection between remained loop and pancreas, we could achieve the adequate dissection without tumor expose of the pancreatic tail although the tumor location was near this mesentery. As the diagnosis of ACC was revealed, node dissection around the common hepatic artery and splenic artery, and the left-side dissection of nerve plexuses of superior mesenteric artery were fully performed. As a result, node metastasis was not observed by the final histological evaluations and the curative distal pancreatectomy could be successfully undergone. As the adequate adjuvant chemotherapy or its regimen for ACC may not been unproven with enough evidences [7,9], or node metastasis was not observed, the adjuvant chemotherapy has not been given for the present case. The tumor-relapse of this patient was not observed by the careful imaging managements for 12 months at a moment. Furthermore, the new multiple carcinogenesis at other organs, as well as detecting tumor relapse of ACC and breast cancer, were necessary by the multi-modality of diagnostic tools for long-period of his life.

4. Conclusion

Curative pancreatectomy and good operative course could be obtained in patients with rare pancreatic malignancy, acinar cell carcinoma, which was diagnosed by the preoperative endoscopic FNA diagnosis via the esophago-jejunostomy after previous total gastrectomy. To our knowledge, such a case report using multimodal diagnosis, histology and surgical treatments for ACC has not been reported yet.

Conflict of interest

Not applicable.

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There is no funding for this manuscript.

Ethical approval

In our facility, no approval of ethics for this paper was necessary.

Consent

The Ethics Committee at our institute does not require approval of submission and, however, the written, informed consent for case reports was obtained from a patient.

Submission declaration

The authors declare that the work described has not been published previously, that it is not under consideration for publication elsewhere, that its publication has been approved by all authors and either tacitly or explicitly by the responsible authorities where the work was carried out, and that, if accepted, it will not be published elsewhere, including electronically in the same form in English or any other language, without the written consent of the copyright holder. The present work has been reported in line with the SCARE criteria [19].

Author contributors

T.H., A.N., M.H., N.I., K.Y., and Y.F. equally contributed to operation and patient peri-operative management.

Y.K., T.B., and H.K. contributed to EUS-FNA.

Y.S. contributed the pathological diagnosis.

A.N. was the Chairman and Director of the Department and the instructed assistant operator for T.H. All authors approved the final version of the manuscript to be submitted.

Conceived and designed the experiments: TH AN. Performed the experiments: TH AN. Analyzed the data: TH AN TN. Contributed reagents/materials/analysis tools: TH AN KY YS MH NI ST YT ST YF TN. Wrote the paper: TH AN. Gave many suggestions in the formation of the manuscript: AN ST YF TN. All authors have read and approved the final manuscript, and ensure that this is the case.

Guarantor

Takeomi Hamada, Atsushi Nanashima

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