

Case Report

Critical ventriculo-peritoneal shunt failure due to peritoneal tuberculosis: Case report and diagnostic suggestions for abdominal pseudocyst

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Abstract

Background: Tuberculous peritonitis (TBP) is a well-known complication of ventriculo-peritoneal (VP) shunt treatment for hydrocephalus resulting from tuberculous meningitis (TBM). However, a case of hydrocephalus unrelated to TBM resulting from VP shunt malfunction due to TBP has not been reported.

Case Description: A 21-year-old male presented with nausea, abdominal pain, and headache. VP and cysto-peritoneal (CP) shunts had been inserted to treat hydrocephalus due to a suprasellar arachnoid cyst, replaced the VP and removed the CP in his childhood. Computed tomography demonstrated acute hydrocephalus and an abdominal pseudocyst surrounding the distal end of the peritoneal tube. Initial laboratory data showed elevated white blood cell count and C-reactive protein level, but no causative pathogen was identified. External drainage of cerebrospinal fluid (CSF) and of the fluid in the peritoneal cyst was established, and empirical antibiotic therapy was initiated. Bacterial cultures eventually revealed *Mycobacterium tuberculosis* infection, and TBP was diagnosed. The patient responded well to antituberculosis (anti-TB) agents and insertion of a ventriculo-pleural shunt.

Conclusion: This case highlights the possibility of CSF shunt failure and concomitant neurological sequelae from TB infection even when the pathogen has not invaded the central nervous system, as in TBM. Moreover, TBP is rare in developed countries and therefore may be misdiagnosed because of nonspecific clinical features and low sensitivity of common TB screening methods.

Key Words: Abdominal complication, cerebrospinal fluid pseudocyst, shunt malfunction, tuberculous peritonitis, ventriculo-peritoneal shunt

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INTRODUCTION

Tuberculous peritonitis (TBP) is still a common and

serious communicable disease in developing nations and is occasionally seen in patients with specific risk factors, such as malignant diseases, diabetes mellitus,

renal failure with hemodialysis, or immunodeficiency diseases, in developed countries.^[3,10,17,20,23] While TBP is rare, accounting for only 3-16% of extrapulmonary tuberculosis (TB) cases and 0.007-3.8% of total TB cases,^[1,6,8,13,16,24-26] it is prone to misdiagnosis because of nonspecific clinical features and the insensitivity of common TB screens.

In tuberculous meningitis (TBM)-induced hydrocephalus cases treated by ventriculo-peritoneal (VP) shunt, TBP is a well-known cause of shunt failure. However, no case of TBP unrelated to TBM has been reported as the cause of cerebrospinal fluid (CSF) shunt failure and resulting hydrocephalus. This study describes a case of TBP associated with shunt malfunction 8 years after the last shunt inserting surgery in a patient with no history of TBM or obvious risk factors for TB. Pathogenesis and clinical features of TBP as well as possible complications are described in detail to facilitate early identification and effective treatment.

CASE REPORT

Patient history and initial examination

A 21-year-old Japanese male presented at the outpatient department with sporadic nausea, abdominal discomfort, and worsening headache. Cysto-peritoneal (CP) and VP shunts were implanted 11 months after birth by another doctor for the treatment of hydrocephalus due to a suprasellar arachnoid cyst. His VP shunt had been replaced twice by the age of 13 years. At the time of last surgery, the CP cranial tube was removed while the extra-cranial tube was left in place because it passed through the mediastinal space rather than the subcutaneous tissue and adhered to the surrounding organs. During this course of treatment, the patient had been diagnosed with slit ventricle syndrome but had no current restrictions on daily activity except for mild mental retardation. He had no remarkable medical history to suggest systemic abnormalities or risk factors for infection and had not traveled to foreign countries.

Nausea was reported for 2 months and abdominal pain for 1 month before admission. A few days before presentation, the patient developed fever and mild consciousness disturbance. Initial physical examination revealed a fever of 38.9°C, restlessness, and significant abdominal distension but no meningeal irritation or focal signs of neurological dysfunction. White blood cell count was 10,900/mm³, and serum C-reactive protein level was 31.8 mg/dl. Chest X-ray was negative but brain and abdominal computed tomography (CT) scans revealed enlarged lateral ventricles, a huge peritoneal cyst surrounding the distal end of the peritoneal tube, and fluid in the sheath surrounding the peritoneal tube, indicating possible shunt blockage and a concomitant increase in intracranial pressure (ICP) [Figure 1]. In

contrast, chest CT revealed no abnormalities in bilateral lung fields or subcutaneous tissues of the chest. The tentative diagnosis was shunt malfunction resulting from some undefined peritoneal abnormality.

