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Case Report

Upward migration and peroral extrusion of a peritoneal shunt catheter: Case report and review of the literature

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Abstract

Background: Various complications after ventriculoperitoneal (VP) shunt surgery have been reported, but peroral extrusion of peritoneal catheter is an extremely rare complication, and only 20 cases have been reported since 1987. The pathophysiology still remains unclear and the management is challenging.

Case Description: A 5-year-old boy presented with a catheter coming out of his mouth. The boy had a posterior fossa tumor surgery and had VP shunt insertion 1 year earlier. Clinical signs and imaging studies showed that the distal end of the catheter had perforated the gaster and migrated upward and extruded through the mouth. Emergency removal of the shunt and proper treatment were done and he made uneventful recovery.

Conclusion: Peroral extrusion of VP shunt is extremely rare. Clinicians should be aware of this complication. With early diagnosis and proper management, the prognosis for good recovery is excellent, with only two deaths being reported in the literature. Complication of shunt extrusion is difficult to avoid, but knowing the risk factors, pathophysiology and proper management will decrease the morbidity and mortality of such cases.

KeyWords: Management, patophysiology, peroral extrusion, risk factors, VP shunt

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INTRODUCTION

Ventriculoperitoneal (VP) shunt is the most widely used procedure to treat hydrocephalus.[19] VP shunt surgery is associated with a high rate of complications (24–47%), including infection, obstruction, cerebrospinal pseudocyst, bowel perforation, and shunt migration.[18,21,22] Bowel perforation, albeit rare, is a serious complication which can sometimes lead to a fatal outcome.[2,17,19] The incidence of bowel perforation is reported to be 0.1–0.7%

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of all peritoneal shunting procedure,[15] with the most common site of perforation being the colon (70%), followed by the stomach (16%) and small bowel (14%).[17] Extrusion of the peritoneal catheter occurred in about half of the cases of bowel perforation.[15] Extrusion of the catheter may occur in any natural orifices, the most common being through the anus (61.9%) or not at all (31.4%).[17] Cases with peroral extrusion of the peritoneal catheter is very rare, and most commonly associated with gastric perforation.[5] We describe here a case report and review of the literature for all reported cases of peroral extrusion of a VP shunt catheter. To our knowledge, there were 21 cases of peroral extrusion of the peritoneal catheter that have been reported in the literature since 1987, including the present case.

CASE HISTORY

A 5-year-old boy presented with peroral extrusion at the distal end of a VP shunt catheter. The boy had been diagnosed by magnetic resonance imaging as having posterior fossa tumor and hydrocephalus when aged 4 years. A surgery was performed and VP shunt catheter was inserted using a Chhabra-slit-in-spring silicone shunt system. After the surgery, the patient remained well with the exception of mild gait disturbance. One week before admission to our hospital, the boy complaint of upper abdominal discomfort with emesis. In the following day, he regurgitated and severed peritoneal catheter exiting through the mouth.

Examination

At the time of admission, the boy was afebrile and fully conscious. We found no evidence of meningitis or increased intracranial pressure. The abdomen was soft and bowel sounds were normal. There was no sign of inflammation along the shunt tract. The peritoneal catheter was found extruding from his mouth [Figure 1]. There was no flow of cerebrospinal fluid (CSF) from the end of the catheter, which meant that an obstruction had occurred in the shunt catheter system. Laboratory results indicated no evidence of infection or any other abnormality. Head computed tomography scan showed no enlargement of the ventricles and the ventricular catheter was in proper position [Figure 2a]. Skull X-ray showed the peritoneal catheter coming up through the pharynx and extruded through the mouth [Figure 2b]. Chest X-ray revealed the migration of the peritoneal catheter into the stomach and esophagus [Figure 3].

