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Case Report

Intriguing case of giant intra-abdominal pseudocyst: Diagnostic dilemma

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ABSTRACT

We present a case of a giant intra-abdominal pseudocyst in a 24-year-old male as a complication of ventriculoperitoneal (VP) shunt. Ultrasonography and computed tomography abdomen detected a 20 × cm 14.5× cm 9 cm thin-walled cystic lesion with few septae occupying a large space in the left side of the abdomen with a VP shunt tip within it. Histopathological findings suggested a pseudocyst. However, multiple epithelioid cell granulomas on cyst wall resulted in a diagnostic dilemma.

Keywords: Granulomatous inflammation, intra-abdominal pseudocyst, tuberculosis, ventriculoperitoneal shunt

Introduction

Non-pancreatic intra-abdominal pseudocysts are uncommon. The reported incidence is approximately 1/100,000 to 250,000 hospital admissions. They are mostly associated with major intra-abdominal surgeries, trauma, ventriculoperitoneal (VP) shunts, or intra-peritoneal catheter infections. They are an important complication of VP shunt surgery. The prevalence of intra-abdominal pseudocyst secondary to VP shunt has been reported to be from 0.25% to 10%.[1]

We present a case of a giant intra-abdominal pseudocyst formation as a complication of VP shunt.

Case Report

A 24-year-old male presented to the hospital in March 2019 with diffuse pain in abdomen, abdominal distension and multiple episodes of non-bilious, non-projectile vomiting for 3 days. The abdomen was markedly distended with diffuse tenderness, rigidity, and with a vague palpable lump.

The patient was admitted in Oct 2018 with giddiness and loss of balance. Magnetic resonance imaging and computed tomography (CT) brain showed communicating hydrocephalus with periventricular ooze with a possibility of tuberculous meningitis (TBM). There was no evidence of space-occupying lesion. Cerebrospinal fluid (CSF) examination revealed normal protein (42.68 mg/dl) and mildly reduced sugar (41.98 mg/dl). Routine microscopy of CSF showed 14 cells/cu.mm (only lymphocytes). CSF was sent for Gram stain, Ziehl Neelsen (ZN) stain, culture, mycobacteria growth indicator tube, and GeneXpert for tuberculosis, all of which were negative. Anittuberculous treatment was not started. VP shunt was placed for drainage of excess CSF and the patient was discharged.

The patient presented again in February 2019 with pain in abdomen and nausea. Ultrasonography (USG) abdomen revealed a 2.5 cm × 1.4 cm ×1 cm pseudocyst in the left iliac fossa, around the distal end of the VP shunt. The shunt was repositioned to the right iliac fossa and a draining tube was placed into the cyst.

During a recent visit, his total white blood cell count was normal (5400/cu.mm). USG and CT abdomen detected a 20 cm × 14.5 cm ×9 cm thin-walled cystic lesion with few septae, occupying the left iliac fossa and extending into the pelvis with tip of VP shunt within it [Figure 1]. The possibility of CSF pseudocyst was suggested.

The cyst was drained, partly excised, and the VP shunt was converted to a ventriculoatrial shunt. Cyst fluid examination revealed normal protein (25.04 mg/dl) and mildly reduced sugar (45.76 mg/dl). Routine examination of cyst fluid showed only 2 cells/cu.mm (all lymphocytes). Microbiological analysis of cyst fluid did not show any growth of microorganisms. GeneXpert of CSF for tuberculosis was negative.

Tissues from the cyst wall were sent for histopathological examination. Gross findings revealed 7 cm × 5 cm × 0.2 cm light brown, membranous tissue bits, with a smooth glistening surface. No solid areas or papillary excrescences were identified. Microscopy revealed a cyst wall composed of fibrocollagenous and fibrofatty tissue without definite lining epithelium. The wall showed many non-caseating epithelioid cell granulomas with Langhans giant cells along and mixed inflammatory infiltrates [Figure 2a and b].

Periodic acid-Schiff stain was negative. No foreign bodies were found during the extensive screening of the histopathology slide; hence, foreign body etiology was ruled out too. ZN stain was also negative.

Discussion

VP shunt is the most popular treatment for hydrocephalus. It helps to drain excess CSF into the peritoneal cavity which is the best site for CSF absorption. Infection, multiple shunt revisions, and shunt obstruction results in the wrapping of mesentery around the peritoneal catheter and prevents CSF from gaining access to the rest of the peritoneal cavity resulting in the formation of intra-abdominal pseudocyst.

