

ASYMPTOMATIC SHUNT FRACTURE IN A PATIENT WITH HISTORY OF TUBERCULOUS MENINGOENCEPHALITIS: A CASE REPORT

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ABSTRACT

Ventriculoperitoneal (VP) shunt is the most frequently performed procedure in patients with hydrocephalus, but can cause serious complications. Shunt fractures, is a rare complication of VP shunt and can be damage for patient. The question of whether asymptomatic patients should or should not be operated on remains to be answered.

The authors report a case of a pediatric patient who had an asymptomatic shunt fracture with a history of tuberculous meningoencephalitis (TBM).

We report the case of a 7-year-old girl with a shunt fracture and a history of hydrocephalus due to TBM. She presented to the hospital in 2021 without symptoms of increased intracranial pressure and was fully conscious. Three weeks later, the patient experienced a gradual loss of consciousness. The result of the examination revealed that the hydrocephalus had become larger than before the operation in 2015. The peritoneal shunt had completely migrated into the peritoneal cavity. An emergency shunt revision was performed at the left Kocher point. After the operation, the patient regained consciousness and lived life without any complications.

Although the decision to re-operate in an asymptomatic patient with a shunt fracture is debatable, shunt revision should be considered. Early revision of the shunt fracture does not pose a serious hazard to the patient.

KEY WORDS: shunt fracture, shunt complications, hydrocephalus, tuberculous meningitis, child health

INTRODUCTION

Hydrocephalus is a condition that should be carefully assessed [1]. Although the ventriculoperitoneal (VP) shunt is still the mainstay of hydrocephalus management today, this procedure most often requires a lifelong commitment to monitoring its function and to prevent any of its many complications [2, 3]. A shunt fracture is a rare one complications of shunt procedures but are the second most common cause of shunt failure in children [3–6]. Because patients with shunt fractures do not always show signs of intracranial hypertension, these asymptomatic patients are often treated non-operatively and are even considered shunt [6, 7]. However, it is important to note that hydrocephalus in these patients may still develop over weeks to years [8, 9]. A similar condition can be observed in tuberculous meningitis (TBM) where hydrocephalus can develop without the need for a shunt procedure [10, 11]. TBM causes hydrocephalus through obstruction of the arachnoid granulation by exudate. In a more advanced phase, scar tissue will form and interfere with cerebrospinal fluid (CSF) absorption, thereby exacerbating hydrocephalus [12–14]. In a previously reported case study, hydrocephalus in tuberculous meningitis may appear up to 14 months after starting anti-tuberculosis therapy (ATT) [10, 11].

Here, we report a case of pediatric patient who had asymptomatic shunt fracture with history of tuberculous meningoencephalitis.

CASE REPORT

HISTORY AND EXAMINATION

A 7-year-old girl with a history of VP shunt insertion 5 years ago came to our hospital in 2021. She complaining of a lump in the shunt line on her right neck (Figure 1) that had been felt for 3 days earlier. The patient was previously diagnosed with TB meningoencephalitis and acute communicating hydrocephalus. The one-year ATT regimen was completed and the patient was declared free of TB. When he came to the clinic, the patient had no complaints or symptoms related to an increased intracranial pressure (ICP). The patient was fully conscious with Glasgow Coma Scale (GCS) E4V5M6, normal bilateral pupillary reflexes and no papilledema. There were no outstanding findings on physical examination apart from a lump in the shunt pathway. Therefore, conservative treatment is considered appropriate. The patient was discharged from the clinic with a take-home message about dangerous clinical signs associated with elevated ICP.

After 3 weeks, the patient came to the emergency room due to loss of consciousness and vomiting. The GCS presentation was E2V2M4 with bilateral decreased

pupillary reflexes. Funduscopy examination revealed papilledema in both eyes. Meningeal signs are absent. A computed tomography (CT) scan of the head showed hydrocephalus with a significantly larger lateral and 3rd ventricle compared to a preoperative head CT in 2015 (Figure 2). A discontinuity of the shunt tube was seen on the cervical radiograph (Figure 3) and was further confirmed by the absence of a shunt catheter on the thoracoabdominal film. A shunt fragment was identified in the left iliac region (Figure 4), indicating that the peritoneal shunt had completely migrated into the peritoneal cavity.

