Large Abdominal Cysts: A Complication of Peritoneal Shunts

Report of Three Cases

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Cerebrospinal fluid shunts into the peritoneal cavity are frequently used at Children's Hospital Medical Center, despite their limitations. Considerations leading to a ventriculo-peritoneal shunt include operative scarring of neck veins, cardiac disease, and blood stream or other infection. Similarly, lumbar-peritoneal shunts have been utilized for small or infection-scared ventricular systems, to avoid unnecessary instrumentation of intact brain and because revision to accommodate body growth seems to be required less often with the shunt tubing lying transverse to the body axis.

It is well known that peritoneal shunts often become obstructed by omentum or peritoneal adhesions. This report presents three instances of large, intra-abdominal, cerebrospinal fluid cysts as an unusual, and occasionally dramatic, sequel to peritoneal shunts. Jackson and Snodgrass¹ made brief allusion to one of their 62 patients who formed a "huge omental cyst" around the tip of a peritoneal catheter. Harsh,² in his series of 12 patients with fallopian tube peritoneal shunts, reported one in whom two abdominal procedures for shunt were followed by the formation of a "large thin-walled cyst about the fimbria." Unnecessary and extensive evaluation of an abdominal mass may be avoided by awareness and suspicion of this complication of shunting; moreover, its presence may influence future management.

Case Reports

Case 1. This 3-year-old girl had had a ventriculo-atrial shunt inserted in Peru at 5 weeks of age for aqueductal stenosis. Between 2 and 6 months of age she had had a series of fevers of undetermined etiology with hepatosplenomegaly, leading to a bone marrow aspiration and liver biopsy, both of which were normal.

First admission. The patient was transferred to Children's Hospital Medical Center at 6 months of age because of continuing fever. Staphylococcus aureus, coagulase negative, was cultured from blood as well as from the shunt when it was removed. One month later, while she was still on antibiotics, a ventriculo-peritoneal shunt was inserted, placing the Holter silicone tubing in the suprahepatic space.

Second admission. The patient did well until 2 years and 3 months of age when she returned with intermittent vomiting, lethargy, and deterioration of her gait. The Holter valve pumped well, but she had a cracked pot percussion sound and a head circumference above the 97th percentile. The liver could be felt three fingers' breadth below the right costal margin. In addition there was a firm, midline, upper abdominal mass. An intra- or suprahepatic neoplasm was strongly suspected. The AU¹⁹⁸ hepatic scan showed an anterior indentation of the liver with decreased uptake in the left lobe and adjacent right lobe. Celiac arteriogram suggested a suprahepatic mass. Liver function tests were normal. Plain abdominal x-rays (Fig. 1) showed marked displacement of abdominal contents and located the shunt tubing anterior to the liver. Aqueductal stenosis and markedly dilated ventricles were seen in the ventriculogram.

Operation. Several hundred cubic centimeters of clear fluid were found in the suprahepatic space, the liver appearing normal. After an unsuccessful attempt to perform a ventriculo-atrial shunt, a lumbar-peritoneal shunt was inserted, placing the tubing in the right lower quadrant of the abdomen.

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The child did well for approximately 3 months, when signs of obstruction recurred. A successful ventriculo-atrial shunt was performed in Peru, without removal of the peritoneal shunt. This has functioned well for 10 months.

**Case 2.** At 2 years and 9 months of age, this boy had had a silicone tube lumbar-peritoneal shunt inserted for communicating hydrocephalus. Postoperatively, marked tube tract swelling occurred. At reoperation 2 months later, the tube was found to lie outside the peritoneum and was reinserted. When he was 3 years old, an inguinal herniorrhaphy was performed.

**Examination.** At 5 years (6 months later) he was readmitted. For 2 weeks he had had flank pain and inability to eat normal amounts of food; for 4 days he had shown massive enlargement of the abdomen; for 36 hours he had had a headache. Electrolytes and liver chemistries were normal. X-ray studies showed displacement of all intestinal gas to the right lower quadrant.

**Operation.** The shunt tubing was found entering a red thick-walled cyst (Fig. 2) containing approximately 1500 cc of clear fluid. The tubing was placed in another part of the peritoneum. He did well for 4 weeks, when all his symptoms recurred. The shunt was then removed from the peritoneal cavity and placed in the ureter, after nephrectomy. The patient has had no further difficulty in the 7 months since that procedure.

