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ACCEPTED CASE REPORT

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Authors:	Frederick Ejumudo, Edward Jolayemi, Onyeonoziri Achibiri, John Gbaji, Omotayo Ojo, Okezie O. Kanu
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CASE REPORT

Peroral extrusion of a ventriculoperitoneal shunt in an infant managed at a tertiary hospital in Lagos, Nigeria

Running title – Peroral extrusion of a VP shunt in Nigeria

Frederick Ejumudo[1], Edward Jolayemi[1], Onyeonoziri Achibiri[1], John Gbaji[1], Omotayo Ojo [1,2], Okezie O. Kanu*[1,2]

1. Department of Surgery Lagos University Teaching Hospital, Lagos State, Nigeria

2. Neurosurgery Division, Department of Surgery, College of Medicine, University of Lagos, Idi-Araba, Lagos State, Nigeria

***Corresponding Author**

Okezie Obasi Kanu, FWACS

Neurosurgery Division, Department of Surgery

College of Medicine University of Lagos

PMB 12003, Surulere, Lagos State, Nigeria

(drkanu@gmail.com)

Abstract

Ventriculoperitoneal (VP) shunts are commonly employed in the surgical treatment of hydrocephalus despite the expanding frontiers of neuroendoscopy. The risk of VP shunt failure is highest in the first year and is estimated to be between 11% and 25%. VP shunt- related morbidities may occur and these include rare complications such as gastrointestinal perforation by the shunt hardware and extrusion through natural orifices. We report the case of an 11-month-old male who presented five months post-VP shunt insertion with extrusion of the VP shunt hardware through the mouth. We discuss the presentation, investigations, management, and review relevant literature.

Keywords: hydrocephalus, ventriculoperitoneal shunt, peroral extrusion, intestinal perforation, Nigeria

Introduction

Ventriculoperitoneal (VP) shunt is the mainstay of surgical treatment of hydrocephalus despite advances in endoscopic surgery[1–3]. Shunt- related complications such as hardware obstruction, infection, pseudocyst formation and bowel perforation can occur. VP shunt failure rate has been estimated at approximately 11% to 25% within the first year of initial shunt placement with significantly higher number of shunt revisions and replacements among pediatric patients compared to adults.[4]

Bowel perforation by the shunt hardware is rare with a reported estimated occurrence of 1 per 100 to 10,000 VP shunt insertions. The VP shunt peritoneal catheter is extruded in about half of these cases[1].

Peroral extrusion of the peritoneal catheter of the VP shunt is an extremely rare complication. The authors describe a case of peritoneal catheter extrusion through the mouth.

Case presentation

Patient information

An 11-month-old male brought to the paediatric emergency room with history of one episode of non- bilious vomiting and consequent protrusion of a tube from the mouth. He had had VP shunt implanted five months prior to index presentation as treatment for non-communicating hydrocephalus. His mother gave a history of him being irritable. There was no history of fever or abdominal distension. The child currently stands and attempted to walk with support.

Clinical findings

At presentation, he was irritable, afebrile, and not dehydrated. His occipitofrontal circumference was 52cm and anterior fontanelle was open and soft. His vital signs were normal, and he had no neurologic deficits. An approximately 5cm length of distal VP shunt catheter was protruding from his mouth. The shunt was actively draining clear cerebrospinal fluid (Figure 1A). His chest was clinically clear and there was no sign of inflammation along the shunt tract. The abdominal scar was at the right paraumbilical region and had normal appearance His abdomen was soft, non- tender, and bowel sounds were normoactive.

Diagnostic assessment

A shunt series done demonstrated the peritoneal catheter looping back on itself and coursing through the posterior mediastinum to exit the mouth (Figure 1B, 1C). There was no fracture or disconnection of the shunt parts as the extruded part was actively draining CSF.

Therapeutic intervention

Parenteral antibiotics (Ceftriaxone and Vancomycin) were administered for 72 hours. He was placed on nil by mouth and following parental consent, we removed the entire shunt assembly in the operating room. The ventricular catheter was removed along with the reservoir via the cranial incision and the CSF sample collected at this point after dividing it below the reservoir chamber. The abdominal incision over the previous surgical scar was used to divide the peritoneal catheter and remove the subcutaneous part while the peritoneal catheter was removed from the mouth. The abdomen was monitored clinically with no symptomatology and signs of acute abdomen. He was commenced on graded oral feeds 24 hours post-shunt hardware retrieval which he tolerated without any complication. His abdomen remained soft and demonstrated no clinical signs of peritonitis.

Cerebrospinal fluid (CSF) analysis showed normal chemistry and culture-negative for (insert summary of biochemistry, microscopy, and negative culture).

