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CASE REPORT

Management of a Complicated Pancreatic Pseudocyst: Report of a Case and Review of the Literature

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ABSTRACT

Case report Herein we present a case of a huge complicated pancreatic pseudocyst following an episode of acute necrotizing pancreatitis, the treatment approaches and a short review of the literature. **Methods** An on-line survey in PubMed was conducted using the terms "acute pancreatitis-complications", "pancreatic pseudocyst" and "pancreatic pseudocyst treatment". We also used the data from the file of the patient regarding his treatment. **Results** Treatment of a pancreatic pseudocyst is a challenging process requiring a close follow-up of the patient and a fully equipped institution with medical team ready to cope against any possible complications of the different therapeutic interventions including open surgery. **Conclusion** Pancreatic pseudocyst is a complication of acute or chronic pancreatitis which should be managed closely for a longstanding period and treated effectively by endoscopic and surgical means, either alone or in combination, so that the outcome of this complication to be in favor of the patient.

INTRODUCTION

Pancreatic pseudocyst is a fluid-filled cavity after acute episodes of pancreatitis that result in tissue necrosis or disruption of a pancreatic duct. According to the Revised Atlanta classification, as a pseudocyst should be characterized every acute pancreatic fluid collection that develops an enhancing capsule earlier than four week after onset of acute pancreatitis. The communication with the pancreatic ductal system is initially always present and may further remain or seal off spontaneously during the clinical course [1]. Most pseudocysts present minor symptoms and are uncomplicated. The vast majority of pseudocysts (less than 6 cm) have thin wall and usually resolve spontaneously. Large pseudocysts are often in continuity with the pancreas, and thick-walled rarely communicate with the pancreatic ductal system [2].

Most large pancreatic pseudocysts are likely to remain requiring intervention only in the presence of complications (bleeding, infection, splenic vein thrombosis etc.), and obstructive symptoms of duodenum, bile duct, or stomach. The most common complaints of the patients are early satiety, nausea and vomiting after meals [3].

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In this case we present a patient with a huge pancreatic pseudocyst complicated after endoscopic drainage procedures and finally resolved by open surgery, and also a short review of the literature of the different treatment strategies of the pancreatic pseudocysts.

CASE REPORT

A sixty-two-years-old man was referred to our hospital, 5 months after an episode of severe acute necrotizing pancreatitis in a district hospital. The patient was diagnosed with pancreatic pseudocyst, three weeks after the remission of the episode. The pseudocyst was 18 cm in diameter and was causing restrictive symptoms, mostly of early satiety and loss of weight (15 kg) during his hospital stay. The patient's history includes goiter, hyperlipidemia and chronic obstructive pulmonary disease.

On CT scan with pros and IV contrast agent there was found a large fluid collection of 18 cm in diameter with debris (hemorrhagic fluid and necrotic material) in the area of the lesser sac. Also, there was found a pancreatic necrosis of 70-80%.

In our hospital, the patient continued to present symptoms of early satiety and epigastric discomfort after meals and was severely debilitated. On a second CT scan the pancreatic fluid collection was further enlarged **(Figure 1)**. As a first-line treatment was decided the endoscopic drainage of the pseudocyst, so the patient was referred to another hospital for interventional endoscopy. The endoscopic EUS showed a cystic mass behind the stomach pressing its lumen with a large amount of debris inside. Drainage of the pseudocyst through the stomach

