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Case Report

Enterocutaneous fistulae presenting as a late complication of a non-functioning Ventriculo- Peritoneal shunt catheter.

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Abstract: A patient with spina bifida and hydrocephalus who had undergone multiple shunt revisions, presented with a 9 month history of chronic discharging sinuses related to a retained shunt catheter not visible on x-ray. This case report demonstrates the importance of clinical history and investigation in patients with retained catheters presenting with cutaneous sinuses.

Key Words: Ventriculoperitoneal shunt, retained ventricular catheter, shunt complications, congenital hydrocephalus, CSF shunt complication.

INTRODUCTION

Since the 19th century the peritoneal cavity has been used as a site for the secondary absorption of CSF (cerebrospinal fluid) via ventriculo-peritoneal shunts.[1] This practice has given rise to multiple complications including abdominal wall perforation, ascites, CSF fluid fistula, hernia, hydrocele, scrotal extrusion, ileus, intramuscular cysts, intussusception, migration of the peritoneal catheter, torsion, peritonitis, pseudocyst/tumour, vaginal and viscus perforation and volvulus. [1] Abdominal complications of ventriculo-peritoneal shunts are reported from 10-30% [2], and bowel perforation in 0.1-1.0% of cases. [3, 4] This case discusses a rare complication of enterocutaneous fistulae via a non-functioning peritoneal shunt catheter.

CLINICAL DETAILS

We report a 39 year-old wheelchair bound man with a background history of spina bifida and myelomeningocele. He suffers from poikilothermia, restrictive lung disease and chronic renal failure. His initial treatment was undertaken elsewhere with closure of the spinal defect at birth and a ventriculo-peritoneal shunt inserted shortly after. He subsequently underwent 13 shunt revisions in the first 2 years of life. His family subsequently moved to the United Kingdom where he underwent a further 5 shunt revisions for shunt blockage and infection. His last shunt revision was performed at the age of 19 years. History from the patient's family revealed that he had 3 shunt catheters in situ; one on the right side which had been tunnelled over the scapular region and then down around his flank into the peritoneum, the second was a ventriculo-atrial (VA) shunt and the third which was a ventriculo-peritoneal (VP) shunt was inserted anteriorly down the right side. The reason for the unusual catheter course over his scapula is unknown. Most of his early neurosurgical records were unavailable at this presentation.

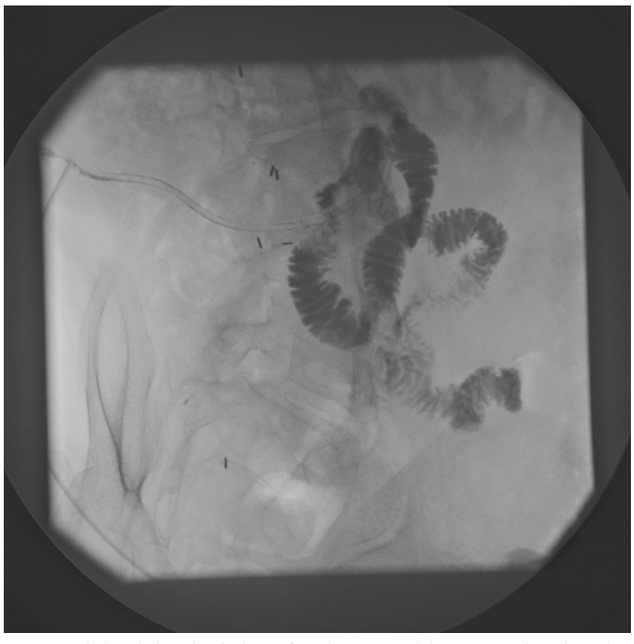

Fig 1. Right abdominal sinus fistulogram with arrows showing the outline of the shunt tubing entering the small bowel and contrast within the small bowel.

He presented to our unit with 2 discharging sinuses. The first sinus had been present for 9 months and was found on the anterior abdominal wall in the right hypochondrium. The second sinus began discharging 3 months prior to admission and was located suprascapularly on the right side. These were initially dressed on a daily basis; with later attempts of laying open the sinuses by general surgeons. Due to their persistent nature and the fact that he posed a poor anaesthetic risk he was referred to neurosurgery for further advice.

He had no new neurological deficit on admission, there was nothing to suggest that the working shunt had malfunctioned in any way, and he had no systemic evidence of infection. He was apyrexial, he had no meningism, there was no evidence of inflammation along any of the shunt tubing tracts and his abdomen was soft and bowel function normal. The peripheral white cell count and inflammatory markers were in the normal range.