Treatment

The boy underwent emergency shunt removal. During intubation the distal catheter was seen coming out of the esophagus. Surgical incision was made in the previous scar in the scalp and in median abdomen. The distal catheter was cut at the abdomen site before entering the peritoneal cavity. The ventricular catheter

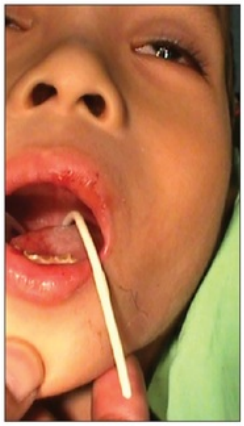

Figure 1: Pretreatment photograph during intubation

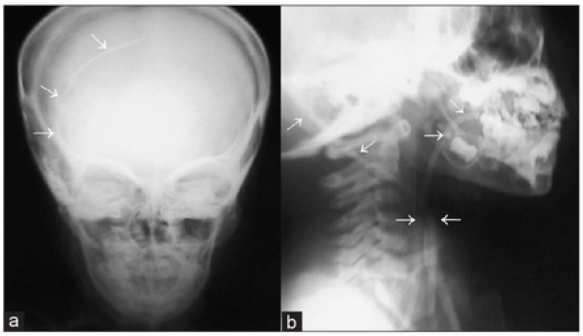

Figure 2: Anteroposterior and lateral films of the skull showing the position of the ventricular catheter (a) and the presence of the distal catheter in the pharynx and mouth (b)

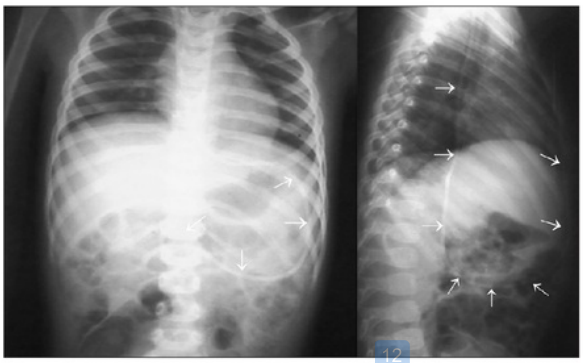

Figure 3: Chest film showing the upward migration of the distal catheter into the stomach and esophagus

was disconnected from the chamber, and there was a flow of CSF from the ventricular catheter. Analysis of the CSF did not reveal any sign of infection. The ventricular end and the chamber were removed through the scalp incision, and part of the distal catheter under thoracic tract was removed through the median abdomen incision. The distal catheter in peritoneal cavity, which had perforated the stomach wall, was removed easily

by pulling out through the opening of the mouth. We removed the whole catheter and observed the patient for 3 days. External drainage was not performed. The boy was nil peroral and maintained on intravenous fluids and antibiotics for 3 days. During the observation there was no sign of meningitis or peritonitis. There was no signs and symptoms of increased intracranial pressure, which meant the boy had become shunt independent, thereby no replacement shunt system was required. After 5 days of treatment, the boy was discharged in satisfactory condition and is currently doing well.

DISCUSSION

We conducted literature search of all cases of peroral extrusion of a VP catheter shunt via PubMed and MEDLINE, and identified 20 cases (excluding the present case) of peroral extrusion of a VP shunt catheter, which are summarized in Table 1. Eleven (52.4%) patients are females and 10 (47.6%) are males; 19/21 patients (90.5%) are children aged below 12. The mean patient age is 7 years and 5 months. The youngest patient age documented is 8 months old and the oldest is 47 years old. Duration between catheter placement and peroral extrusion range from 3 months to 10 years. Most of the major complaints was vomiting in 11 (52.38%) cases, shunt out of the mouth suddenly in 4 (19.04%) cases, abdominal pain in 1 case, and also respiratory distress in 1 case with a perforation of the trachea. Perforation site is the gaster in 15 (71.42%) cases, jejunum in 1 (5%) case, gastroesophageal junction in 1 (5%) case, trachea in 1 (5%) case, and the rest (19%) is unidentified probably because no open surgery or no good radiological study to visualize or predict the site of perforation was performed. Outcome is more favorable when there is no accompanying CSF infection or peritonitis. Peritonitis and meningitis markedly increase mortality rate.[5] According to our literature review, all patients are alive including the present case except for two cases where both had an accompanying CSF infection.

