Case Report

Extrusion of Ventriculoperitoneal Shunt Cathether Through a Herniotomy Wound in an Infant: Case Report and Review of Literature

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ABSTRACT

Cerebrospinal fluid (CSF) diversion through the insertion of ventriculoperitoneal shunt (VPS) is the standard treatment for hydrocephalus. Many complications have been reported following VPS insertion. A case of extrusion of a VPS through a herniotomy wound is reported in a 4-month old male infant, who had a herniotomy performed on him by a General Practitioner (GP) for scrotal swelling. The extruded VPS was first externalized to the chest to aid healing of the groin wound and maintain CSF diversion. Later the VPS was removed to aid sepsis control. Although the parents did not comply with follow up instructions following their request for discharge, the patient remained well during telephonic communication 7 weeks after discharge. The case demonstrated higher risk for abdominal complications in the setting of background spina bifida and presence of intraperitoneal catheter. The need for optimal management of such complications by appropriate specialists cannot be over emphasised.

KEYWORDS: *Herniotomy, infant, sepsis, shunt extrusion, ventriculoperitoneal shunt*

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INTRODUCTION

Hydrocephalus is a common condition that frequently occurs in infants. It undergoes a chronic course and therefore requires continual medical surveillance and surgical intervention for maintenance of optimal quality of life.[1] Increasing prevalence have been demonstrated for hydrocephalus due to increasing survival of preterm infants with post-haemorrhagic hydrocephalus.[1] The therapeutic choice for hydrocephalus is the placement of ventriculoperitoneal shunt.[2,3] Despite the availability of endoscopic third ventriculostomy (ETV), VPS insertion is still a common procedure,[4] and more so in resource poor settings like ours. Various complications are associated with ventriculoperitoneal shunt such as blockage, migration, extrusion out of the body, over drainage of CSF and infection.[1-4] Some of these complications can become burdensome to the neurosurgeon as well as life threatening to the patient. This case of extrusion of the peritoneal catheter through a herniotomy wound is presented to highlight the peculiar challenges of management in our practice environment.

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CASE REPORT

A 4-month-old male infant, was admitted on account of extrusion of a ventriculoperitoneal shunt through a right groin wound in November 2017. The patient was first seen and admitted on the 4th day after birth with a diagnosis of sacral spina bifida which was repaired the following day. On the 3rd post-operative day the anterior fontanelle became full and tense. Computed tomography (CT) scan of the brain was performed on the 4th post-operative day and revealed a non-communicating hydrocephalus [Figure 1]. On the 5th postoperative day, a Chabra medium pressure VPS was inserted through a right occipital approach. He made very good recovery, was discharged home, but the parents were non-compliant with outpatient appointments. Rather the patient presented as an emergency at 12 weeks after discharge with complaints of extrusion of the VPS catheter from the right groin

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after undergoing bilateral herniotomy. A scrotal swelling of gradual onset was noticed 8 weeks after discharge home, which became painful, distressing, not associated with fever or vomiting. This necessitated urgent medical attention at a nearby hospital to their residence, where a diagnosis of hernia was made by the attending General Practitioner (GP). Despite being informed of the presence of a VPS in the abdomen, the GP reassured the parents and performed bilateral herniotomy. On the 3rd day after the herniotomy, the shunt tube was seen protruding from the right herniotomy wound, necessitating urgent referral to the Neurosurgeon same day.