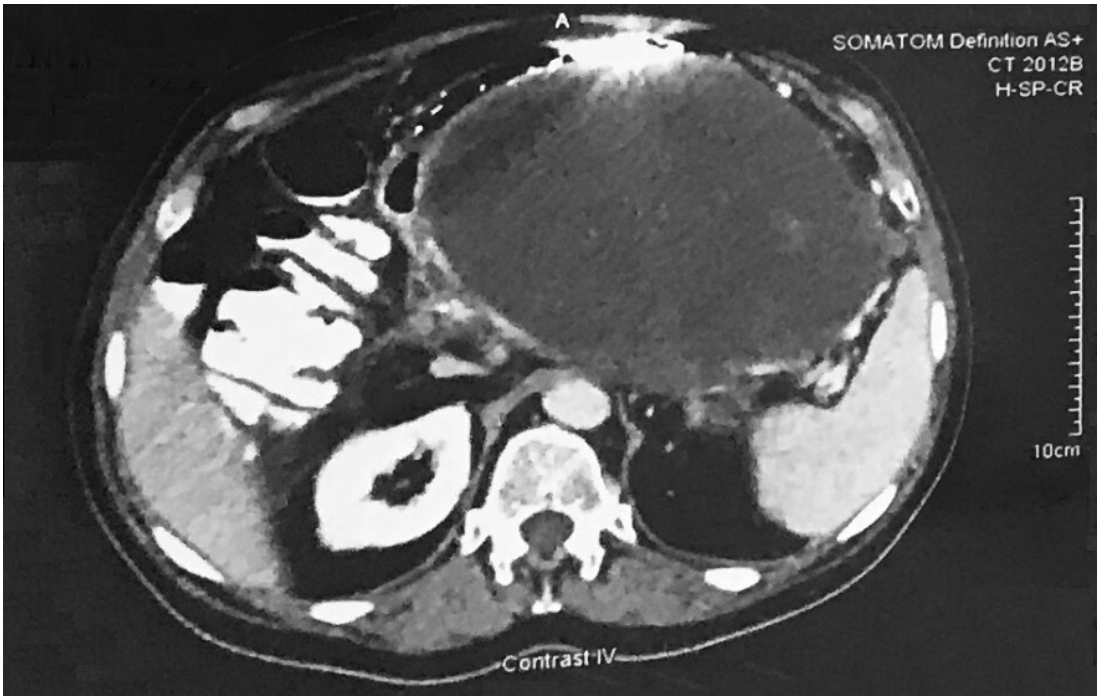

Figure 1. Abdominal CT scan showing a huge (18 cm of diameter) chronic lobulated pancreatic pseudocyst located mainly on the body and tail of the pancreas, surrounded by a thick, dense wall and containing debris with necrotic material within its fluid-filled cavity.

was performed with 500cc of necrotic fluid and solid material (debris), and a metallic stent was inserted into the cyst cavity. A nasogastric tube was placed in order to avoid aspiration.

The patient returned back to our department. The next day the patient was febrile. The site of infection was attributed to aspiration because of inadequate drainage of the pancreatic pseudocyst. Another attempt, for more efficient drainage of the pseudocyst was made, with a new EUS. During this new examination there was found that the stent remained in place but the debris of the pseudocyst was not fully drained. Attempts were made to remove the necrotic material with a basket. A nasogastric tube was inserted directly into the cavity of the pseudocyst aiming to wash out the cavity with continuous lavage with normal saline, which eventually would shrink in second phase **(Figure 2)**.

The symptoms, however, continued and the patient remained highly febrile. Because the clinical status of the patient was getting worse and he became septic it was decided to take him in the operating room for open surgery and drainage of the pseudocyst through the stomach. At laparotomy there was found a huge pancreatic pseudocyst strictly attached to the posterior wall of the stomach and to surrounding tissues. Through a posterior gastrotomy an opening of the pseudocyst was performed and was drained a large amount of infected pancreatic fluid with necrotic debris inside. Also, an extensive necrosectomy of the body and tail of the pancreas was performed **(Figure 3)**, followed by a large anastomosis between the pseudocyst and the stomach. Post-operatively the patient's clinical status was improved rapidly and his recovery was uneventful. His symptoms regressed and he remained afebrile. Repeated CT scan showed that pseudocyst regressed on imaging, the necrotic pancreatic bed excised was clean and the remnant pancreas was healthy. The patient started having normal meals and he was discharged from the hospital in good clinical status. Ten months later the patient remains in very good conditions.

