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CASE REPORT

Lymphoepithelial Cyst of the Pancreas: A Case Report

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ABSTRACT

Lymphoepithelial cysts are relatively common lesions in the lateral neck region, but a lesion with identical morphology and clinical behavior may sometimes occur in the pancreas (although rare). Lymphoepithelial cysts are characterized histologically according to [illegible] mass in the pancreatic tail. Histopathological examination revealed the rare diagnosis of a pancreatic lymphoepithelial cyst. Pancreatic lymphoepithelial cyst is often diagnosed microscopically in a resected specimen, after a partial pancreatectomy performed on suspicion of a neoplastic cyst. The most clinically important differential diagnosis of lymphoepithelial cysts are mucinous neoplastic cysts of the pancreas: mucinous cystic neoplasia and intraductal papillary mucinous neoplasia demanding surgical treatment, whereas in case of asymptomatic lymphoepithelial cyst, the "watch and wait" approach should be preferred. Preoperative diagnosis of lymphoepithelial cyst remains a challenge. Reviewed is literature pertaining to clinical, cytological and histological examination.

INTRODUCTION

[illegible] lesions in the lateral neck region, most often derived from remnants of the second branchial apparatus [1]. Patients usually present with painless swelling. On gross examination, the cysts are unilocular and contain clear to grumous material. Sizes are variable and can reach 10cm. Cytological examination usually demonstrate neutrophils, [illegible] review of the relevant literature along with the differential diagnosis and clinical implications.

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Keywords Carcinoembryonic Antigen; Cysts; cytology; Goblet Cells; Pancreas; Pancreatectomy; Preoperative Period
Abbreviations [illegible]

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CLINICAL COURSE

A seventy-four-year-old female patient with a clinical history of arterial hypertension, osteoporosis and smoking, was referred to the Gastroenterology Department of the 2nd [illegible] & Faculty Hospital Kralovske Vinohrady, because of the [illegible] cystic formation in the pancreatic tail described using computed tomography (CT). The CT was originally performed to investigate cysts [illegible] by urologist. Further endosonographic examination of the pancreas revealed a hypoechogenic multi-cystic lesion on the border of the pancreatic body and tail (Figure 1) , with [illegible] septa, sediment and central module. The clinical diagnosis [illegible] needle aspiration biopsy was performed establishing [illegible] background and particles of mucus without nucleated cells [illegible] CA 19-9, CA 125, CA 72-4) were negative. The patient was recommended to go through with an additional biopsy, but she refused. As a result, she maintained regular follow-ups at the gastroenterology department and the cystic lesion was closely monitored using ultrasonography. However, over the next 10 months the size of the cyst increased for approx. 10 mm. At this point, the patient agreed to undergo a second biopsy. During the endosonograhic examination, [illegible] and sent for analysis. Biochemical analysis of the cystic

Figure 1. • † ‘ • ‘ ‰ ” ƒ ’ Š › • Š ‘ ™ ‹ • ‰ ƒ Š › ’ ‘ ‡ … Š ‘ ‰ ‡ • ‹ … • — Ž – ‹ … › • – ‹ … Ž ‡ • ‹ ‘ • ™ ‹ – Š • ‡ ’ – ƒ ƒ • † … ‡ • – ” ƒ Ž • ‘ † —