Follow up and outcome

The parents were counselled for shunt replacement but opted for an approach of watchful waiting by serial measurements of his occipitofrontal circumference and they were counselled explicitly to look out for symptoms and signs of peritonitis and meningitis and to return to the emergency room if any of these should arise. He was subsequently discharge to the clinic and has remained stable on serial follow up with static occipitofrontal circumference (OFC).

Discussion

VP shunt insertion is the most performed surgical procedure for hydrocephalus[5]. The reported incidence of shunt extrusion attests to its rarity[6].

Common complications of VP shunt insertion include shunt hardware infection, shunt obstruction, abdominal pseudocyst, CSF ascites, bowel perforation, shunt migration, peritonitis, intra-abdominal abscess, shunt kinking and disconnection[7–9].

Several factors have been postulated to predispose patients with VP shunts to shunt-related gastrointestinal perforation. These include the thin bowel wall and weak intestinal musculature of children, chronic bowel irritation by the shunt hardware, stiff peritoneal catheters, use of abdominal trocars. Patients of younger age are also believed to be most affected due to their tendency to more vigorous peristaltic activity[10–12]. Other risk factors are male gender, malnutrition, silicon allergy, length of peritoneal catheter, shunt infection and previous abdominal surgery[13]

Upon bowel perforation, certain factors have been postulated to be responsible for the migration of the distal end of the catheter such as intestinal peristalsis, continuous water-hammer effect of CSF-transmitted arterial pulsations, and intermittent rise in the intra-abdominal pressure[6].

In our patient, the peroral extrusion of the distal end of the VP shunt occurred five months after its insertion and was most likely a sequelae of delayed bowel perforation by the shunt hardware as opposed to iatrogenic bowel injury at VP shunt insertion surgery which will present with acute features of localized or generalized peritonitis. Our patient is male and 11 months old, with no signs of malnutrition but the shunt was long enough to recoil and migrate through the oral cavity aided by peristaltic bowel motions. Young age as a risk factor for bowel perforation has been attributed to the weak intestinal musculature and stronger peristaltic activity in these group of patients[12]. The exact pathogenesis of the bowel perforation has remained elusive. Some authors have described the formation of an encasing fibrosis both at surgery and autopsy[21]. This fibrosis may have been induced by subtle silicon allergy. The encasing fibrosis may have had an anchoring effect on the shunt hardware with subsequent pressure on the bowel and eventual necrosis, leading to bowel perforation[34]. Forceful repeated retching and vomiting may cause a catheter that had perforated the gastrointestinal tract to undergo retrograde migrate to the oral cavity[35]. The formation of fibrosis along the tube or around the site of perforation may prevent the spillage of bowel contents into the peritoneal cavity thereby delaying presentation. Whereas shorter peritoneal catheter may prevent peroral extrusion, it does not prevent bowel perforations which may be concealed.

The diagnosis of bowel perforation may not be apparent if the shunt does not extrude to the exterior. The abdomen may be clinically silent as was the case in our patient.

Retrograde central nervous system (CNS) infection may ensue and become fully established thereby increasing the risk of morbidity and mortality. Ancillary radiological investigations such as shunt series are necessary to clinch the diagnosis when bowel perforation is suspected.

A review of published literature revealed 22 cases including this present case. This is summarized in Table 1. The age range was 6 months to 47 years with an average of 7.2years. The male to female ratio was 1:1. The average duration between procedure and complication was 2 years (3months to 10 years). The varied management protocols for this clinical condition are outlined in the table and the outcome was good in many of the cases with only overall mortality rate of 9%.[2,19,23,29,33,36] There remains no consensus on the treatment algorithm for these patients. Our patient was managed non-operatively and differed from the other studies in that the hardware device was removed via the previous cranial wound and through the mouth with comparatively good outcome

The management principles consist of prompt removal of the shunt hardware, CSF sampling for analysis and cultures, prophylactic broad spectrum antibiotic cover in the presence of signs of peritonitis, local shunt infection or meningitis. Expedited shunt revision is necessary in patients that are shunt dependent. Oral feeds should be restored as soon as possible if there is no contraindication.

Conclusion

Ventriculoperitoneal shunt insertion has remained the mainstay of management of hydrocephalus despite expanding frontiers in the field of neuroendoscopy. Bowel perforation is a rare complication and rarer still is the peroral extrusion of the peritoneal catheter through the mouth.

Prompt diagnosis of bowel perforation and shunt extrusion is required to mitigate the adverse effects of retrograde CNS infections and/or peritonitis. Transoral extrusion of shunt is a neurosurgical emergency that requires prompt clinical diagnosis and radiologic evaluation, removal of the whole shunt assembly under antibiotic cover, microbiology of the aspirated CSF, early commencement of oral feeds if signs of peritonitis are in abeyance, and shunt revision if the patient is shunt dependent and there is no contraindication to it.