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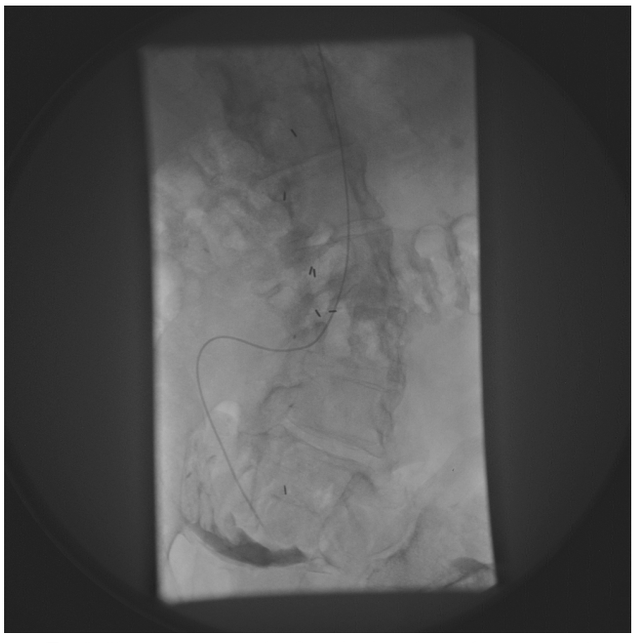

Fig 2. Shuntogram via the functioning shunt shows normal peritoneal drainage of contrast. Arrows show the catheter and drainage of contrast into peritoneum.

A shunt series of x-rays did not show any shunt tubing, although these were palpable in parts. A computer tomography scan of the brain showed evidence of a Chiari type 2 malformation, marked ventricular enlargement and a thickened skull vault. Swabs taken from both sinuses cultured pseudomonas, with MRSA (Methicillin-resistant Staphylococcus aureus) isolated from the abdominal wound only. We performed a fistulogram study using water-soluble contrast media for the abdominal sinus, which showed a well-defined lumen in keeping with the abdominal portion of the shunt tube, and a connection between the skin surface and a loop of small bowel distally with opacification of small bowel loops in the midline (FIG-1). Furthermore, it was noted that there was retrograde flow of contrast up the peritoneal shunt catheter. The supra-scapular sinus was not amenable to catheterisation.

To facilitate further investigation a shuntogram was performed via the valve on the functioning shunt, which showed no contrast extravasation into the skin or bowel with normal drainage into the peritoneal cavity (FIG-2). A small bowel series did not show the previously noted fistula.

As the patient had a sensory level at T6 he underwent exploration of the abdominal fistula without anaesthesia. The tract was explored and the tubing identified. The offending catheter was removed completely with surprising ease, following which the tract was curetted and wound packed with kaltostat. After this procedure the dorsal scapula fistula appeared dry and both fistulae began to close spontaneously.

DISCUSSION

Although the pathogenesis of intra-abdominal complications of VP shunt is uncertain, various possible mechanisms have been proposed. These include chronic low-grade inflammatory reaction and fibrous encapsulation secondary to irritation from catheter, infected CSF or sterile xanthochromic CSF.[2] Encasing fibrosis which has an anchoring effect on the tubing with resultant decubitus ulceration of the bowel wall and eventual bowel perforation has been described as one of the possible mechanisms of bowel perforation. [4] It has also been suggested that children with myelomeningocele and congenital hydrocephalus may have poorly innervated bowel leading to weakness and therefore a susceptibility to perforation. [4] Bowel perforation associated clinical peritonitis was documented in less than 15-25% of cases, as reported by Sells CJ, et al, and Yousfi MM, et al, making clinical diagnosis of this pathology difficult. [4,5,6]

The reports relating to bowel perforation secondary to ventriculo-peritoneal shunts have implicated the spring-coiled Raimondi peritoneal catheter as a cause and incidence has been reduced secondary to softer, more flexible Silastic® catheter usage.[4] The use of these catheters have perhaps contributed to the rarity of fistulae formation with relation to retained shunt hardware.

We are reporting a rare and unusual delayed complication of ventriculo-peritoneal shunting. The fact that the functioning shunt was intact and the retained old non-functioning peritoneal catheter was perforating the small bowel makes our case extremely rare. A literature review identified two previous cases of small bowel perforation in patients with functioning shunts. In both instances the shunt continued to function with no abdominal signs or symptoms. One patient presented with recurrent gram negative ventriculitis and the second patient was asymptomatic. [7] In the majority of cases removing the redundant catheter is difficult because of degradation of the tubing, calcification and fibrous tissue anchorage. In our case chronic low-grade infection had a role in releasing the catheter from surrounding fibrous tissue and facilitated its removal. In order to prevent long-term bowel complications removal of redundant catheters may be considered. In the majority of cases, abdominal shunt complications concerning patients with functioning shunts present with shunt blockage or infection at early stages. Our case which had translucent shunt tubing, an unusual catheter course and no previous documentation available highlights the importance of clinical history and examination accompanied with appropriate radiological investigations for patients with retained non-functioning peritoneal catheters presenting with a cutaneous sinus.

The authors have no conflict of interest.

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