Initial treatment

Continuous external drainage was established to remove CSF from the lateral ventricles and peritoneal cyst. The CSF was clear and the fluid in the abdominal cyst was xanthochromatic. Chemical screening of CSF was negative. The cyst fluid contained 1495 cells/mm³ (69 monocytes and 1426 polycytes/mm³) and 1.4 g/dl total protein. Antibiotic therapies for abdominal infection were empirically administered and bacterial cultures analyzed, but no causative pathogen was identified.

Postoperative course

Preoperative clinical symptoms immediately improved after surgery to insert an external drainage system. However, the cyst fluid gradually recollected and signs of mild inflammation remained despite antibiotic therapy. In the second stage of treatment, transcutaneous drainage of the cyst under CT guidance was performed; however, again cyst fluid recollected [Figure 2]. In the third stage of treatment, laparotomy was performed, which revealed no membrane surrounding the abdominal cyst (defining it as a pseudocyst), no malignancies within the omentum, and no ascites within the abdominal cavity.

Throughout the course of treatment, all cytological examinations for malignancies were negative and bacterial cultures did not reveal a causative pathogen. Two months after initial surgery, inflammation of his left knee joint became apparent. The fluid taken from the joint was xanthochromatic, similar to the pseudocyst fluid, and *Mycobacterium tuberculosis* was detected in a culture from the fluid. Thus, the patient was diagnosed

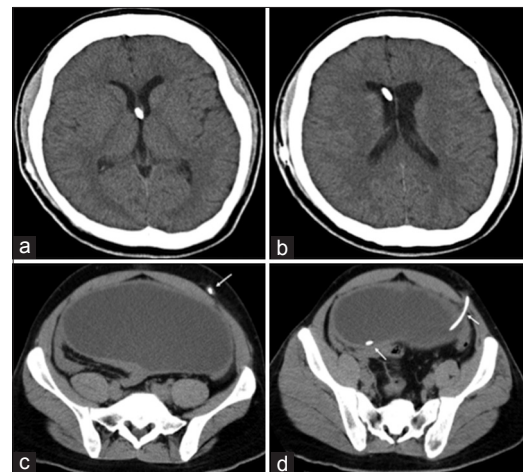


Figure 1: Neuro-radiological findings on presentation. Brain CT showing enlarged lateral ventricles with slit ventricle (a, b), and abdominal and pelvic CT demonstrating the course of the ventriculo-peritoneal shunt catheter (arrows) and the massive cerebrospinal pseudocyst surrounding the peritoneal end (c, d)

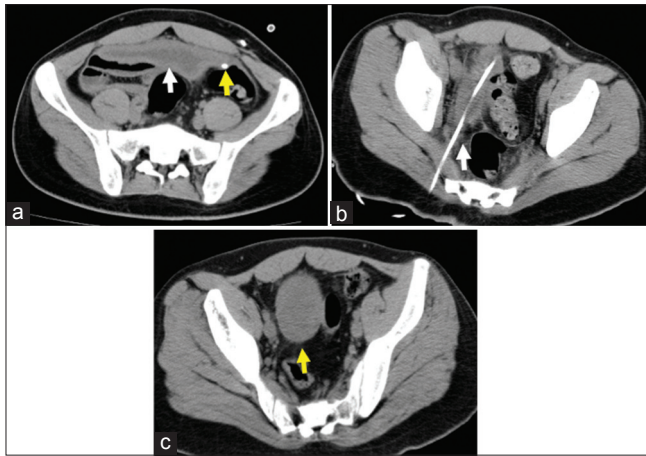


Figure 2: Abdominal and pelvic CT after the first (a) and second (b, c) surgeries. Distal part of the shunt tube inserted for external drainage (yellow arrow) of the pseudocyst. Although the fluid volume decreased, it was not completely cleared (white arrow) (a). CT showing the transcutaneous drainage tube (white arrow) and the recollected cyst fluid (yellow arrow) (b, c)

with an abdominal pseudocyst associated with TBP and successfully treated by anti-TB agents following insertion of a ventriculo-pleural shunt [Figure 3]. At follow-up 2 years after treatment, he again presented with signs of inflammation, and the abscess around the CP shunt was strongly suspected as the cause. He was again treated with anti-TB agents, and all signs of inflammation subsided after the removal of the previous CP shunt tube by thoracoscopic surgery.

