CLINICAL CASE

Cutaneous scrotal fistula as a complication of acute necrotizing pancreatitis


Urology Service. Hospital Civil Viejo de Guadalajara Fray Antonio Alcalde, Universidad de Guadalajara, Guadalajara, Jalisco, Mexico

Received 15 October 2013; accepted 25 June 2014
Available online 23 May 2015

KEYWORDS
Hemorrhagic necrotic pancreatitis; Cutaneous scrotal fistula; Pancreatic hydrocele

Abstract
Acute pancreatitis is an inflammatory disease that usually presents as a benign and self-limited pathology, with a 1% mortality rate. Nevertheless, 20-25% of the patients can develop an aggressive form of the disease, with an increase in mortality of up to 30%. Those patients that survive are susceptible to presenting with a series of locoregional and distant complications, mainly pancreatic collections, organized pancreatic necrosis, and pancreatic abscesses and pseudocysts. They can also present with less common and little-reported complications affecting the genitourinary tract, involving the scrotum in the form of hydrocele secondary to the accumulation of pancreatic enzyme-rich fluid whose main action is fat necrosis of the scrotal soft tissue. This can be difficult to differentiate from symptoms of acute scrotum, resulting in unnecessary surgical management that increases patient morbidity and mortality.

Accompanied by a literature review related to the case reported on herein, this article describes a 23-year-old man with a pancreatic hydrocele secondary to hemorrhagic necrotic pancreatitis due to alcohol ingestion. He also developed a cutaneous scrotal fistula, an extremely rare complication of this pathology that is unreported in the literature.

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Resumen

La pancreatitis aguda es una enfermedad inflamatoria que presenta un curso benigno y autolimitado, con una mortalidad del 1%, sin embargo, del 20-25% de los pacientes pueden desarrollar un curso agresivo de la enfermedad, con aumento en la mortalidad de hasta un 30%. Aquellos pacientes que sobreviven son susceptibles a presentar una serie de complicaciones lorregionales y a distancia, predominantemente colecciones pancreáticas, necrosis pancreática organizada, abscesos y seudoquistes pancreáticos, además de complicaciones poco comunes y escasamente reportadas, incluido el tracto genitourinario, donde el escroto se encuentra involucrado en forma de hidrocele secundario al acúmulo de fluido rico en enzimas pancreáticas, cuya acción principal consiste en la necrosis grasa de tejido blando escrotal, que puede ser difícil de diferenciar de un cuadro de escroto agudo, y de este modo, realizar manejos quirúrgicos innecesarios que aumentan la morbimortalidad en estos pacientes.

Se presenta reporte de caso y revisión de la literatura de hidrocele de origen pancreático secundario a pancreatitis necroticohemorrágica por ingesta etílica en paciente masculino de 23 años de edad, así como fistula escroto-cutánea, entidad patológica sumamente rara, no reportada en la literatura hasta el momento.

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Introduction

Acute pancreatitis is an inflammatory disease in a previously healthy pancreas that is generally benign, with a mortality rate of 1%. Twenty to 25% of the patients can develop aggressive disease with evidence of multiple organ failure, which is classified as severe pancreatitis according to the 1992 Atlanta Classification and has a 7-30% mortality rate. That pathology has multiple etiologies; 80-90% develop after alcohol ingestion or are secondary to cholelithiasis, 10% are idiopathic, and the rest are due to other causes, of which hypertriglyceridemia, anatomic duct anomalies, drugs, trauma, iatrogenic origin (CPRE), or hypercalcemia stand out.

Those patients that survive an acute episode may present with expected subsequent complications such as pancreatic collections, organized pancreatic necrosis, and pancreatic abscesses and pseudocysts. Pancreatic necrosis is regarded as a predictive indicator of said complications.

The presence of hydroceles as a complication of pancreatitis is rare and few cases are reported in the literature due to their low incidence or scant recognition on the part of medical services.

The aim of this report was to describe the case of a 23-year-old man with hemorrhagic necrotic pancreatitis, pancreatic hydrocele, and cutaneous scrotal fistula, as well as to provide a review of the literature.

Case presentation

A 23-year-old man, 3 months before hospital admission for alcohol ingestion presented with severe burning pain in the epigastric region that was intermittent and localized. It diminished and then exacerbated 10 weeks after the onset of his current illness, associated with an inability to eat food, fever, asthenia, adynamia, respiratory difficulty, and jaundice. He went to a secondary care hospital where an exploratory laparotomy was performed due to acute abdomen that revealed data of hemorrhagic necrotic pancreatitis. The patient only underwent Penrose drain placement and was referred to our hospital unit. He was admitted to the ICU with Hb 7.8, leukocytes 20 thousand, PLT 273, creatinine 3.6, urea 201, glycemia 292, TB 2.7, DB 1.2, IB 1.5, CRP 36.7, LDH 2066, amylase 3, and lipase 5,840. A tomography scan identified Balthazar D (figs. 1 and 2), associated with the presence of left retroperitoneal fluid descending downwards until reaching the scrotal cavity through the ipsilateral inguinal canal (figs. 3-5). Integrated management of the patient was begun. However, 5 days after his admittance there was an increase in the left scrotal volume, with changes of coloration and temperature. After interconsultation, the finding associated with the spontaneous leakage of scant,

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odorless, bloody, chocolate-brown material (fig. 6) was corroborated. Due to the unstable status of the patient, the patient was given local anesthesia and a raphe incision was made and left scrotal drainage produced 450 cc of material with the above-described characteristics (fig. 7). The testis was macroscopically normal with necrotic zones on the scrotal wall (fig. 8). The inguinal canal was permeable, a Penrose drain was placed and retained for 2 days draining approximately 600 cc of the fluid described above, making a total of 1,050 cc. The scrotal fluid obtained had lipase of 1,834 and its culture was negative. The patient’s clinical status deteriorated, he presented with multiple organ failure, and then died.