Ð Ž — ‹ † ” ‡ ˜ ‡ ƒ Ž ‡ † ‡ Ž ‡ ˜ ƒ – ‡ † Ž ‡ ˜ ‡ Ž • ‘ ^ … ƒ ‹ ” † ‡ • ‹ ‘ Ð ‹ „ ‡ † ‘ • ‹ • … – ƒ Š ‡ ‹ ‰ ‹ ‘ ‡ • ‡ ” ’ ƒ • … ” ‡ ƒ – ‹ …
± z z t Á ‰ Ž ” ‡ ^ ä ´ t ä w Á ‰ Ž ƒ • † … s { æ { s { æ { ± s z y • Ž ” ‡ ^ ä ´ u r ä { • Ž ” ‡ ^ ä ´ u r ä { • additional CT examination elucidated a slightly lobulated ^ ‘ ” • ƒ – ‹ ‘ • ™ ‹ – Š ‘ — – … ƒ Ž … ‹ Ð ‹ … ƒ – ‹ ‘ • • ‹ • – Š ‡ • „ ‘ • † › ™ ‡ ” – ‡ Š ‡ Ž ‹ ƒ ‡ † … ” ‡ ™ ƒ • Š • ‡ ” ƒ – ‹ • ‹ œ ‹ • ‰ bending cranially to the wall of stomach, while the rest of the pancreas was structurally normal without expansion of the main duct(Figure 2) . Owing to the aforementioned enlargement of the cyst, as well as the clinical suspicion ‘ ^ ƒ • — … ‹ • ‘ — • – — • ‘ ” ‡ Ž ‡ ˜ ƒ – ‹ ‘ • ‘ ^ ‹ • … › • – Ð Ž — ‹ † á — • … ‡ ” – ƒ ‹ • … ‘ • • — • ‹ . duct), a distal pancreatectomy was recommended by the multidisciplinary team at the hospital. Prior to surgery, a • Š ‘ ” – ’ ƒ • … ” ‡ ƒ – ‹ … • – ‡ • – ™ ƒ • ‹ • – ” ‘ † — … the postoperative pancreatic leak. Concurrent exploration ^ ‘ ” ƒ … ‘ • • — • ‹ … ƒ – ‹ ‘ • ™ ‹ – Š † — … – — • ‹ ” ‡ • – ‰ ‹ • ‡ … Ž ‹ • ‹ … ƒ Ž † ‹ ƒ ‰ • ‘ • ‹ • ‘ ^ ä ‡ • ‡ … – ‹ ‘ • ‰ ƒ Ž Ž ‡ „ Ž ƒ † ‡ Š ‡ ‘ • • – ” ƒ – ‡ † • ‘ • ‹ ‰ Ð – ƒ ‹ Ž ‘ ^ – Š ‡ ’ ƒ • … ” ‡ ƒ • ™ ƒ • • — „ • ‡ “ — ‡ • – Ž changes. ‘ ” • ‡ † – ‘ ‰ ‡ – Š ‡ ” with splenectomy and cholecystectomy in the General — ” ‰ ‡ ” › Ž ‹ • ‹ … ƒ – – Š ‡ u ” † ƒ … — Ž – › Hospital Kralovske Vinohrady. One month later, the pancreatic stent was endoscopically removed.

Histopathological Examination

A gross specimen measuring 92×33×20 mm, consisting of the pancreatic tail and spleen (weight 60 g) was delivered to the Department of Pathology for dissection. An encapsulated mass measuring 40×30×23 mm was

section, multiple cysts containing viscous grumous material were observed, with dirty yellowish areas in the • — ” ‘ — • † ‹ • ‰ ’ ‡ ” ‹ ’ ƒ • … ” ‡ ƒ – ‹ … ^ ƒ – – ‹ • • — epithelium without cellular atypia and focally with admixed mucinous PAS-positive goblet cells (Figure 3) . The cystic … ‘ • – ‡ • – ™ ƒ • … ‘ • ’ ” ‹ • ‡ † ‘ ^ † ‡ • “ — ƒ • ƒ – ‡ † debris and foamy macrophages. Abundant lymphoid tissue with germinal centers was found within the cyst wall, in hematoxylin-eosin stain (Figures 4, 5) . Older fat necrosis with cholesterol crystals, giant multinuclear macrophages and scarring were present in the surrounding adipose tissue. The findings were compatible with the diagnosis ‘ ^ ƒ ’ ƒ • … ” ‡ ƒ – ‹ … á ™ ‹ – Š ’ ‘ • – æ ‹ • Ð Ž ƒ

DISCUSSION

‡ † ‹ … ‹ • ‡ ¬ ƒ … — Ž – › ƒ • … ” ‡ ƒ – ‹ … ™ ƒ • Ð ‹ ” • – † ‡ • … ” ‹ „ ‡ † … Š ” ‹ ‡ ^ ‡ ” • ‹ • s { z w u ä • … ‘ • – ” ƒ • – – ‘ ” ‡ ‰ ‹ ‘ • á ‹ • ƒ ” ƒ ” ‡ † ‹ ƒ ‰ • ‘ • ‹ • ‹ • – Š ‡ case reports or small groups of patients, including recent references, have appeared in the literature [3, 4, 5, 6, 7, 8, 9, 10, 11, 12, 13, 14, 15, 16, 17, 18, 19, 20, 21, 22, 23, 24, t w á t x á t y á t z á t { á u r á u s á u t á u u á u v middle-aged patients, predominantly but not exclusively

Figure 2. CT scan after intravenous contrast media administration showing exophytic cystic lesion arising from the superior contour of the pancreatic body. – Š ƒ • —•‹Ž‘…—Ž ƒ ” ƒ ’ ’ ‡ ƒ ” ƒ • … ‡ ™ ‹ – Š •Ž ‹ ‰ Š – Ž › Ž ‘ „ — Ž ƒ – ‡ † … ‘ • – ‘ — ” • á – Š ‡ … ‘ • – ‡ • – ‹ • Ž ‘ ™ † ‡ • • ‹ – › pancreatic duct is not dilated.