SURGERY AND POSTOPERATIVE COURSE

Emergency shunt revision was performed at the left Kocher point. The initial pressure was 20 cmH₂O with macroscopically clear CSF (Figure 5). CSF analysis was unremarkable for signs of infection. Because of the difficulty in evacuating the peritoneal shunt through the previous abdominal incision, the shunt fragment remains in the peritoneal cavity. On the first postoperative day (POD), the patient regained consciousness. Pupil diameter and pupillary light reflex were normal. The patient was discharged with POD-3 without complications.

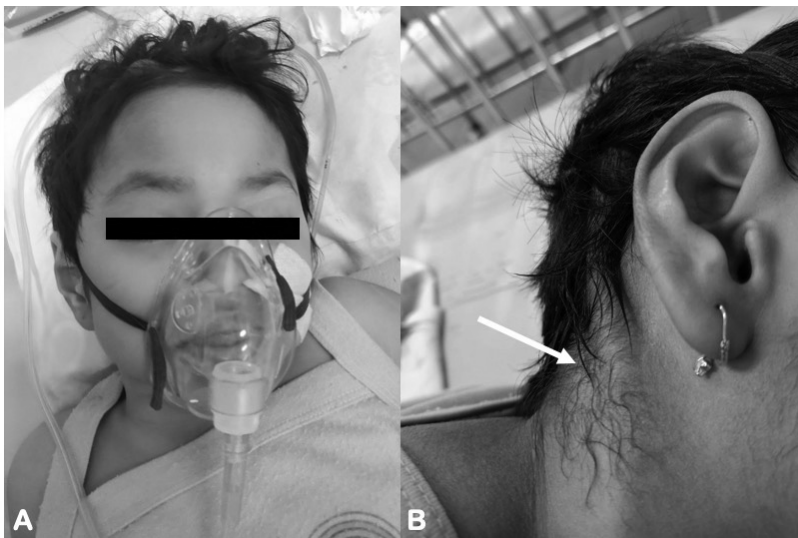


Fig. 1. A 7 years old girl presents with decreased consciousness (A). Lump on the neck as a sign of shunt fracture (white arrow) (B).

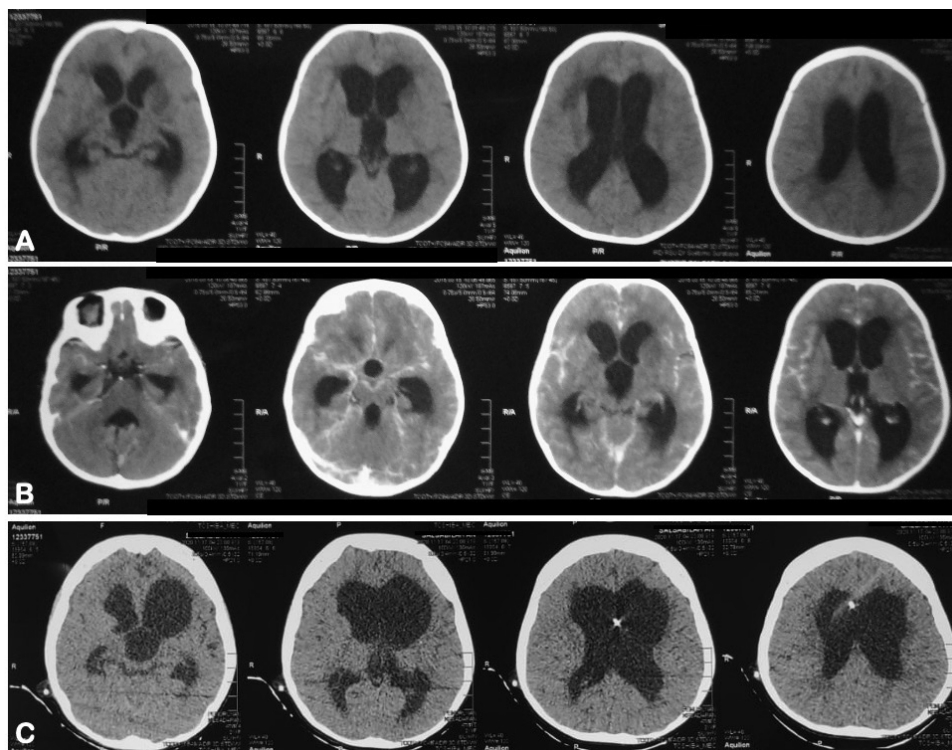


Fig. 2. Head CT-Scan in 2015 with hydrocephalus, showing dilated temporal horn and ballooning of the lateral ventricle, and 3rd ventricle (A and B). Head CT-scan with contrast, depicting leptomenigeal enhancement, especially of the basal cranium (B). Head CT-Scan in 2020 (C), it appears lateral ventricles and the 3rd ventricles widened compared to previous head CT-Scan in 2015 (A and B).

