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Abdominal cerebrospinal fluid pseudocyst: a complication of ventriculoperitoneal shunt in a Brazilian Amazon woman. Case report

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SUMMARY: **Abdominal cerebrospinal fluid pseudocyst: a complication of ventriculoperitoneal shunt in a Brazilian Amazon woman. Case report.**

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Ventriculoperitoneal shunt (VPS) is the most common treatment for hydrocephalus, however it is not free of complications. Abdominal cerebrospinal fluid pseudocyst (ACP) is an uncommon, but potentially life-threatening, complication of VPS. It is characterized by a fluid filled collection of cerebrospinal fluid (CSF) in the peritoneal cavity containing the distal end of the VPS catheter and is surrounded by a wall composed of fibrous tissues without an epithelial lining.

We report the case a Brazilian Amazon woman that presented ACP fifteen years after the placement of a VPS. Physicians should be aware of this possible complication once early diagnosis would improve outcome and reduce patient's suffering and distress.

RIASSUNTO: **Pseudocisti addominale di liquido cefalorachidiano come complicanza di derivazione ventricoloperitoneale in una paziente dell'Amazzonia brasiliana. Case report.**

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La derivazione ventricoloperitoneale (VPS) è il trattamento più comune per i casi di idrocefalo, ma non è esente da complicanze. La pseudocisti addominale di liquido cefalo-rachidiano (ACP) è una complicanza rara del VPS, potenzialmente fatale. L'ACP è caratterizzata da una raccolta di liquido cefalorachidiano nella cavità peritoneale contenente l'estremità distale del catetere del VPS, rivestita da tessuto fibroso senza epitelio.

Riportiamo il caso di una paziente dell'Amazzonia brasiliana che ha presentato una ACP 15 anni dopo l'inserimento di VPS. Occorre fare attenzione a questa possibile complicanza dal momento che la diagnosi precoce può migliorare la prognosi e ridurre la sofferenza dei pazienti.

KEY WORDS: Ventriculoperitoneal shunt - Cerebrospinal fluid abdominal pseudocyst.
Derivazione ventricoloperitoneale-liquor - Pseudocisti addominale.

Introduction

Ventriculoperitoneal shunt (VPS) is a surgical procedure performed to relieve high intracranial pressure caused by hydrocephalus of diverse etiologies in children and adults (1, 2). A variety of extracranial complications of VPS may be seen, such as omental clogging, abdominal viscera perforation and bowel obstruction (2), however abdominal cerebrospinal fluid pseudocyst (ACP) have been poorly described in the English literature (3, 4). It accounts for about 0.33% to 6.8% of all VPS and, although many cases have been reported in children (5, 6), this process is extremely rare in adults (7-9).

The purpose of this article is to describe the case of a Brazilian Amazon woman with a giant ACP that arose as a complication of VPS.

Case report

A 28 years-old woman, born and residing in the Brazilian Amazon region, was admitted presenting a 3-weeks history of diffuse abdominal pain associated with progressive abdominal distention. Her past medical history was remarkable for a communicating hydrocephalus after bacterial meningitis treated with systemic antibiotics. She had undergone placement of VPS fifteen years ago and after that no other abdominal surgery was performed. The patient had no history of malignancy, pancreatic or liver diseases.

On physical examination, the patient was without fever, dehydrated and presented mild mucocutaneous paleness, abdominal tenderness and distention. A large mass was palpable occupying the upper abdomen. Laboratory examination revealed elevation of the whi-

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te blood cell count (14.200/mL). Computed tomography of the abdomen showed a homogeneous hypodense fluid collection, measuring 24.9x12.8 cm on its larger diameters, with visible walls limit and no contrast capitation (Fig. 1).

Exploratory laparotomy was indicated and, during surgery, a giant abdominal fluid collection was identified located at the upper abdomen containing the distal end of the VPS catheter (Figs. 2 and 3). Adhesions were removed and more than 4L of fluid were drained off. The shunt was repositioned within the peritoneum. All cultures yielded no bacterial growth.

The patient had an uneventful recovery and was discharged home on the 7th postoperative day in good clinical conditions. A follow-up sonography showed no collections of intra-abdominal fluids or any other disease one month after surgery.

Discussion

Ventriculoperitoneal shunting (VPS) is currently the most common treatment for hydrocephalus (8, 9). Shunt complications are reported to occur at a rate of approximately 26% (10) and those associated with the peritoneal tip of the VPS usually include cerebrospinal fluid (CSF) loculation and cyst formation, viscera perforation, migration of the shunt, bowel obstruction secondary to adhesions and metastatic spread via the shunt (2, 11). Abdominal cerebrospinal fluid pseudocyst (ACP) is an uncommon but important complication of VPS. It was first described by Harsh, in 1954, and accounts for about 0.33% to 6.8% of all VPS operated (5, 6, 11, 12).

ACP consists of a collection of CSF in the peritoneal cavity at the distal end of the VPS catheter and is surrounded by a wall composed of fibrous tissues without an epithelial lining (13). The underlying mechanisms involved in the formation of ACP are still unknown, however inflammatory process, either sterile or infectious, is usually regarded as the main causative factor (5-7, 13). The infection rate of ACP ranges from 17% to 80% and *Staphylococcus epidermis* and *Staphylococcus aureus* are the two most commonly cultured microorganisms (4, 5). Additionally, other predisposing factors have been described, such as peritonitis, prior surgical peritoneal adhesions, distal shunt migration, multiple shunt revisions, malabsorption of CSF and allergic reactions (5, 14, 15). In the present report, although culture of the material obtained at surgery yielded no microorganism, we believe that adhesions caused by peritoneal fibrosis were responsible for the reduced absorption of CSF once many fibrous bands were found during surgery.