DISCUSSION

Pancreatic pseudocysts are well defined fluid collections surrounded by a wall composed of collagen and granulation tissue without epithelium. The prevalence has been reported to range from 6% to 18% in the acute and from 20% to 40% in chronic pancreatitis respectively. Pseudocysts may be drained using a variety of approaches or a combination of techniques, especially if an internal drainage is feasible. The decision making concerning treatment indication and optimal approach is quite challenging. The Revised Atlanta classification categorizes accurately the peripancreatic collections accompanying or following an acute pancreatitis [4].

Acute pancreatic fluid collection (APFC) that develops early in the course of acute pancreatitis (without cystic wall) and/or necrosis may or may not be present. Acute necrotic collection (ANC) is the development of necrotizing pancreatitis which contains both fluid and necrotizing material. The distinction between those two conditions is difficult in it he first week. As the disease progresses CT images in ANC become more complex. Pancreatic pseudocysts which are encapsulated fluid collections without necrosis and form after 4 weeks. Walled-off necrosis (WON) is another condition which usually appears after 4 weeks of the ANC episode and develops a thickened nonepithelialized wall between the necrosis and the adjacent tissue. Like ANC, WON may involve pancreatic parenchymal tissue. The distinction between WON and pancreatic pseudocyst is very important as a pseudocyst can be treated effectively by draining the fluid in most cases.

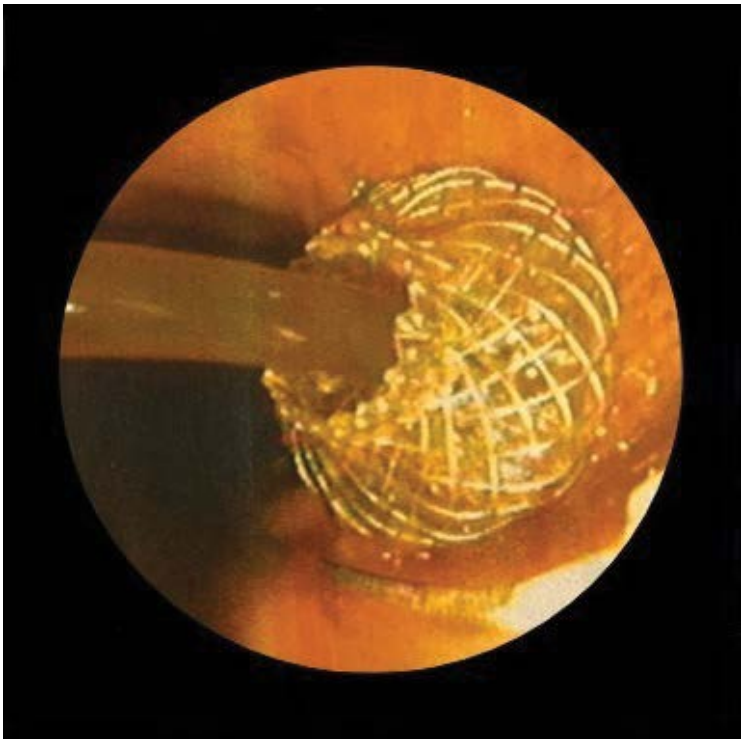

Figure 2. Endoscopic image of the stent in the stomach: transgastric stenting using endoscopic approach into the pseudocyst cavity with a naso-gastric tube inserted through the cysto-gastrostomy transmural stenting.