DISCUSSION

As not all patients with asymptomatic shunt fractures develop hydrocephalus, early shunt revision in such cases has been debated [4, 8, 15, 16]. Non-operative management is considered reasonable as long as follow-up is adequate and the patient is appropriate. good information about the warning signs of intracranial hypertension. In TBM, not all cases develop hydrocephalus. Schoeman et al reported that 83% of cases of TBM in the pediatric population developed hydrocephalus while only 12% of adult cases followed a similar course [10, 12–14, 17]. In line with these findings, Güneş et al reported that hydrocephalus in children with TBM was found in 90.3% of cases [18].

In this case report, the patient initially presented with no symptoms of increased ICP even though the shunt had fractured 3 days before the outpatient visit. This is most likely due to the flow of patent CSF through the distal frac-

tured tube. That said, the neurological worsening that followed was indeed due to an increase in ICP as evidenced by the presence of papilledema. Neck motion has been thought to close the distal end of a fractured catheter [4, 15], although the process will not be so severe as to cause hydrocephalus. Therefore, the hydrocephalus in this case indicates that the patient has permanent impairment of CSF absorption due to previous TBM.

The cause of the shunt fracture in this case is uncertain, and the most likely explanation is excessive neck movement [4, 15]. A calcified shunt was considered by us but was never confirmed by microscopic examination of the shunt. The patient's family refusal was based on their personal belief that the shunt fragment should be buried in its entirety. A torn shunt due to a faulty shunt passer is also a possibility. This hypothesis is based on our unpublished experience in which the

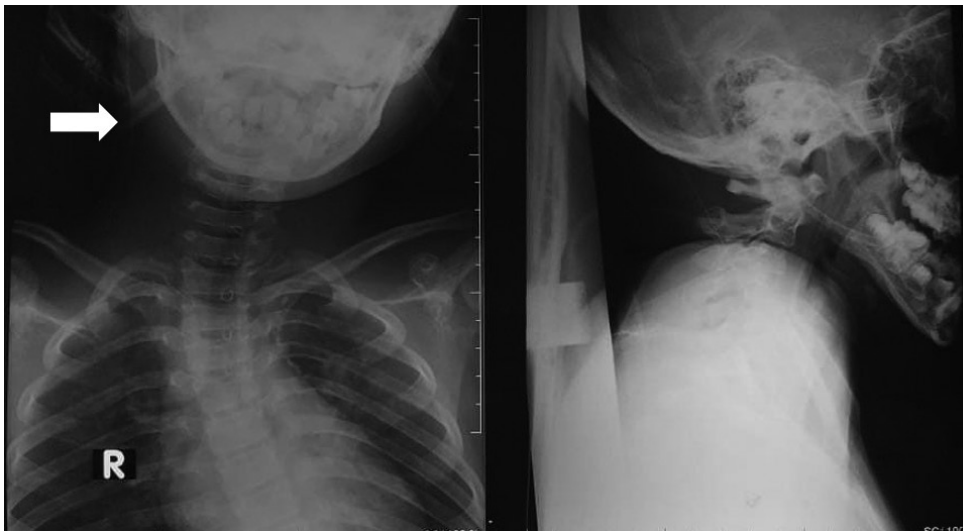


Fig. 3. Cervical AP/Lateral with discontinuities shunt (white arrow).