On examination, the patient was awake and alert, pale and afebrile (36.9º) with a sunken anterior fontanelle. There was bilateral oblique groin incision wounds with non-absorbable skin sutures in-situ, and with an extruded shunt tube from the right groin wound [Figure 2a]. There was drainage of clear CSF from the distal fenestrations of the extruded shunt tube. The scrotum was swollen [Figure 2a] and translucent with both testicles palpable. The abdomen was soft and non-tender. He was admitted and urgent routine blood analysis showed anaemia of haemoglobin concentration 7.0g/dl, and blood transfusion was administered for optimization. There was leucocytosis (high total White Blood Cell count (WBC)) of 22.2 × 10^9/L (4.0-10.0) and high lymphocyte count of 53.6% (20–40%) but reduced neutrophil count of 35.7% (50-70). He was commenced on broad spectrum antibiotics (3rd generation cephalosporin) including metronidazole and analgesics. Partial wound break down of the right groin wound was noticed on the second day of admission, discharging serous fluid and by the 4th day, the wound completely broke down, and an urgent Paediatric surgical review recommended removing all sutures and daily wound dressing. The CSF drainage was excessive resulting in markedly sunken anterior fontanelle, hence the extruded shunt was clamped while the anterior fontanelle was closely monitored to guide release of the clamp whenever necessary. The total WBC and lymphocyte count remained high, while the neutrophil count remained low throughout the admission of the patient as follows: 5th day of admission (total WBC-12.4 × 10^9/L, lymphocytes-59%, neutrophil-38%); 8th day of admission (total WBC-13.2 × 10^9/L, lymphocytes-55.1%, neutrophil-32.6%); 13th day of admission (total WBC-12.6 × 10^9/L, lymphocytosis-68.6%, neutrophil-20%); and 22nd day of admission (total WBC-15.7 × 10^9/L, lymphocytes-67.1%, neutrophil-21.1%). On the 8th day of admission, the scrotal swelling had subsided, the left groin wound had healed while the right groin wound was clean and granulating [Figure 2b]. To aid healing of the right groin wound and maintain controlled CSF drainage, the extruded shunt was revised on the 9th day of admission by externalizing it at the chest wall [Figure 2c]. The extruded portion of the shunt tube was removed gently from the right groin wound after cutting the shunt tube at the lower chest wall through a small incision. Due to financial constraint to purchase a new VPS, a connector was used to reconnect the extruded distal shunt tube to the proximal portion of the shunt tube outside the chest. However, strict asepsis was maintained during the procedure in the theatre. On the 22nd day of admission, the right groin wound was now completely healed. The anterior fontanelle remained normotensive despite being clamped from the 4th day to the 23rd day of admission. To further evaluate the brain and ventricles, CT brain scan was performed and revealed moderately dilated occipital horns of the lateral ventricles but with no obvious pressure changes [Figure 3]. The VPS system was removed the following day, by disconnecting the extruded shunt tube from the connector; pulling down a few millimetres of the distal portion of the remaining proximal shunt tube within the chest wall, transecting it,

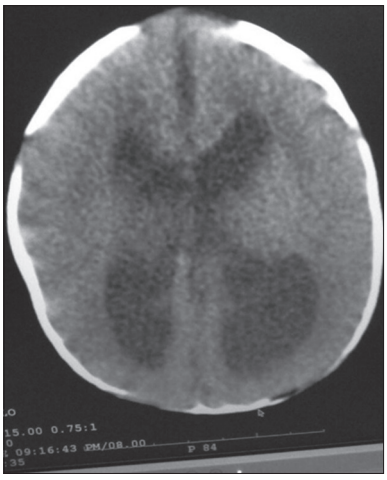

Figure 1: Axial section cranial noncontrast computed tomography scan performed at day 4 postspina bifida repair at first presentation showing dilated ventricles with effaced surface sulci

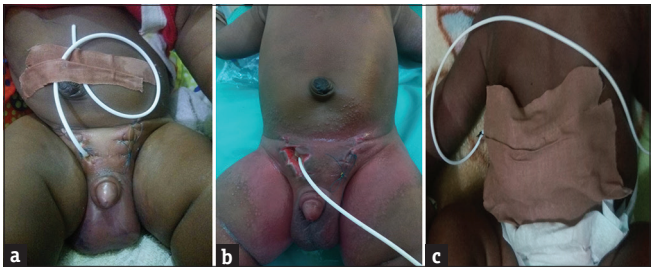

Figure 2: (a) Bilateral oblique groin incision wounds with nonabsorbable skin sutures *in situ* revealing extruded distal shunt tube from the right groin wound, and scrotal swelling at presentation. (b) Right groin wound breakdown although clean and granulating, and healed left groin wound at day 8 post admission. (c) Revision of extruded shunt tube by externalizing it at the chest wall at day 9 postadmission