Pathophysiology

According to our case literatures review, all had a delayed presentation, which meant that the perforation is caused by a chronic process rather than acute injury (e.g., during the procedure). An important part of the pathophysiology of perforation is local inflammation and repeated pressure on the bowel wall. Inflammation leads to formation of an encasing fibrosis, which anchored the catheter to the serosal surface of the bowel wall. The site of catheter adherence to the bowel wall is then subjected to repeated pressure, due to the "pushing" effect of intestinal movements,[6] leading to the development of ulcer, and eventual perforation.[18,19] Bowel perforation may or may not lead to extrusion of the catheter, whereas when it did occur, mostly with downward migration that occur in accordance to direction of normal peristalsis, upward migration and extrusion through the oral orifice is very rare. Peroral extrusion of the catheter required it to move retrogradely against normal peristaltic movement across the gastroesophageal junction toward the oral cavity, which may be due to abnormal peristalsis[8] or bulk movement upwards caused by repeated vomiting episodes.[1] Sites of perforation also occurred in the upper part of the gastrointestinal system. From the literatures report, found in nearly all cases, site of perforation occurred in gaster; only two cases with unusual site of perforation is in the trachea and in the jejunum. In cases of perforation of the trachea, it is quite interesting because the catheter penetrates the diaphragm, enter the thoracic cavity, and finally into the trachea. The most distal perforation site of the gastrointestinal tract in oral extrusion cases is jejunum. In case of jejunum perforation, physiologically, the catheter should be pushed downward in accordance to the intestinal peristalsis. This phenomenon could be because of the position and direction of catheter penetration and decrease of peristalsis that leads to contra-mechanism movement, thereby resulting in upward migration. In our case, based on the radiological results, perforation is in the gaster as reported in most cases, with migration and peroral extrusion have been occurred.

Risk factors

The etiology of migration and extrusion of a VP shunt catheter has not been fully understood. From the literatures that we have reviewed, we identified the risk factors associated with extrusion of the peritoneal catheter and divided them into internal (ensuing from the host) and external factors. Internal factors include: (1) age, (2) nutritional status, (3) bioreactivity, (4) previous abdominal surgery and (5) chronic immobilization. Younger age has been mentioned as a prominent risk factor for bowel perforation in much literatures,[17,18,20] with children aged 10 years or less constituting 70.1% of bowel perforation cases and the male to female ratio is 3:2.[17] Plausible theory for this occurrence might be because children had weaker bower musculature and more vigorous peristaltic activity.[18] Organ penetration by catheter is also facilitated by malnutrition.[8] Out of the cases we reviewed, 3/21 (14.3%) patients presented with malnutrition.[1,8,10] Malnutrition may also occur as a result of recurrent emesis caused by the presence of the peritoneal catheter in the esophagus and gaster. Bioreactivity, such as silicon allergy[3] or mechanical irritation of the bowel wall by the catheter tip,[1,14] subsequently leads to local inflammation and perforation. Scarring or adhesion from previous abdominal surgery has been mentioned as a risk factor.[2,8] Two reported cases (9.5%) had history of previous abdominal surgery.[11,14] Three (14.3%) cases presented with myelomeningocele,[2,5,15] which has been suggested as a risk factor for bowel perforation due

Table 1: Summary of the literature review of cases with peroral extrusion of a ventriculoperitoneal shunt catheter