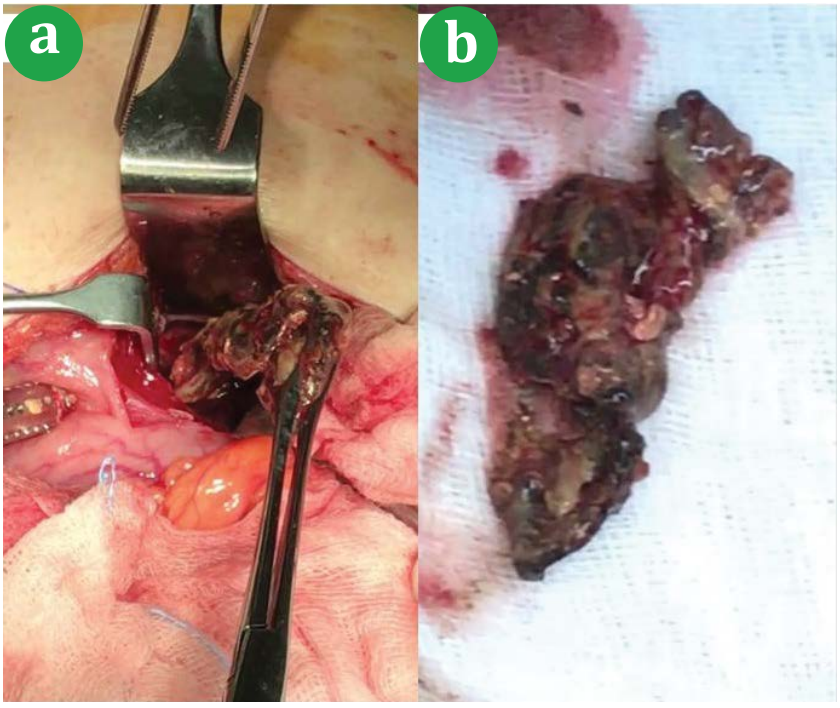

Figure 3. Open surgical procedure. **(a).** Transgastric drainage of the pseudocyst; **(b).** Removal of extensive necrotic tissue from the body and tail of the pancreas.

All these entities can be distinguished by patient's history, imaging studies or even biochemical analysis of needle aspirate if necessary.

Diagnostic Evaluation

The initial diagnosis of a pancreatic pseudocyst can be initially set upon the findings of transabdominal ultrasonography. CT scan is often the method of choice for establishing the diagnosis, with sensitivity up to 100% and specificity of 98%. EUS is another imaging method that helps the clinician to distinguish the characteristics of a pseudocyst. Aspiration of fluid content with concomitant cytology and estimation of tumor markers may help towards distinguishing cystic malignancies from simple pseudocysts.

The clinical course of a pseudocyst highly depends on its size and the time that has elapsed since the diagnosis. Cysts caused by an episode of acute pancreatitis, measured to have a diameter of up to 4 cm, regress spontaneously and require no treatment, given that they remain

asymptomatic. On the other hand, chronic pancreatic pseudocysts show a low regression's rate (less than 10%).

Indications of Treatment

In general, every symptomatic pancreatic pseudocyst that has been present for more than six weeks and has not regressed under conservative treatment should be treated [5].

Among the possible symptoms and complications, there are some specific situations that should be highlighted. Firstly, a firm compression of the stomach or duodenum may lead to symptomatic obstruction, while compression of the common bile duct may cause stenosis and symptomatic cholestasis. A possible compression of major vessels may manifest with angina intestinalis, occult intestinal bleeding due to ischemia, impaired intestinal motility and serum lactate elevation. Furthermore, infection or hemorrhage into the cyst or even formation of a pancreaticopleural fistula may complicate the clinical course of the pseudocyst. The indication of treatment of any asymptomatic pseudocyst with diameter greater than 4 cm that does not resolve after the period of 6 weeks is relative. The supporters of such an approach claim that these cysts do have an increased possibility for complications in the future. A further relative indication is the chronic pancreatitis in presence of abnormalities or stones in pancreatic dust. Pseudocysts that correlate to these conditions, show significantly lower rate of spontaneous regression.

Treatment Options

Management of pancreatic pseudocysts include conservative treatment (watchful monitoring), surgical drainage (open or laparoscopic), or endoscopic drainage. In endoscopic drainage, a stent is inserted in order to achieve a connection between the pseudocyst and the stomach (usually) or even the proximal part of the small bowel. The stent placement may take place under EUS guidance, while an additional insertion of a nasocystic tube (through the stent) may be performed. The optimal treatment of pancreatic pseudocysts remains unclear, as the data extracted from current literature are inconclusive. Hookey *et al.* supported that the treatment of pancreatic pseudocysts always necessitates an interdisciplinary approach [6].