Figure 3. ‹ • – ‘ ’ ƒ – Š ‘ Ž ‘ ‰ ‹ … ƒ Ž ‹ • ƒ ‰ ‡ • Š ‘ ™ ‹ • ‰ ^ ‘ … ƒ Ž ’ ” ‡ • ‡ • … ‡ ‘ ^ ‰ ‘ „ Ž ‡ – … ‡ Ž Ž • ‹ • – Š ‡ ‘ – Š ‡ ” ™ ‹ • ‡ • “ — ƒ

Figure 4. ‹ • – ‘ ’ ƒ – Š ‘ Ž ‘ ‰ ‹ … ƒ Ž • … ƒ • • Š ‘ ™ ‹ • ‰ • – ” ƒ – ‹ Ð ‹ ‡ † • “ — ƒ • ‘ — • ‡ ’ ‹ – Š ‡ Ž ‹ — • ƒ • † Ž › • ’ Š ƒ – ‹ … ‰ ‡ ” • ‹ • ƒ Ž

Figure 5. ‹ • – ‘ ’ ƒ – Š ‘ Ž ‘ ‰ ‹ … ƒ Ž • … ƒ • • Š ‘ ™ ‹ • ‰ • … › • – ‹ … … ƒ ˜ ‹ – › … ‘ • – ƒ ‹ • ‹ • ‰ – ‹ • • — ‡ † ‡ „ ” ‹ • ™ ‹ – Š ˆ ‘ ƒ • › • ƒ … ” – ‹ • • — ‡ ™ ‹ – Š • ‡ … ‘ • † ƒ ” › Ð ‹ „ ” ‘ – ‹ … … Š ƒ • ‰ ‡ • ä á z ä

‹ • • ‡ • ± v s ä Š ‡ • ‘ • – … ‘ • • ‘ • • › • ’ presentation is abdominal pain. Other complaints include nausea, vomiting, anorexia, weight loss, back pain, fatigue, ˆ ‡ ˜ ‡ ” á ƒ • † … Š ‹ Ž Ž • ä ƒ • › … ƒ • ‡ • ƒ ” ‡ † ‹ ƒ ‰ › • • $ † ‹ ‡ — ” ˆ Ž • ‰ … ƒ Ž ‡ • æ ” ” ‘ — • † ‡ † „ › ƒ • • ƒ Ž Ž ” ‘ — • † „ Ž ƒ • † Ž › • ’ Š ‘ … › – ‡ • ä • • ‘ up for other diseases.

” ‘ • • Ž › á ’ ” ‡ • ‡ • – • ƒ • ƒ • — Ž – ‹ Ž ‘ … — Ž ƒ • — • ‹ Ž … — Ž ƒ ä intrapancreatic or peripancreatic cystic mass with the • ‡ ƒ • • ‹ œ ‡ ‘ ˆ v ä z … • ” ƒ • ‰ ‡ s ä t æ s y … • situated in the tail of the pancreas, the cysts are round and Ð ‹ Ž Ž ‡ † ™ ‹ – Š ƒ … Š ‡ ‡ • › á ‰ ” ƒ • — Ž ƒ ” á › ƒ Ž Ž æ „ ƒ • • • — „ • — ƒ • : ‡ ä • ƒ ˆ ‡ ™ … ƒ • ‡ • á – Š ‡ … › • – • … ‘ • – ƒ ‹ • ‡ † – Š ‹ Ž • • ‡ ” ‘ ’ – • Š Ð Ž — ‹ • ‡ • ‹ • • – ‡ ƒ † • ‹ • • ‘ – • ‘ ™ of the keratinaceous debris [4].The inner surfaces of the cysts are granular, devoid of any prominent projections [4].

‹ … ” ‘ • … ‘ ’ ‹ … ƒ Ž Ž › á • Š ƒ ˜ ‡ ƒ • “ — ƒ • ‘ — • • Ž ‹ ä ‹ • ‰ Š ‡ ƒ ‡ † ƒ ƒ ‡ • ‡ ˜ ‡ ” ƒ Ž ’ ” ‘ ’ ‘ • ‡ † • ‡ dense subepithelial rim of lymphoid tissue [4, 5, 10, 11, 12, 20, 25, 27, 28, 29, 30, 31], the lining may also be focally attenuated. Occasionally, columnar mucinous cells or goblet cells are found [4, 12], at the luminal surface of ‘ – Š ‡ ” ™ ‹ • ‡ • “ — ƒ • ‘ — • ‡ ’ ‹ – Š ‡ Ž ‹ — • ä Š ‡ ” ‡ • ƒ • ƒ ‡ „ ‡ ’ ƒ ” ˆ • ” • ‘ ^ ‘ ^ – ‡ ” ƒ – ‘ • ƒ ‘ ” ƒ „ ” ƒ • …

foci with sebaceous differenti ƒ – ‹ ‘ • ‹ • [7, 8, 9].