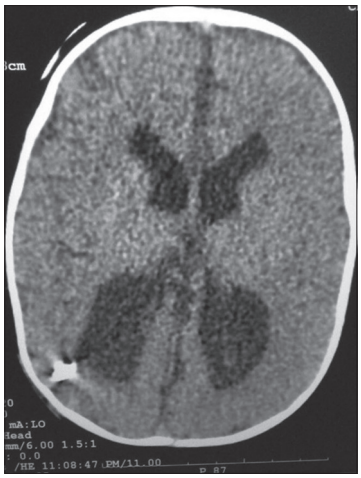

Figure 3: Axial section cranial noncontrast computed tomography scan after 20 days of clamping the extruded shunt tube showing moderately dilated occipital horns of the lateral ventricles but with no obvious pressure changes

while the remaining portions of the proximal shunt tube and ventricular catheter assembly were removed through exploration of the previous occipital surgical scar. The tip of the ventricular catheter and CSF obtained in a sterile container from the shunt tube were sent for microscopy, culture and sensitivity (m/c/s), which yielded no bacterial growth. Throughout the patient's admission, temperature remained relatively normal with the highest recorded temperature being 37.3º at 7th, 16th and 17th day post admission. The patient was discharged home 48 hours after removal of the VPS (26th day of admission) on request from the parents. He was placed on oral 3rd generation cephalosporin, with instruction for urgent neurosurgical consultation if the anterior fontanelle becomes full and tense. Also the parents were instructed on the need for weekly FBC until the WBC normalizes, after which the plan was to repeat the CT brain and re-insert the VPS. A repeat FBC was performed at another Laboratory a week after discharge, and showed a normalizing total WBC of 11.1 × 10^9/L (4-11 × 10^9/L), raised lymphocytes of 54.1% (20-46), reduced neutrophil of 33.9% (40–75), although these were much improved compared to previous results. The patient was well, all wounds were healed and the anterior fontanelle remained normotensive. However, the parents did not comply with further outpatient appointments, although the patient was said to be well during telephonic conversation with the mother at 7 weeks after discharge.

Discussion

Ventriculoperitoneal shunt (VPS) insertion remains the most common neurosurgical procedure performed worldwide for hydrocephalus, and aims at diverting CSF from dilated cerebral ventricles to the peritoneal cavity where absorption of CSF occurs.[4,5] Hydrocephalus may be communicating or non-communicating, and is associated with morbidity and mortality if there is delay in intervention. This is worse with non-communicating hydrocephalus, as in our patient. Complications of hydrocephalus related to VPS insertion are well known and may occur anywhere along its course from the cranial location in the ventricle, along its subcutaneous tract and at its distal location in the peritoneal cavity.[2,4-6] Although these VPS related complications remain a persistent problem in clinical practice, extrusion of components of shunt apparatus is very rare and an unusual complication of VPS insertion.[6-8]

About 25% of the complications caused by VP shunt insertion have been attributed to abdominal complications.[6,7] Common complications include mechanical obstruction of the distal peritoneal catheter by omentum or other intra-abdominal structures with resultant shunt failure, formation of abdominal pseudocyst, spontaneous bowel perforation, intestinal obstruction, inguinal hernia and development of liver abscess.[4,6,7] Rare abdominal complications include migration of the distal peritoneal catheter into the stomach, gallbladder, urinary bladder, liver, bowel, colon, scrotum and diaphragm.[4,6,7] However, spontaneous extrusion of VPS components is unusual and rare and have been reported in the chest wall, abdominal wall, through the mouth, anus, urethra and vagina.[4,6-10]