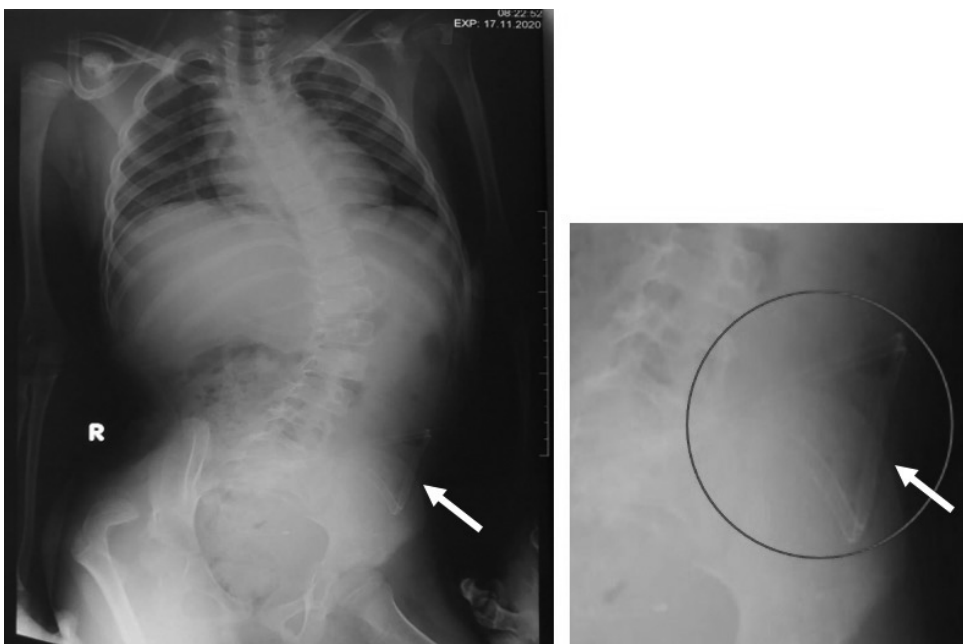


Fig. 4. Thoracoabdominal x-ray showed no shunt tube along the shunt track, shunt fragments in the peritoneal cavity of the left iliac region (white arrow).

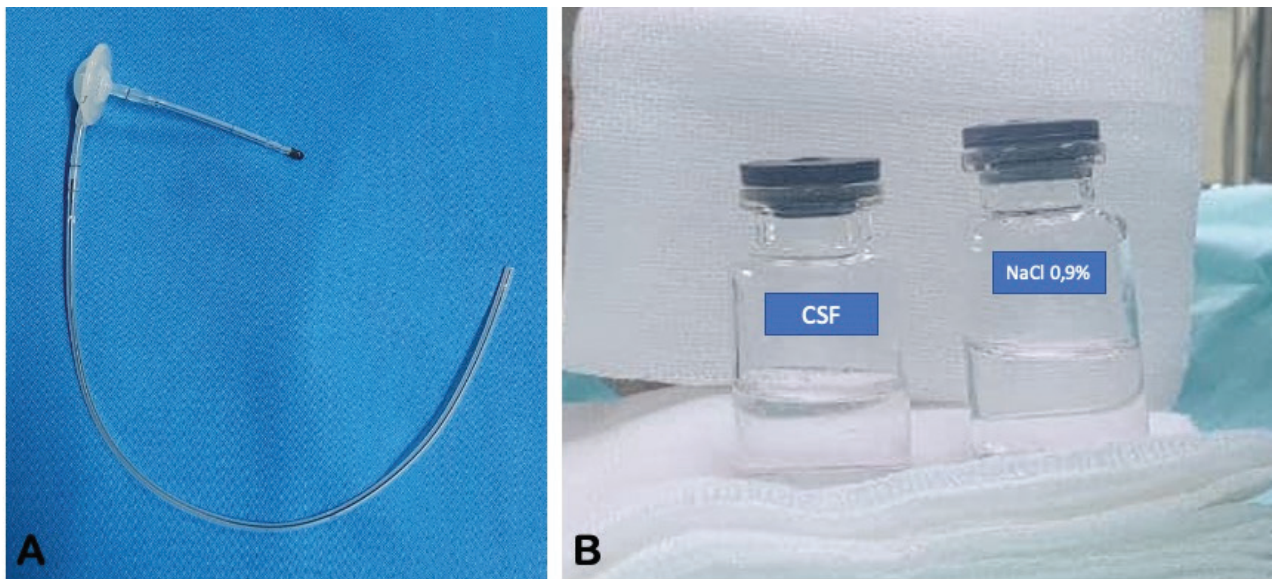


Fig. 5. Ventricular shunt after shunt revision (A). Macroscopic CSF compared to 0.9% NaCl (B).

shunt pass hole, through which the distal shunt catheter is connected, has small metal protrusions due to imperfect smithery. Rough manipulation of the shunt catheter during surgery can also cause small cracks or even full-thickness tears in the tube, which will result in fracture at a later date used.

Given the prevalence of hydrocephalus in TBM, children with shunt fractures and a history of tuberculous meningitis should be considered for surgery. The choice of non-operative management due to the absence of neurologic signs or symptoms, and the possibility of the patient being shunt-free, should be weighed against the risk of intracranial hypertension that may develop days or weeks later. Until now the authors have not found a similar case report that specifically discusses asymptomatic shunt fractures in TBM, especially in children. Hopefully this case report can shed light on a new perspective for readers to consider when dealing with the case.