It should be noted that, prior to any intervention, the pancreatic ductal system should always be initially investigated. If the pancreatic pseudocyst does have a connection to Wirsung's duct, the transpapillary stent insertion for internal drainage represents the preferable treatment option. The main principle is that any pancreatic ductal disruption should be bridged with endoscopic stenting, because a remaining disruption of pancreatic duct decreases the rate of cyst resolution after drainage. In cases where the transpapillary drainage of the pseudocyst is not feasible, the endoscopic approach represents the alternative to surgical procedures.

If technically feasible, endoscopic cystgastrostomy for pseudocysts is as effective as surgical cystgastrostomy, but with shorter hospital stays, lower costs and higher patient physical and mental health scores [7]. However other authors as Melman and Gurusamy report that the endoscopic approach is inferior in terms of primary success in comparison to surgical approach as additional interventions were needed in the endoscopic group. As the existing literature is conflicting the results are still controversial as far as the efficacy of the two approaches [8, 9]. Furthermore, after gaining an access in the cavity of a WON, the endoscopic debridement should ideally goal to revealing and removing the granulation tissue covering the pseudocyst wall. Regarding the endoscopic treatment of pseudocysts and WONs, following recommendations have been stated through several authors. Firstly, the distance between the pseudocyst and the gastric or duodenal wall should ideally be less than 1 cm, in order to facilitate an effective stent placement [10]. Furthermore, in the absence of EUS guidance, the chosen approach should be through the site of greatest impression by the pseudocyst on the adjacent gastric or duodenal wall [11, 12]. The greater success rates of endoscopic therapy are noted in cases of single, large (>5 cm), mature cysts that do not communicate with the pancreatic duct [13].

It should be mentioned that the presence of malignancy or the formation of pseudoaneurysm should be ruled out, before any endoscopic attempt. Especially in the second condition, every endoscopic maneuver may lead to troublesome bleeding.

The next question that should be answered is which type of stent should be used. Some authors favor the use of pigtail catheters in contrary to straight stents, because their complication rate is markedly lower. Moreover, the placement of multiple wide stents raises the success rate of endoscopic intervention, without affecting the morbidity or mortality [14].

Traditional self-expanding metal stents (SEMS) are designed to anchor in place in a stricture; however, when used for treatment of pancreatic fluid collection, there is a significant migration risk, as the size and shape of available biliary and esophageal SEMS are not specifically designed for management of pancreatic collections [15]. Much promising is the introduction of the so called lumen-apposing metal stents (LAMS), which make the transmural intervention more effective [16]. This kind of stent ensures a larger lumen for drainage and endocystic access, while ancors firmly in both lumens (intestinal and cystic) showing low migration rates. The first referral to LAMS was published in 2012 and since then LAMS do become steadily more popular in endoscopic cystgastrostomy and pancreatic necrosectomy interventions [17, 18]. The safety and efficacy of LAMS is supported by several recent studies [19, 20]. Their application instead of double pigtail stents (DPS) gains constantly field in drainage of WON. There is strong published evidence that LAMS are associated

with higher regression rate, fewer needed endoscopic interventions and shorter hospital stay [21, 22, 23, 24].

Despite the existing enthusiasm, we should be aware of all possible complications that may accompany the application of endoscopic stents (Pigtails, but mostly SEMS and LAMS).

More specific acute bleeding during drainage procedure or even delayed bleeding (due to stent related erosion) may occur. Factors predisposing to bleeding are the placement of wide metal stents, the presence of pseudoaneurysm or varices, and the blind puction without EUS guidance as well. The bleeding may be quite troublesome and difficult to control, especially if the injured site is located in the cystic cavity. In exceptional cases, an emergency angiography or even laparotomy may be indicated.

Migration of the stent represents a further potential complication. A stent dislocation into the cyst cavity can be more problematic, as the cystgastrostomy tract may partially or completely close. An unusual complication of cardia occlusion after stent migration has been recently reported [25].

Finally, mucosa growth over the stent may lead to its occlusion, predisposing to cyst retention, enlargement or even infection. Furthermore, the stent removal may become quite challenging. Other causes of stent occlusion and drainage impairment may be the interposition of food debris or cyst contents, requiring additional endoscopic interventions [26].

