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Gigantic Abdominal Pseudocyst: An Unusual Evolution of the Ventriculoperitoneal Shunt

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Abstract

Background: The ventriculoperitoneal (VPS) shunt is the most common procedure in the treatment of hydrocephalus in children. Abdominal cerebrospinal fluid pseudocysts are a rare complication of the ventriculoperitoneal shunt with an incidence ranging from less than 1% to 10% and are more prevalent in children. The malfunction of the ventriculoperitoneal shunt can cause headaches, nausea, vomiting, altered level of consciousness and abdominal pain due to the accumulation of cerebrospinal fluid. There is no consensus on which type of treatment is better in this case, but there are several available methods. **Aim:** To report an unusual case of a giant abdominal cerebrospinal fluid pseudocyst as a complication of the VPS. **Case Presentation:** Female 1 y/3 months old patient, less than 7 kg, that has been diagnosed with hydrocephalus prenatally, confirmed postnatally associated with an esophageal atresia and distal tracheoesophageal fistula (AE/FTE, Gross III), was admitted to our service with progressive abdominal distention without obstructive intestinal signs or peritoneal inflammatory signs. The CT scan of the abdomen showed a large liquid collection (estimated volume of 600 ml), centered on the umbilical region, diagnosed as a giant abdominal cerebrospinal fluid (CSF) pseudocyst from the VPS. All of data and information were obtained from her medical records at the infirmary of the Conjunto Hospitalar de Sorocaba (CHS), São Paulo. **Conclusion:** Abdominal CSF as a cause of these giant pseudocysts should be considered as a diagnostic hypothesis for cases of large abdominal distensions without intestinal involvement in patients with a VPS. This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Keywords

Abdominal Cerebrospinal Fluid Pseudocysts, Ventriculoperitoneal Shunt

1. Introduction

Hydrocephalus is characterized by the accumulation of cerebrospinal fluid (CSF) in the cerebral ventricles in children. It is usually treated using ventricle shunts to the peritoneum, atrium, pleura or even external [1]. The use of ventriculoperitoneal shunt (VPS) is the most common neurosurgical procedure in the treatment of hydrocephalus in children [1], in order to decrease intracranial pressure [2], avoiding huge head volume. VPS can avoid several types of complications including infections, abscesses, intestinal perforation and abdominal CSF pseudocysts [2] [3] [4] [5] [6].

Despite abdominal pseudocyst was first reported by Harsch in 1954 [2], during the use of shunts for the peritoneal cavity [7], one year later, Jackson and Snodgrass mentioned similar complication as well [8] [9].

CSF pseudocyst has an uncertain etiology but comes from infections in the catheter or due to CSF malabsorption by the peritoneum [2] [5] [10] [11]. The time between placement of the VPS and cyst formation varies from 3 weeks to 5 years [2] and can be mobile or attached to abdominal organs [3]. Larger cysts described in this report tend to be sterile but smaller ones tend to be infected [12] [13]. It is usually preceded by recent inflammatory or infectious state, or recent surgery, frequently in early childhood and adolescence [11] [12] [13].

The diagnosis of abdominal CSF pseudocysts is made more commonly by ultrasound or abdominal CT [3] [4] [5] [10]. The diagnosis must be quick in order to avoid complications [2], which can be: abdominal obstruction, infections, insufficient or excessive drainage of CSF and consequent collapse of the ventricles, or intracranial hypertension [5] [6] [11]. The patient in the present clinical case did not present any of them.

Treatment depends on the clinical condition presented, but percutaneous puncture can be performed with or without repositioning the catheter in an alternative location [5] [10] [13].

The clinical case below aims to report the possibility of the formation of giant peritoneal CSF pseudocysts in the presence of a ventriculoperitoneal shunt, and to differentiate them from clinical conditions relating to bowel obstruction in children with other digestive tract malformations.

2. Case Report

One year and 3 months old female patient, weighing no more than 7 kg, that has been diagnosed with hydrocephalus prenatally, confirmed postnatally associated with an esophageal atresia and distal tracheoesophageal fistula (Gross III). The diagnosis of hydrocephalus was confirmed at birth, and she evolved with an increasing cranial circumference for four days, when a ventriculoperitoneal shunt (VPS) was placed on the left side of her head. The following day the child underwent surgery for the esophageal atresia and distal tracheoesophageal fistula, with an esophagoplasty and suture of tracheoesophageal fistula. At twenty-eight

days the VPS was obstructed and the system was switched to the right side.

When the child was 1 y/3 months old, she was referred to our service with a history of progressive abdominal distension, without obstructive intestinal signs or peritoneal inflammatory signs. Her food and water intake were normal, but there was an increase in her temperature during this period (above 38˚C) and weight gain (500 g in 1 week). The ultrasound showed an image compatible with huge abdominal volume, and that is why she was transferred to the Neurosurgery and Pediatric Surgery service at the Sorocaba Regional Center Hospital, São Paulo, Brazil.