CONCLUSIONS

There are no guidelines defining surgical decision making in asymptomatic patients with shunt fractures. Practitioners can still consider conservative therapy with routine observation, with the possibility that the patient can or has achieved a shunt-free state. However, given the risk of worsening hydrocephalus, surgical management even before neurologic symptoms become apparent is equally safe and feasible. The authors prefer to consider surgical management as the primary treatment for asymptomatic shunt fractures in children with a history of tuberculous meningitis, without waiting for a clinical improvement in ICP to appear.

DISCLOSURE OF ETHICS

Written informed consent was obtained from the patient for publication of this case report and accompanying images. Medical data had been de-identified before publication to ensure patient confidentiality.

REFERENCES

1. Tully HM, Dobyns WB. Infantile hydrocephalus: a review of epidemiology, classification and causes. *Eur J Med Genet* 2014; 57: 359–368.
2. Narayan VB. Treatment of hydrocephalus: Challenges and the way ahead. *Indian J Anaesth* 2014; 58: 456–457.
3. Krishnan SR, Arafa HM, Kwon K, et al. Continuous, noninvasive wireless monitoring of flow of cerebrospinal fluid through shunts in patients with hydrocephalus. *npj Digit Med* 2020; 3: 29.
4. Erol FS, Ozturk S, Akgun B, et al. Ventriculoperitoneal shunt malfunction caused by fractures and disconnections over 10 years of follow-up. *Child's Nerv Syst ChNS Off J Int Soc Pediatr Neurosurg* 2017; 33: 475–481.
5. Park M-K, Kim M, Park K-S, et al. A Retrospective Analysis of Ventriculoperitoneal Shunt Revision Cases of a Single Institute. *J Korean Neurosurg Soc* 2015; 57: 359–363.
6. Hanak BW, Bonow RH, Harris CA, et al. Cerebrospinal Fluid Shunting Complications in Children. *Pediatr Neurosurg* 2017; 52: 381–400.
7. Lee L, Low S, Low D, et al. Late pediatric ventriculoperitoneal shunt failures: A Singapore tertiary institution's experience. *Neurosurg Focus* 2016; 41: 1–9.
8. Hersh DS, Kumar R, Klimo P, et al. Hydrocephalus surveillance following shunt placement or endoscopic third ventriculostomy: a survey of surgeons in the Hydrocephalus Clinical Research Networks. *J Neurosurg Pediatr* 2021; 28: 139–146.
9. Vinchon M, Fichten A, Delestret I, et al. Shunt revision for asymptomatic failure: surgical and clinical results. *Neurosurgery* 2003; 52: 346–347.

10. Mullanfiroze K, Shah I. Hydrocephalus in tuberculous meningitis--how fast does it develop? *Scandinavian journal of infectious diseases* 2014; 46: 475–477.
11. Sharma D, Shah I, Patel S. Late onset hydrocephalus in children with tuberculous meningitis. *Journal of family medicine and primary care* 2016; 5: 873–874.
12. Rajshekhar V. Management of hydrocephalus in patients with tuberculous meningitis. *Neurol India* 2009; 57: 368–374.
13. Raut T, Garg RK, Jain A, et al. Hydrocephalus in tuberculous meningitis: Incidence, its predictive factors and impact on the prognosis. *J Infect* 2013; 66: 330–337.
14. Solari L, Soto A, Agapito JC, et al. The validity of cerebrospinal fluid parameters for the diagnosis of tuberculous meningitis. *Int J Infect Dis* 2013; 17: e1111–e1115.
15. Langmoen IA, Lundar T, Vatne K, et al. Occurrence and management of fractured peripheral catheters in CSF shunts. *Child's Nerv Syst ChNS Off J Int Soc Pediatr Neurosurg* 1992; 8: 222–225.
16. Clyde BL, Albright AL. Evidence for a patent fibrous tract in fractured, outgrown, or disconnected ventriculoperitoneal shunts. *Pediatr Neurosurg* 1995; 23: 20–25.
17. Schoeman J, Donald P, van Zyl L, et al. Tuberculous hydrocephalus: comparison of different treatments with regard to ICP, ventricular size and clinical outcome. *Dev Med Child Neurol* 1991; 33: 396–405.
18. Güneş A, Uluca Ü, Aktar F, et al. Clinical, radiological and laboratory findings in 185 children with tuberculous meningitis at a single centre and relationship with the stage of the disease. *Ital J Pediatr* 2015; 41: 75.

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CONFLICT OF INTEREST

The Authors declare no conflict of interest.

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