Š ‡ Ž › • ’ Š ‘ ‹ † – ‹ • • — ‡ ‘ ˆ ’ ƒ • … ” ‡ ƒ – ‹ … elements ordinarily present in lymph nodes: scattered granulomas and microscopic foci of fat necrosis in the lymphoid areas or in the areas immediately adjacent to the pancreas were present [4]. older fat necrosis with cholesterol crystals and giant cell granulomas in the peripancreatic adipose tissue.

The e ‹ ‘ ’ ƒ – Š ‰ • ‡ ‡ ‹ • ‹ • • – ‡ ƒ † • ‹ • • ‘ – • ‘ ™ – Š ƒ – ƒ ” ‡ – › ’ ‹ … ƒ Ž Ž › ƒ • • ‘ … ‹ ƒ – ‡ † ™ ‹ – Š • — … Š ƒ • ƒ Š ‹ • – ‘ ” › ‘ ^ Œ Ú ‰ ” ‡ • † ‹ • ‡ ƒ • lymphoma have not been reported in cases of pancreatic – Š ‡ ’ ƒ – Š ‘ ‰ ‡ • ‹ … „ ƒ … • ‰ ” ‘ — • † ‘ ^ • — • “ — ƒ • ‘ — • • ‡ – ƒ ’ Ž ƒ • ‹ ä x ä – Š ƒ • ƒ Ž • ‘ „ from epithelial remnants in lymph nodes [5], or cystic transformation of pancreatic ducts that have undergone

that have been displaced and fused with the pancreas during embryogenesis [3]. The sebaceous glands included [illegible] 9], may support the hypothesis concerning teratomas.

[illegible] can be diagnosed preoperatively, the option of "watch and wait" may be clinically acceptable [4].

The most clinically important differential diagnosis [illegible] Another important differential diagnosis can be solid pseudopapillary neoplasia, occurring predominantly in young women. Solid pseudopapillary neoplasia is regarded as well. On the other hand, like in case of asymptomatic case of serous adenoma, a fully benign pancreatic tumor representing another important differential diagnosis. [illegible] histopathological examination of a resected specimen following partial pancreatectomy performed on suspicion of a neoplastic cyst.

The most common clinical and imaging mimicry of this is a cystic neoplasm of pancreas, either benign or malignant [illegible] There is prevailing male sex and tail-localization in [illegible] described round shape and exophytic location in the pancreatic body and tail as [illegible] test for the diagnosis of a mucinous cyst [12, 13], but it does not distinguish non-neoplastic from neoplastic [illegible] associated with increased plasmatic CA 19-9 level as well and proper cytological assessment as the only tool that can achieve a diagnosis without resection [17, 21, 22, 23]. [illegible] cells, multinucleated giant cells, mature lymphocytes on a background of keratinaceous debris, and a lack of neoplastic cells [29]. Cytological examination may aid in the correct diagnosis if tissue elements characteristic of a [illegible] tissue fragments are found [30]. However, cytologic [illegible]

Some authors do not recommend needle biopsy for cystic lesions of the pancreas because of risk of the dissemination of tumor cells or the development of pseudomyxoma [32].

[illegible] mentioned above, there are several papers describing characteristics include mosaic pattern depending on the degree of keratin amount in ultrasonography, enhancement of the wall and septa of the cyst without pancreatic duct [illegible] et al. present male gender and elevation of serum CA19-9 [illegible] were not present in our case. [illegible] biopsy of the cystic mass is recommended by some [illegible] which has been commonly performed in many previous case reports, including our case. However, Arumugam et al. state there is no reliable preoperative diagnostic method and surgical excision and pathological analysis remain the gold standard in symptomatic patients and when malignancy cannot be excluded [35].

[illegible] epidermoid cysts in intrapancreatic accessory spleens, lymphangiomas, and pseudocysts; nevertheless, after [illegible] which has been commonly performed in many previous case reports, including our case. The reliability of preoperative diagnostics remains controversial. There are several references favorizing imaging methods for cytological analysis of [illegible] fully benign lesion, often treated by partial pancreatectomy performed on suspicion of a neoplastic mucinous cyst, like in our case. The reliability of preoperative diagnostics remains controversial. There are several references favorizing imaging methods for cytological analysis of [illegible] enucleation of the cyst. However, surgical excision and histopathological analysis remain the gold standard in symptomatic patients and when malignancy cannot be excluded, like in our case.

CONCLUSION

[illegible] fully benign lesion, often treated by partial pancreatectomy performed on suspicion of a neoplastic mucinous cyst, like in our case. The reliability of preoperative diagnostics remains controversial. There are several references favorizing imaging methods for cytological analysis of [illegible] enucleation of the cyst. However, surgical excision and histopathological analysis remain the gold standard in symptomatic patients and when malignancy cannot be excluded, like in our case.

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CONFLICT OF INTEREST

[illegible]

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