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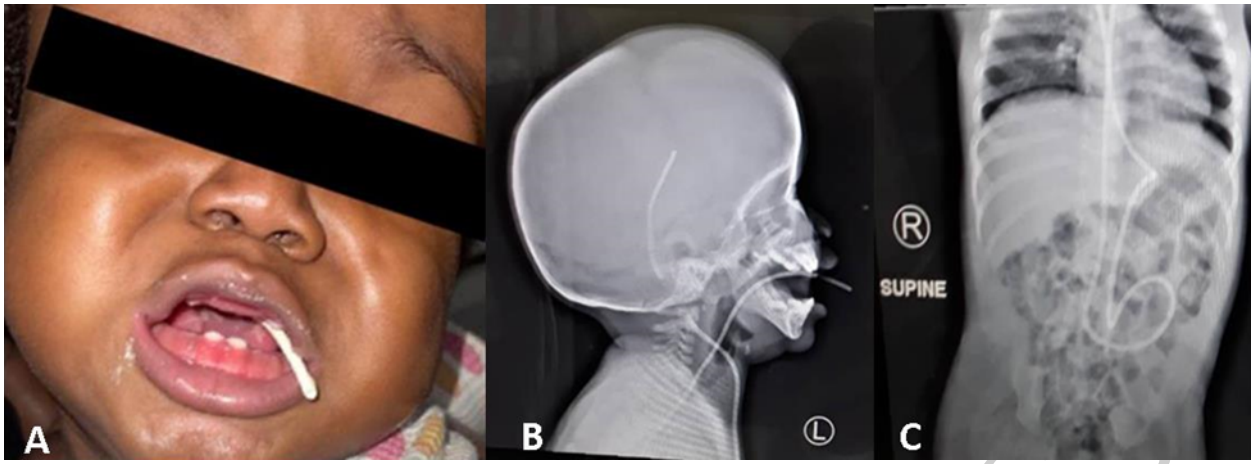

Figure 1: Clinical photograph and shunt series of an 11-month-old male with extruded VP shunt hardware in his mouth.

A: *The distal end of the peritoneal catheter is seen protruding from his mouth; B* ***and C:*** *Shunt series shows the course of the peritoneal catheter and its extrusion through the mouth.*

Table 1: A summary of published case reports of peroral extrusion of VP shunt

First author	Publication date	Country	Patient gender	Patient age	Time of diagnosis	Treatment	Outcome
Griffith JA[14]	1987	USA	F	9.5 year	3 months	Shunt exteriorization, IV antibiotics and venticuloatrial position	Deceased
Danismend [15]	1988	USA	F	9.5 year	10 months	Laparotomy + VA shunt	Alive
Fermin [16]	1996	Indonesia	F	8 months	6 months	Laparotomy	Alive
Park[17]	2000	Korea	F	5 years	4 years	Externalization of peritoneal shunt + upper GI endoscopy removal	Alive
Jimenez[18]	2001	Spain	F	5 years	1 year	Unexplained	Alive
Kothari [19]	2006	India	M	1.5 year	17 months	Incision behind the ear + shunt removal	Alive
Murali [20]	2008	India	M	6 years	5.5 years	Externalization + shunt removal	Alive
Odebode TO[21]	2007	Nigeria	F	15 months	6 months	IV antibiotics, laparotomy/shunt removal and subsequent VP shunt re-insertion	Alive
Berhouma [22]	2008	Tunisia	M	2 years	15 months	Externalization	Deceased
Sridhar K[23]	2009	India	F	8 months	6 months	Shunt removal, NPO, IV antibiotics	Alive
Sinnadurai M[24]	2009	India	F	27 years	2 weeks	Shunt externalization, IV antibiotics and distal shunt revision	Alive
Low [25]	2010	Singapore	M	1 year	11 months	Shunt removal + external ventricular drain	Alive
Dua R[26]	2011	India	M	8 months	7 months	Proximal shunt exteriorization/removal, IV antibiotics, NPO and shunt re-insertion	Alive
Agarwal [27]	2011	India	M	1 year	8 months	Shunt removal through the mouth	Alive
Gupta M[28]	2012	India	M	4 years	3years 6months	Shunt removal and IV antibiotics	Alive
Kundal [29]	2012	India	M	7 years	1 year	Shunt removal through the mouth	Alive
Yilmaz [30]	2013	Turkey	F	47 years	10 years	Laparotomy	Alive
Gupta R [2]	2014	India	M	11 years	10 years	Shunt removal through incision behind the ear	Alive
Mandhan [31]	2015	New Zealand	F	11 years	-	Laproendoscopic shunt removal + UGI tract endoscopy	Alive
Ghritlaharey [32]	2015	India	F	2 years	1 year	Externalization + shunt removal through the mouth	Alive
Fauzi [33]	2017	Indonesia	M	5 years	1 year	Shunt removal through the mouth	Alive
Present case	2023	Nigeria	M	11 months	6 months	Proximal shunt removal through the previous cranial incision + distal shunt removal through the mouth	Alive