

A rare case of peripancreatic Castleman's disease diagnosed preoperatively by endoscopic ultrasound-guided fine needle aspiration

Castleman's disease is a rare lymphoproliferative disorder of unknown etiology and has three pathologic variants: hyaline-vascular (HV), plasma cell (PC), and mixed type [1]. Two clinically relevant subtypes exist, the unicentric and multicentric variants. Unicentric pancreatic Castleman's disease is very rare with only 13 reported cases [2–14]. We report on the endosonographic features of pancreatic Castleman's disease and its preoperative diagnosis by endoscopic ultrasound-guided fine needle aspiration (EUS-FNA).

A 27-year-old woman had a 4.2 × 4.3 cm, exophytic, hypervascular mass on arterial phase computed tomography (CT) (▶ Fig. 1), which washed out on the venous phase images.

EUS revealed a hypoechoic and hypervascular peripancreatic mass with a punctate calcification (▶ Fig. 2).

EUS-FNA was carried out and cytologic examination with flow cytometric analysis revealed polymorphous lymphocytes with a predominance of B lymphocytes, comprising a mixture of κ- and λ-bearing cells. Occasional morphologic features on cytologic smears (presence of variably sized and partially intact lymphoid follicles with traversing capillary vessels) and on cell block section (numerous lymphoid follicles with characteristic concentric rimming of lymphocytes) were compatible with a diagnosis of Castleman's disease (▶ Fig. 3).

Laparotomy with excision of pancreatic mass was carried out without the need for pancreatic resection. Pathologic examination was consistent with an enlarged lymph node. The overall nodal architecture was preserved and there were numerous small follicles. Many of the follicles were atretic, and some contained two or more germinal centers. There was prominent concentric layering of peripheral lymphocytes around the follicles, creating an onion-skin pattern (▶ Fig. 4). Hyaline deposits were present in many of the follicles. Interfollicular vascular proliferation was present, with some of the vessels penetrating the follicles, creating "lollipop" lesions. These characteristic pathologic features were consistent with

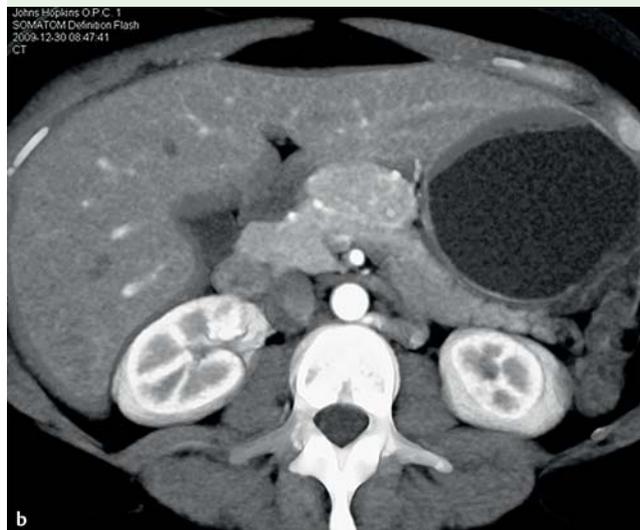


Fig. 1 Dual-phase computed tomography (CT) angiogram with three-dimensional (3D) mapping demonstrating a hypervascular mass arising from near the neck/body of the pancreas with increased vascularity: **a** maximum intensity projection and **b** volume rendered arterial phase images. The focus of calcification was also defined.



Fig. 2 Endoscopic ultrasound (EUS) showing a 41.6 × 25.1 mm, hypoechoic, homogeneous, and round peripancreatic mass with sharp borders. **a** The mass had a punctate calcification (arrow) with posterior shadowing.



Fig. 2 b EUS-guided fine needle aspiration (FNA) of the mass showed hypervascularity with power flow.

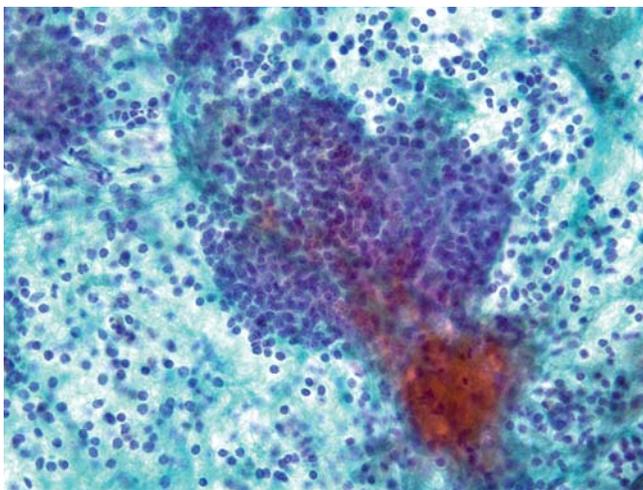


Fig. 3 Fine needle aspiration cytology shows a partially intact lymphoid follicle with a piercing fine capillary vessel. Background has polymorphous lymphocytes (Papanicolaou stain, $\times 200$).