**Case 3.** This 15-year-old boy was born with hydrocephalus. A ventriculogram performed when he was 2½ months old demonstrated non-filling of the third and fourth ventricles and was followed by an exploration that revealed a large venous lake covering the cerebellum and both occipital lobes. A series of six shunt or shunt-revision operations (left ventriculo-cisternal, lumbar-ureteral, and ventriculo-atrial) ensued; there was one episode of Staphylococcus aureus ventriculitis. The child subsequently showed mental retardation but adequate control of the hydrocephalus.

When the patient was 14 years old he underwent abdominal exploration for ob-
struction of the ventriculo-ureteral shunt. It was found that the tubing led into a retroperitoneal space filled with brown fluid. Because the ureter could not be found, the distal polyethylene shunt tubing was led into the peritoneal cavity. One month later he developed a Staphylococcus aureus coagulase-negative ventriculitis, the ventricular pressure by tube tap measuring 190 mm H₂O. He was placed on penicillin and the lower end of the peritoneal shunt explored. A kinked, twisted polyethylene tube was removed and a Holter silicone tube inserted over the dome of the liver. The upper end of the shunt was revised three times in the following 4 months. A few months later he began complaining of headaches, vomiting, and abdominal pain. An upper gastrointestinal series suggested extrinsic pressure on the lesser curvature of the stomach by the left lobe of the liver. X-ray studies showed that the shunt tube was no longer over the dome of the liver and that there was an epigastric mass in the abdominal wall.

Operation. At abdominal exploration a 250 to 300 cc cystic mass was found in the outer abdominal wall; there was also a multiloculated intraperitoneal cystic mass on the inner abdominal wall. Both contained clear fluid. The Holter tubing was redirected into the right lower quadrant.

Two months later the distal end of the shunt became obstructed by omentum and was removed. A right ventriculo-auriculostomy was performed, using a transthoracic, direct cardiac approach. Because of a Staphylococcus aureus coagulase-negative septicemia, this shunt was removed 1 month later and the ventriculo-ureteral shunt re-established. Since then there has been a 3-year follow-up without complication.

Discussion

Pathological examination of the cyst wall in Case 2 (Fig. 2) showed a thick fibrous wall with chronic inflammatory cells on the inner surface. There were fat cells near the outer surface, possibly where the wall was formed partially by retroperitoneal fat or omentum. The cause of this chronic inflammatory complication must remain speculative. However, infection and especially previous abdominal procedures are strongly suspected as being significant contributory factors. Table 1 summarizes the major clinical features of our patients. In Cases 1 and 3, shunt infections were prominent in the history. Case 2 and Harsh’s case² were both
free of infection. Jackson and Snodgrass did not offer any data on their patient. All three of our patients and that of Harsh had at least one other abdominal procedure in addition to the original insertion of the peritoneal shunt. Our Case 1 had a liver biopsy prior to the insertion of her shunt. Case 2 had a peritoneal shunt revision 2 months after the shunt insertion and an inguinal herniorrhapsy 19 months later, 6 months prior to the development of his cyst. Case 3 had a peritoneal shunt revision 1 month after its insertion and 10 months before the onset of cyst symptoms. Harsh’s patient had a revision of the lumbo-salpingo-peritoneal shunt with insertion of a new tube 3 months prior to the formation of the cyst.

Had a cerebrospinal fluid cyst been suspected from the outset in Case 1, an extensive work-up could have been shortened by more direct and earlier attempts to establish the diagnosis. This could have been done by early surgical exploration of the abdominal portion of the shunt or by the introduction of RISA into the ventricles or shunt tubing with subsequent counting over the course of the shunt.\(^1\)

All three of our patients had significant abdominal masses and symptoms of increased intracranial pressure of varying intensity and duration. In all, the abdomen was explored, the cyst evacuated, and the peritoneal tubing placed in another part of the abdomen. None of the shunts functioned longer than 3 months after cyst evacuation, and in each patient diversion was performed into another space, the blood stream in Case 1, the ureter in Cases 2 and 3. This suggests that the formation of a cyst is a poor prognostic sign regarding the usefulness of the peritoneal cavity for shunting.

### Summary

Three patients have been presented, each of whom developed a large intra-abdominal cyst of cerebrospinal fluid as a consequence of ventriculo- or lumbar-peritoneal shunts. We believe this was an inflammatory phenomenon related to other abdominal procedures or infection. Once such a cyst is suspected, early diagnosis by operation or other means is important. Following surgery for this complication, subsequent peritoneal shunts did not succeed beyond 3 months.

### References