DISCUSSION

The present case illustrates that TBP alone, in the absence of TBM, can cause CSF shunt failure with ensuing elevations in ICP. Clinicians should be aware of this possibility and the potential for neurological damage. While TBP is a relatively rare type of TB infection in industrialized countries (and thus extremely rare in patients with CSF shunts), it is still a common infection in many regions of the world, and delayed diagnosis or misdiagnosis can be fatal.^[2] TBP usually yields lower bacterial counts, and both the clinical and radiological features are nonspecific,^[6,7,12,16,26] making it one of the most difficult forms of TB to correctly diagnose. This is especially true in patients without obvious risk factors, such as exposure to TB, cirrhosis, alcoholism, renal failure with peritoneal dialysis, diabetes mellitus, malignancy, immunosuppressive medication (e.g. corticosteroid or antitumor necrosis factor agents), or acquired immune deficiency syndrome, which collectively account for 16.7-42% of all TBP patients.^[4-6,14,25] This case suggests that TBP may be particularly dangerous in patients with CSF shunts.

Many cases of abdominal pseudocyst formation after insertion of CSF shunts have been reported since the first description by Harsh *et al.* in 1954.^[10] Abdominal pseudocyst formation occurs in 0.33-6.8% of shunt

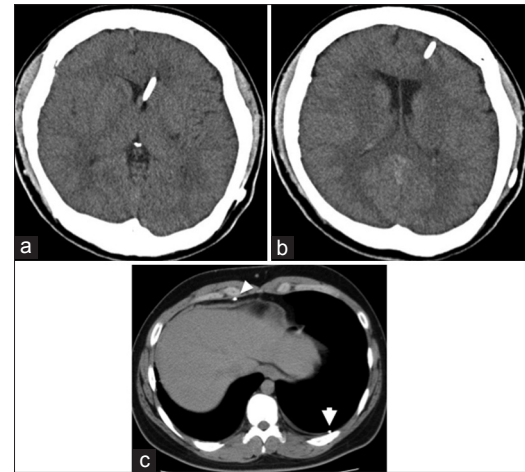


Figure 3: CT after insertion of the ventriculo-pleural shunt. Brain CT showing decreased lateral ventricle volume (a, b). Chest CT showing the distal part of the shunt (arrow), a small amount of cerebrospinal fluid in the thorax and the distal tube of the previous CP shunt (arrowhead) (c)

procedures due to abdominal infection.^[10,18,19] In majority of these cases, external drainage and antibiotics result in good clinical outcome.^[22] In some of these cases, inflammatory cells on the inner surface of the cyst and bacteria such as staphylococci were detected.^[18,22] In problematic cases like the present one, however, inflammatory signs and symptoms may be mild, absent, or equivocal, and pseudocyst formation may occur several years after surgery.^[21] Even in cases where the role of infection is unclear, antibiotics have been empirically administered (but with little effect). In these treatment-resistant cases, some authors have suggested laparotomy with wide excision of the cyst walls^[9,19] or laparoscope-assisted lysis of the abdominal pseudocyst.^[11] The present case and several previous case series suggest that multiple abdominal shunt surgeries greatly increase the probability of abdominal pseudocyst formation. Kariyatti *et al.* reported that patients with abdominal pseudocysts had an average of 2.9 shunt revisions compared with an average of only 0.75 revisions in a group with ascites.^[15] They also suggested that several abdominal surgeries could cause omentum fibrosis, resulting in reduced absorption of CSF. In the present case, the patient had undergone three shunt surgeries. Thus, although the etiology of abdominal pseudocysts is not well understood, multiple shunt surgeries may increase the risk.

CONCLUSION

The authors described a rare case of abdominal CSF pseudocyst associated with TBP unrelated to TBM. This case indicates the possibility of CSF shunt failure and concomitant neurological sequelae from TB infection even when the pathogen has not invaded the central

nervous system. Moreover, multiple shunt surgeries may increase the risk for pseudocyst formation.

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