The causes of VP shunt extrusion have been considered under various hypotheses, and includes focal wound dehiscence and infection for early VPS extrusion.[6,8] Delayed VPS extrusion may be attributed to ischemic necrosis of dermis overlying shunt components, while other factors considered contributory to VPS extrusion include poor host immunity, factors related to surgical technique such as superficial shunt catheter placement during subcutaneous tunnelling, and bio reactivity of shunt components.[6,8]

In a case report, Ozveren *et al*.,[11] concluded that the development of scrotal swelling or hydrocele in a child with VP shunt should be recognized as a possible shunt complication. It appears therefore that the history of scrotal swelling 2 months after the VPS insertion in our patient may have been a complication of the VPS insertion. Panda *et al*.[5] reported that there is possibility of shunt malfunction if the VPS migrates into the scrotal sac, which may lead to secondary hydrocele and its sequelae. One of the explanations given for this complication is that peritoneal cavity distension due to draining CSF increases the intra-abdominal pressure thereby preventing the obliteration of the processus vaginalis,

and encouraging shunt migration and CSF collection in the scrotum.[5,11] Also spina bifida repair have been associated with intra-abdominal complications, and it has been reported that the repair of a large spina bifida defect may be another factor increasing intra-abdominal pressure besides the hydrocephalus.[11] The male gender of our patient makes him vulnerable to complications arising from the presence of a patent processus vaginalis (PPV) which may be either congenital or because of peritoneal CSF drainage.[5] Also the repair of his large spina bifida defect may have had an additional effect on increased intra-abdominal pressure. When such a complication is encountered, urgent intervention is recommended including bilateral herniotomy as the treatment of choice, with close observation for signs of obstruction in the immediate postoperative and follow-up periods.[5,11,12] In our patient, bilateral herniotomy was performed by a GP despite being aware of the presence of VPS in the patient, and with neither the parents nor the GP consulting the neurosurgeon for any input in the management. The GP reassured the parents and expressed his competence to perform the procedure. Bryant *et al*.[13] noted that an awareness of abdominal complications is necessary in creating an index of suspicion for the primary physician whose patient harbours a VPS and presents with abdominal symptoms. It is hoped that this case report will help to create this awareness in our environment and enhance the application of necessary precautions to prevent abdominal complications during management decisions.

The extrusion of the shunt tube through an intact right groin wound [Figure 2a] indicates poor surgical technique and calls into question the appropriateness of a GP to have performed the procedure. Ricci *et al*.[12] recommended obtaining General (in adults) or Paediatric surgical consultation in VPS-related groin complications. The standard surgical technique involves opening the hernia sac, and if the shunt tube is within the hernia sac, it should be repositioned in the peritoneal cavity; and thereafter the hernia sac transfixed and repair of the posterior wall performed.[5] Historically, it's common to perform herniotomy on the contralateral side as well.[5,11,12] However, where facilities are available, Ricci *et al*.[12] noted that it has now become standard practice to evaluate for a PPV on the contralateral side via laparoscopy, thus avoiding surgical exploration. They also noted that groin complications could be prevented by laparoscopically searching for a PPV at the time of the first VPS insertion.[12] If a PPV or an inconspicuous hernia is discovered, then a herniotomy should be proactively performed at the same operation.[12]