She was immediately admitted to the service and a CT scan of the abdomen was requested, which showed a large liquid collection with an estimated volume of 600 ml (Figure 1). It was then diagnosed as a giant abdominal CSF pseudocyst.

The child underwent abdominal puncture three times and the neurosurgery service decided to exchange the VPS for an external ventricular shunt as an emergency procedure, removing 600 ml of CSF this time. After the last puncture and the removal of the ventriculoperitoneal shunt, the pseudocyst was drained again, and the abdomen gradually reduced.

The patient spent 12 days in the Intensive Care Unit and 18 days at the nursery. She continued without complications and was discharged after 1 month of hospitalization.

3. Discussion

Hydrocephalus generally requires a peritoneal shunt, whose function is to drain

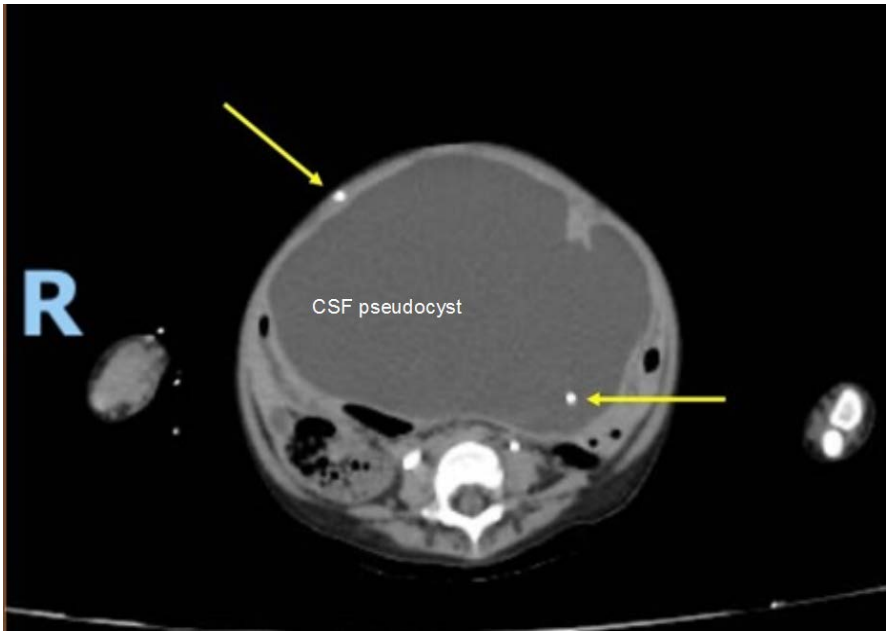

Figure 1. CT image showing the giant abdominal CSF pseudocyst. Arrows show the catheter on the left, still in the intramural pathway of the abdominal wall; and on the right, inside the large cyst.

different amounts of CSF to other sites, such as the peritoneal cavity [6] or to the atrium (VPS) [14]. This type of treatment can present complications with an incidence of 5% to 47%, which include infections, abscesses, intestinal perforation and abdominal pseudocysts [15], such as the present report. Signs of a malfunctioning in VPS are increased intracranial pressure (associated with headache, nausea, vomiting and altered level of consciousness), but hardly ever presents abdominal pain due to the accumulation of CSF in the peritoneal cavity [1] [9]. Over time, this accumulation of CSF can lead to a pseudocyst formation, which is a rare complication, with an incidence ranging from less than 1% [16] to 10% [5] and a higher prevalence in children [9] and depend on a curious formation of the loops around a volume of CSF, forming a pseudocyst that consists of a fragile serous membrane due to a chronic inflammatory process, consisting of adhesions of the adjacent structures and organs (*bowel loops*) [2], with no epithelium [11].

Kashyap S. *et al*., 2017 [5], Tamura A. *et al*., 2013 [10], Pathi R. *et al*., 2004 [12], published clinical cases of abdominal CSF pseudocysts with larger volumes in children such as in the present report: the first one had 1.500 ml, the second 1.260 ml and the third 2.100 ml, respectively [17].

There is no consensus about the best type of treatment for the CSF abdominal pseudocyst, and the decision should follow the symptoms presented by the patient [5] [9]. Paracentesis, CSF aspiration [10] [13], laparotomy or video-laparoscopic approach for debridement of adjacent structures can be performed [9] [18].

Percutaneous drainage with distal repositioning of the catheter in an alternative location (contralateral abdomen wall, pleural space or right atrium) or complete removal of the VPS from the third ventricle are the options for initial treatment of the condition [9] [10] [19]. In this report, the treatment of the pseudocyst was done through percutaneous drainage to decrease the large intra-abdominal volume.

In asymptomatic patients with small volume and those whose volume is not so large, the conduct can be non-operative. The prognosis is poor when taking into account the use of a new VPS, with recurrence rates varying from 7.1% to 62.5% [2].

4. Conclusion

In conclusion, this is a very low incidence case and the diagnostic can be strongly considered in children with a large abdominal distension with absence of intestinal involvement in patients with VPS.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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