The most frequent symptoms and signs of ACP in adult patients are pain, distention and a palpable abdominal mass (16), whereas symptoms derived from shunt malfunction, such as headache, nauseas and vomiting more common in pediatric patients (5). The time between last VPS operation and development of ACP has been reported from 3 weeks to 10 years (17). In our case, the

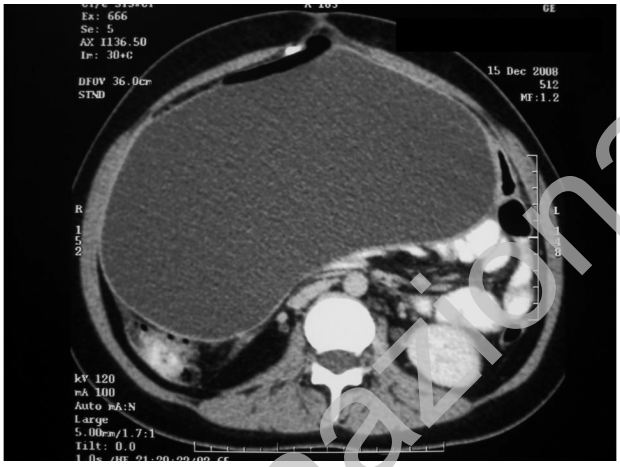

Fig. 1 - Computed tomography of the abdomen showing the intraperitoneal cerebrospinal fluid pseudocyst.

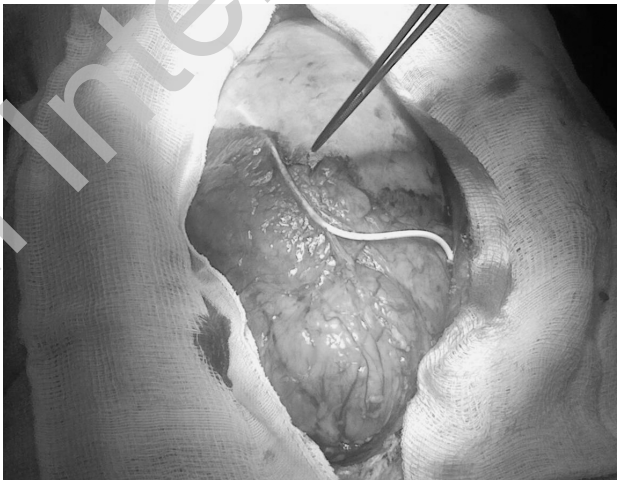

Fig. 2 - Intraoperative finding of the abdominal cerebrospinal fluid pseudocyst.

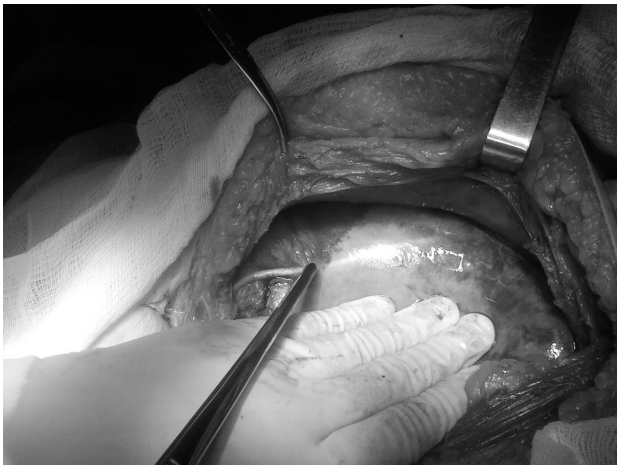

Fig. 3 - Relation between the tip of the VPS and abdominal cerebrospinal fluid pseudocyst.

patient complained mainly of symptoms of pain and abdominal distention. Her last VPS operation was performed fifteen years before the beginning of current symptoms and no other abdominal surgery was performed during that period. This seems to be the longest period reported so far in the English literature.

Ultra-sonography has proved to be the best imaging technique for diagnosing and monitoring possible VPS complications because it is fast and reliable (4, 7). However, computerized tomography (CT) is considered effective in the diagnosis of ACP in adult patients, especially when these are large and deform the normal architecture of the abdomen (7). In the present report, CT revealed a large fluid-filled collection delimited by a thin wall. The catheter tip was adjacent to the fluid collection suggesting the diagnosis of ACP.

The treatment of ACP is controversial. Many therapeutic modalities have been described as curative, for example laparotomy and wide excision of the cystic walls, paracentesis and aspiration of the cystic fluid, CT-guided or sono-guided aspiration of the pseudocyst and, more recently, laparoscopic-assisted lysis of the ACP (3, 4, 6, 18). In the present report, exploratory laparotomy and wide excision of the cystic walls with drainage of the fluid was performed due to a suspicion of peritonitis and intestinal obstruction caused by the abdominal pseudocyst.

In conclusion, the present report highlights that ACP is a uncommon, but possible complication of VPS, and should be included in the differential diagnosis of an acute abdomen complaint.

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