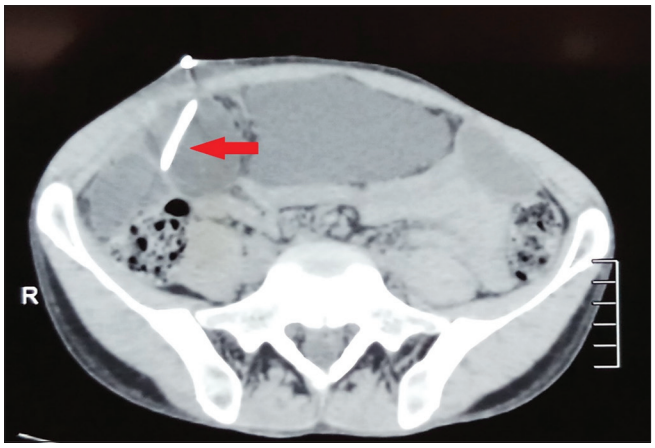

Figure 1: Computed tomography of abdomen and pelvis showing a giant intra-abdominal pseudocyst with tip of the ventriculoperitoneal (VP) shunt within it and the course of the VP shunt catheter (arrow)

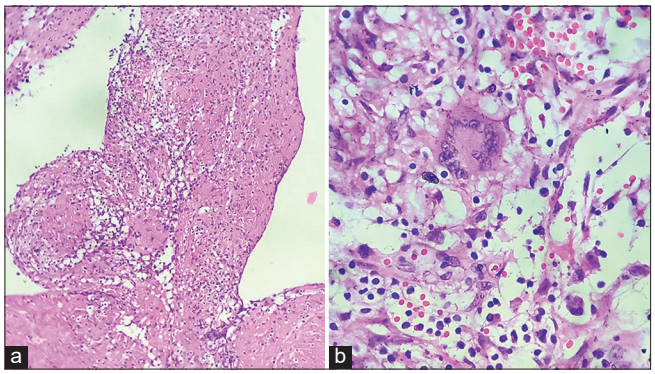

Figure 2: (a) Photomicrograph showing a fibrocollagenous cyst wall without definite lining epithelium (Hematoxylin and Eosin stain, 100×). (b) Photomicrograph showing cyst wall with many non-caseating epithelioid cell granulomas with Langhans giant cells (Hematoxylin and Eosin stain, 400×)

Our patient was diagnosed with intra-abdominal giant pseudocyst on CT scan. They have to be differentiated from various neoplastic and non-neoplastic conditions. de Perrot *et al*.[2] identified the differential diagnosis for intra-abdominal cyst based on histopathological features which included: Cysts of lymphatic origin, cysts of mesothelial origin, cysts of enteric origin, cysts of urogenital origin, mature cystic teratoma, and pseudocysts. Accurate diagnosis of pseudocyst can be obtained by radiological, histopathological, and other laboratory findings, including tumor markers, to rule out malignancy.

Clinical and histopathological findings helped us to rule out cyst of lymphatic, mesothelial, enteric, urogenital origin, and teratoma. However, cyst wall did not show definite lining epithelium; hence, mesenteric cyst was ruled out and the diagnosis of pseudocyst was confirmed. In addition, the wall showed multiple non-caseating epithelioid cell granulomas with Langhans giant cells. It did not show any foreign body/foreign body type giant cell and no fungal elements were revealed even on special stains. ZN stain was negative. It is a well-known fact that the sensitivity of ZN stain to diagnose acid-fast bacilli is low, ranging from 0% to 35% only and the yield of positive culture for *Mycobacterium tuberculosis* is much lower.

Hahn *et al*.[3] studied 26 cases of abdominal CSF pseudocyst associated with VP shunt. The typical presentations were abdominal pain, distension, nausea, and vomiting which were similar to our case. Infection was the etiology in 36% of the cases. Previous shunt revisions were done on an average in 11.2 cases.

Sanal *et al*.[4] studied 8 cases of abdominal CSF pseudocyst associated with VP shunt. They could not find any relationship between the bacterial growth and CRP levels. Spinal protein, CSF cell count, and glucose concentrations were normal in all cases.

As per our knowledge, there is a single case in literature by Takase *et al*.[5] of abdominal pseudocyst associated with tuberculous peritonitis (TBP) presenting after 8 years in a 21-year-old male with hydrocephalus due to a suprasellar arachnoid cyst with multiple shunt surgeries. The bacterial culture of cyst fluid revealed *M. tuberculosis* infection. They concluded that pseudocyst was secondary to TBP without TBM and multiple shunt surgeries may increase the risk of pseudocyst formation.

In a country like India, where the prevalence of tuberculosis is high, the most common cause of hydrocephalus is TBM. Raut *et al*. from India had reported that the incidence of

hydrocephalus due to TBM was 65%.[6] Our case is similar to the case discussed by Takase *et al*.[5] However, a definite diagnosis of tuberculosis could not be obtained in our case in spite of granulomatous inflammation which was seen in the cyst wall and hence, a diagnostic dilemma.

Conclusion

In spite of the high prevalence of tuberculosis and the presence of granulomas on histology, we could not provide a definite diagnosis of tuberculosis. The patient was not started on antituberculosis treatment since none of the laboratory tests were positive for tuberculosis. At present, he is on symptomatic treatment and follow-up has been advised.

Authors' Declaration Statements

Ethics approval and consent to participate

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Patient's informed written consent was taken for publishing the case in a scientific journal.

Availability of data and material

The data used in this study are available and will be provided by the corresponding author on a reasonable request.

Competing interest

We declare that we have no significant competing financial, professional, or personal interests that might have influenced the performance or presentation of the work described in this manuscript. The authors declare that they have no conflicts of interest.

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None to declare.

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