Discussion

Acute pancreatitis incidence varies from 5 to 80 per 100,000 inhabitants per year in the industrialized countries. The mean presentation age is 53 years, mean hospital release is 7 days in uncomplicated cases, with spontaneous symptom resolution. This case report was striking due to the fact that the young adult patient was only 23 years old, with a fulminating sub-acute episode and uncommon complications.

The two most frequent etiologies are alcoholism, whose main action is the formation of esters and aldehydes that produce toxicity directly affecting the pancreatic acinar cells, and cholelithiasis, that when complicated with choledocholithiasis, produces a direct one-way valve effect on the pancreatic ducts. In our case, alcoholism was the triggering factor of said clinical symptoms.

The acute attack of pancreatitis can be divided into: mild, 80% of the cases with no tomographic evidence of parenchymatous necrosis, with minimum or no multiorgan failure, with early recovery and no complications; and severe, with necrotizing pancreatitis in 20% of the patients, tomographic evidence of parenchymatous necrosis, systemic manifestations
Severe acute pancreatitis is an important medical problem associated with significant morbidity and mortality, in relation to its complications. Grade D and E abdominal complications, in accordance with the Balthazar classification, occur predominantly between the second and fifth week.

Acute pancreatitis can be associated with a variety of locoregional complications caused by the interaction of the pancreatic fluids with local structures. Fifty-seven percent of the patients with acute pancreatitis present with fluid collections: 2 areas are involved in 39% of the patients and 3 or more areas are involved in 33% of the patients. The fluid collections with elevated levels of pancreatic enzymes are associated with disruption of the pancreatic duct in patients with chronic pancreatitis, and eventually form pancreatic pseudocysts, ascites, pleural effusion, and hydroceles. Our patient presented with left pleural effusion that clinically manifested as respiratory difficulty, as well as the abovementioned hydrocele.

Among the most common complications are the formation of a pancreatic abscess that develops some weeks after the acute attack, rarely during the second week, and should be suspected in all patients with pain, persistent hyperthermia despite standard medical management, nausea, vomiting, and on occasion, a palpable mass.

When the initial peripancreatic fluid is not reabsorbed, it tends to organize into a pancreatic pseudocyst. It is generally found towards the fourth week from the initial process and is spontaneously resolved in up to 40% of the cases. However, it can remain stable for up to 12 weeks. Complications such as bleeding, rupture, or infection of the abscess have been reported in 18-50% of the cases.

Scrotal inflammation is a rare complication of acute pancreatitis and is thought to be the result of fat necrosis of the soft tissue of the scrotum secondary to the destructive effect of pancreatic fluid, that due to tissue digestion can finally condition the formation of a cutaneous scrotal fistula for draining the accumulated material.

The involvement of the genitourinary system, especially the scrotum, is a rare complication of pancreatitis. The first
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Table 1  Case reports in the literature

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<tr>
<th>Author</th>
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<tr>
<td>Babalich AK, et al.</td>
<td>1979</td>
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<td>Lee et al.</td>
<td>2004</td>
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<td>Nazar et al.</td>
<td>2007</td>
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<td>Atiq et al.</td>
<td>2008</td>
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<td>Liu et al.</td>
<td>2008</td>
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<td>Kim et al.</td>
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The case was reported in 1979 and at present there are few reports published in the literature (table 1). It should be mentioned that there are no reports of cutaneous scrotal fistula due to acute pancreatitis up to now.

There is an anatomic interconnection between the left anterior pararenal space with an inferior extension toward the extraperitoneal pelvic tissue and it later involves the scrotal sac.

Embryologically there are 2 structures of the utmost importance in the formation of the inguinal canal: the gubernaculum and the processus vaginalis of the peritoneum. In males, the testicular gubernaculum forms the scrotum and contributes to the descent of the testes toward the scrotum; and the processus vaginalis of the peritoneum, a peritoneal tubular fold, invaginates into the inguinal canal and ends in the scrotum.

Involvement can be unilateral or bilateral and it is clinically indistinguishable from other scrotal lesions; it can even simulate testicular torsion, scrotal infarct, and incarcerated inguinal hernia that are unnecessarily managed through surgical exploration, increasing patient morbidity and mortality.

Clinically, the signs of Cullen and Grey Turner can guide us to a diagnosis of hemorrhagic necrotic pancreatitis, as part of the manifestations associated with this pathologic entity.

It is striking that in other reports on this pathology, scrotal manifestation occurred on the third, fourth, fifth, sixth, and tenth days, respectively, and there was no other case with a prolonged presentation such as that of our patient. He had scrotal manifestation more than 10 weeks after illness onset.

Therefore, it is important for us to be aware of this uncommon complication of an extragenital pathologic process that contributes to diagnostic suspicion in the management of these patients and aids in the integrated management of severe acute pancreatitis.

Conclusions

Hydrocele secondary to hemorrhagic necrotic pancreatitis can present as a rare complication at any time during the progression of this pathology, as the result of the accumulation of pancreatic fluid drained toward the retroperitoneum and by gravity toward the scrotal cavity. However, a cutaneous scrotal fistula is extremely rare, due to the action of pancreatic enzymes that culminate in scrotal tissue digestion, and has not been previously reported on in the literature.

The clinical presentation of this complication is difficult to differentiate from other symptoms of acute scrotum, often leading to unnecessary surgical exploration. Therefore it is important to be aware of this potential complication for opportune diagnosis and treatment and thus reduce the morbidity and mortality in these patients.

Financial disclosure

No financial support was received in relation to this study/article.

Conflict of interest

The authors declare that there is no conflict of interest.

References