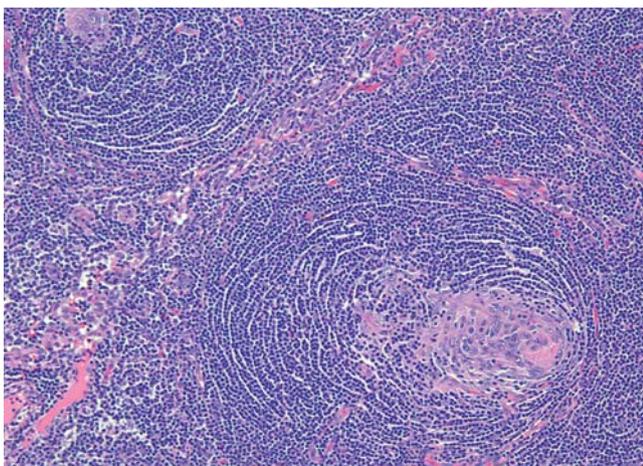


Fig. 4 Histopathologic section displaying follicles with germinal centers lined by concentric layering of lymphocytes ("onion skinning") and containing sclerotic vessels in the center (hematoxylin and eosin stain, $\times 100$).

a diagnosis of unicentric peripancreatic Castleman's disease, HV variant. In all but one of the reported pancreatic Castleman's disease cases (▶ **Table 1**) [12], the diagnosis was established after histopathologic examination of surgically resected specimens.

Rhee et al. described preoperative diagnosis of pancreatic Castleman's disease using EUS-guided trucut biopsy (TCB) [12]. However, EUS-TCB can be technically challenging, especially in the case of pancreatic head lesions, due to the stiffness of the needle. Two other reports described

nondiagnostic preoperative EUS-FNA [9, 10]. The current report is the first study to suggest a positive yield of EUS-FNA for the preoperative diagnosis of Castleman's disease. Preoperative diagnosis of Castleman's disease is important for patient reassurance, avoidance of unnecessary neoadjuvant therapy, and appropriate surgical planning. Castleman's disease should be considered in the differential diagnosis of pancreatic/peripancreatic masses. Radiographic and endosonographic characteristics of Castleman's disease are not specific and cases may display central calcifications. EUS-FNA may be a valuable tool in establishing a preoperative diagnosis.

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Competing interests: None

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Table 1 Summary of reported pancreatic/peripancreatic Castleman's disease cases.

Study	Location	Symptoms	Subtype	Treatment	EUS features	Preoperative impression/diagnosis
Lepke et al. (1982) [2]	BOP	None	HV	DP	NA	None
LeVan et al. (1989) [3]	TOP	Back pain	HV	DP	NA	None
Brossard et al. (1992) [4]	TOP	Systemic	PC	DP	NA	None
Corbisier et al. (1993) [6]	Peripancreatic	Abdominal pain	HV	Excision	NA	None
Baikovas et al. (1994) [5]	Peripancreatic	None	HV	Excision	NA	None
Le Borgne et al. (1999) [7]	Uncinate	Systemic	PC	Whipple	NA	None
Erkan et al. (2004) [8]	Peripancreatic	Abdominal pain	PC	Enucleation	NA	None
Goetze et al. (2005) [9]	TOP	None	HV	DP	Hypoechoic, well circumscribed, calcified (non-diagnostic EUS-FNA)	None
Su et al. (2005) [11]	Peripancreatic	Abdominal pain	HV	Excision	NA	PNET
Wang et al. (2007) [10]	HOP	None	HV	Whipple	Not reported (non-diagnostic EUS-FNA)	None
Tunru-Dinh et al. (2007) [14]	TOP	Abdominal pain	HV	DP	NA	None
Rhee et al. (2008) [12]	Peripancreatic	None	HV	Excision	Hypoechoic, homogeneous, well-delineated, hypervascular	Castleman's disease (by EUS-trucut biopsy)
Charalabopoulos et al. (2010) [13]	BOP	Abdominal pain	PC	DP	NA	None

BOP, body of pancreas; TOP, tail of pancreas; HV, hyaline-vascular type; PC, plasma cell type; DP, distal pancreatectomy; EUS-FNA, endoscopic ultrasound fine needle aspiration; NA, not applicable; PNET, pancreatic neuroectodermal tumor.

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