In terms of all possible complications, the administration of SEMS or LAMS should be timely limited. DeSimone *et al.* suggest early removal of LAMS, immediately after cyst regression. If long term drainage is needed, such as in cases of recurrent pseudocysts, plastic stents or pig tails should be preferred [27].

The complication rates after drainage of infected pseudocysts vary among the published studies. Although few studies present high complication rates after drainage of abscesses or WONs [28, 29], some others report contrary findings [30, 31].

The role of surgery is clear, representing the definitive treatment modality, in cases where all other approaches are proven to be not feasible or inadequate. The first successful operation of pancreatic pseudocyst drainage was described by Bozeman in 1882. Technically the pseudocyst usually is incised and debriefed and the cystic wall is anastomosed wherever possible (stomach, duodenum, small bowel).

The rate of surgical success ranges from 90 to 100 %, while the average mortality and morbidity are 2.5 % and 16 % respectively. The recurrence rate ranges from 0-12 % over the next 5 years, depending mostly on the site of the pseudocyst and the underlying illness. Moreover, the surgical treatment of pseudocysts due to chronic pancreatitis carries a markedly lower mortality and morbidity.

The laparoscopic approach is not always feasible and requires a skilled surgical team. The drainage principles remain the same as in the open procedure. The reported success rate is 90% with 0% mortality and 9% complications rate. Finally, transcutaneous drainage is indicated only as an emergency intervention for symptomatic acute fluid retentions or infected cysts. The recurrence rate ranges as high as 70% and percutaneous fistula is a very common complication [32].

In general, surgical (including minimally invasive) techniques are hard to compare because of an evident selection bias. Patients that require transcutaneous intervention are candidates with high morbidity, thus not eligible for surgical treatment.

Gurusamy *et al.* compared four randomized control trials with a total sample of 177 participants, and analyzed the treatment options of pancreatic pseudocysts, extracting interesting results. The different treatments included endoscopic drainage (without EUS guidance), EUS-guided drainage, EUS-guided drainage with nasocystic drainage and open surgical drainage. The authors mention that the overall quality of evidence was very low for all the outcomes, because the trials included a limited number of participants and additionally were at high risk of bias. The analysis of data referring to mortality and serious adverse events showed comparable results between the various treatments strategies. On the other hand, the short-term health-related quality of life (up to 3 months) was worse and the costs were higher in the open surgical drainage group than in the EUS-guided drainage group. The EUS-guided drainage with additional nasocystic drainage caused fewer adverse events than EUS-guided or endoscopic drainage, and had shorter hospital stay when compared to other modalities. Finally, the group of endoscopic drainage showed the higher incidence of additional invasive procedures needed for cyst resolution [33].

According to the Asian consensus statement on endoscopic management of WON, conservative treatment should represent the initial approach to these patients (watchful waiting approach) [34]. The supportive therapy consists in administration of systemic antibiotics and nutritional support, as well as organ and system support. The use of endoscopy is preserved for the patients who fail to improve, despite the aggressive medical treatment of symptomatic or infected WON. Finally, in cases where the drainage procedures are inadequate, necrosectomy should be performed. If an endoscopic approach is not feasible, a surgical necrosectomy (open or laparoscopic) should be undertaken. The authors comment that additional nasocystic drainage has not been proven to be helpful, so its placement is not recommended.

CONCLUSION

The treatment of pancreatic pseudocysts should always involve an interdisciplinary therapeutic approach ensuring an initial adequate support. Endoscopic interventions,

compared to surgical or radiologic approaches are more often successful when multiple wide stents are placed, and this does not elevate either morbitity or mortality. This is particularly true with regard to placement of pigtail catheters. Pigtails catheters are preferable to straight stents, because their complication rate is markedly lower. Finally, in cases of associated pancreatic duct disruption an attempt to bridge via endoscopic stenting may contribute to pseudocyst resolution. In summary, most pancreatic pseudocysts should be managed principally by endoscopic procedures and laparoscopic or open surgical approach would remain reserved to failures.

Conflict of Interest

The authors declare that no conflicts of interest exist concerning this study.

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