There are no general consensus on the acceptable mode of treatment for extruded VPS catheter. Teegala and Kota[4] noted that the management of these cases is difficult and needs to be individualized. They, however, reported that the standard method of treatment is removal of the extruded shunt system, control of infection, improvement of general condition followed by CSF diversion procedure.[4] Borkar *et al*.[6] made similar recommendation for immediate commencement of prophylactic antibiotics and complete removal of shunt assembly, if a shunt tube extrudes out of the body. Bansal *et al*.[7] removed the entire VPS system in their case of VPS tube extrusion through the anus in a child. The distal end of the catheter was pulled out through the anus after division of the shunt just below the reservoir to avoid catheter track contamination, and the ventricular end was sent for culture and sensitivity.[7] They also administered intravenous antibiotics in antimeningitic doses, closely observed the patient, inserted an external ventricular drain (EVD) when the anterior fontanelle started bulging on 2nd postoperative day, and planned re-insertion of the VPS when the culture result was received.[7] Teegala and Kota[4] in their management of 2 cases; anal migration and vaginal migration; removed the entire VPS system and offered ETV as the CSF diversion procedure. However, Rattan *et al*.[9] in their report of shunt tube extrusion through the urethra removed the shunt tube cranially through an incision in the post auricular area, under local anaesthesia. They considered their approach much simpler than other previously reported ones by identifying the chamber and pulling out the entire shunt assembly through the pre-auricular incision wound.[9] In contrast to other previously reported treatment approaches, a different approach was, however, utilized by Sami *et al*.[14] in their report of 3 cases of anal migration of VPS. Sami *et al*.[14] removed and replaced only the peritoneal catheter in 2 cases, while the entire VPS system was removed and replaced in the third patient who had meningitis.[14] It is to be noted that culture and sensitivity of the ventricular catheter or CSF were negative in most reports,[4,7,8,10,14] except in patients that presented with obvious meningitis.[4,14] It would seem then that removing the entire shunt tube is mandatory in cases of obvious meningitis at presentation, whereas in asymptomatic cases, the management can be individualized. Odebode[15] in a case of jejunal perforation and per oral extrusion of a peritoneal shunt catheter, removed the distal shunt per orally after transecting the tube flush with the jejunum.

In our patient, the challenges in the management included the persistently deranged WBC, the treatment of the right groin wound which completely broke down,

and ensuring controlled CSF drainage. In addition, the parents were financially constrained. Although broad spectrum antibiotics was given to prevent and or control sepsis, the persistently deranged WBC was difficult to explain. The questions considered were the likelihood of the neutropenia reflecting overwhelming sepsis, which could not be collaborated clinically as there were no signs of peritonitis or meningitis while the temperature pattern remained normal (there were no facilities for sepsis markers such as C-reactive protein or procalcitonin). The lymphocytosis may be considered as an immune response and may be responsible for the leucocytosis. The management was individualized for our patient by externalizing the extruded VPS to the chest to ensure controlled CSF drainage with on-going treatment for the groin wound to heal. A significant observation was that despite clamping the extruded shunt tube for 20 days before the entire shunt assembly was removed, the anterior fontanelle remained normotensive, and the patient remained well at the last communication with the parents 7 weeks after discharge home. Further follow up is desirable to know the state of the patient, but unfortunately follow up compliance has been poor. Our findings, however, may be similar to that of Borkar *et al*.[6] and Rattan *et al*.,[9] who managed their patients by removing the entire VPS system. There was no re-insertion of fresh VPS in their patients; one patient was well at last follow up visit 3 months post operatively and a repeat CT brain post operatively showed no hydrocephalus in both cases.[6,9]

Conclusion

The case demonstrated higher risk for abdominal complications in the setting of background spina bifida and presence of intraperitoneal VPS catheter. The development of scrotal swelling or hydrocele in a patient harbouring VPS should be recognized as a possible shunt complication. There is need for a high index of suspicion for such complications among GPs or attending doctors whose patients harbour a VPS and present with abdominal symptoms. The application of appropriate and optimal management decisions is recommended. Specialist (General or Paediatric) surgical consultation combined with neurosurgical input in VPS-related groin complications is vital to ensure application of optimal surgical technique. When shunt tube extrusion occurs, prompt attention should be instituted and the treatment individualized for the patient, based on clinical presentation and guided by the available resources at the centre.

Patient consent

Appropriate patient consent was obtained from the parents of the patient for permission to publish the case, and with the included images. The parents understand that the patient's names and initials will not be published and due efforts will be made to conceal his identity, but anonymity cannot be guaranteed.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understand that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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