No	Author and year	Sex	Age	CC*	Duration*	CSF Infection	Peritonitis	Site of perforation	Previous shunt revision	Management	Outcome	Device	Other findings
1	Griffith *et al*., 1987[8]	F	9,5 year	Fever, Headache	3 months	Meningitis	-	Gaster	Twice	Exteriorized + VA shunt	Deceased	Raimondi coil spring	Shunt track infection
2	Danismend *et al*., 1988[4]	F	1.5 year	Diarrhea	10 months	-	-	Gaster	Three times	Laparotomy + VA shunt	Alive	Raimondi coil spring	-
3	Fermin *et al*., 1996[6]	F	8 months	Severe Cough	6 months	-	-	Trachea/ diaphragm	-	Laparotomy	Alive	Holter valve soft tubing	-
4	Park *et al*., 2000[18]	F	5 year	Abdominal pain	4 years	-	-	Gaster	-	Externalization of peritoneal cath + Upper GI endoscopy removal	Alive	Codman Silicone Catheter	Shunt tract infection
5	Jiménez Moya *et al*., 2001[11]	F	11 year 2 months	Shunt extrusion	1 year	-	-	Gaster	-	Unexplained	Alive	-	Previous surgery with appendicitis gangrenous
6	Kothari *et al*., 2006[12]	M	1.5 year	Shunt extrusion	17 months	-	-	Gaster	-	Incision behind the ear+shunt removal	Alive	-	-
7	Murali *et al*., 2008[16]	M	6 year	Shunt extrusion	5,5 years	-	-	Gaster	-	Externalization + shunt removal	Alive	-	-
8	Odebode 2007[17]	F	15 months	Shunt extrusion	6 months	-	-	Jejunum	-	Laparotomy + shunt removal	Alive	Chhabra slit spring silicone	Shunt tract infection and skin necrosis
9	Berhouma *et al*., 2008[2]	M	2 year	Vomiting	15 months	Meningitis	-	-	-	Externalization	Deceased	-	Myelomeningocele
10	Sridhar *et al*., 2009[21]	F	8 months	Vomiting	6 months	-	-	Gaster	-	Shunt removal through the mouth	Alive	-	Neonatal meningitis
11	Sinnadurai *et al*., 2009[20]	F	27 year	Coughing	15 years	-	-	Gaster	Twice	Shunt removal through the mouth	Alive	-	Arachnoid cyst
12	Low *et al*., 2010[14]	M	1 year	Vomiting	11 months	-	-	Gaster	Once	Shunt removal + external ventricular drain	Alive	-	Exomphalos major post-operative peritonitis
13	Dua *et al*., 2011[5]	M	8 months	Vomiting	7 months	-	-	Gaster	-	Externalization shunt removal via mouth	Alive	Chhabra slit spring silicone	Myelomeningocele
14	Agarwal *et al*., 2011[1]	M	1 year	Vomiting	8 months	-	-	Gaster	-	Shunt removal through the mouth	Alive	-	Malnourished

Contd...

Table 1: Contd...

No	Author and year	Sex	Age	CC*	Duration*	CSF Infection	Peritonitis	Site of perforation	Previous shunt revision	Management	Outcome	Device	Other findings
15	Gupta *et al*., 2012[9]	M	4 year	Vomiting	3,5 years	-	-	Gaster	-	Shunt removal through incision behind the ear	Alive	-	-
16	Kundal *et al*., 2012[13]	M	7 year	Vomiting	1 year	-	-	-	-	Shunt removal through the mouth	Alive	-	Post meningitis
17	Yilmaz *et al*., 2013[22]	F	47 year	Recurrent headache	10 years	-	-	Gaster	Once	Laparotomy	Alive	-	Pseudotumor cerebri
18	Gupta *et al*., 2014[10]	M	11 year	Vomiting	10 years	-	-	Gastro-esophageal junction	Four times	Shunt removal through incision behind the ear	Alive	Chhabra slit spring silicone	Malnourished
19	Mandhan *et al*., 2015[15]	F	11 year	Vomiting	-	-	-	Gaster	-	Laparoendoscopic shunt removal + UGI tract endoscopy	Alive	-	Myelomeningocele
20	Ghritlaharey 2015[7]	F	2 year	Vomiting and cough	1 year	-	-	-	-	Externalization + Shunt removal through the mouth	Alive	Chhabra slit spring silicone	-
21	Present case	M	5 year	Vomitting	1 year	-	-	Gaster	-	Shunt removal through the mouth	Alive	Chhabra slit spring silicone	-

*CC: Chief complaint, Duration: From operation to extrusion

to neurogenic weakness of the bowel wall. In our case, age factor seems to be a very influential factor on the occurrence of perforation and migration of shunt tube.

External factors include: (1) surgical error, (2) infection, (3) shunt type, and (4) shunt length. Perforation caused by surgical error is mainly associated with an acute presentation, which was not shown in any of the cases we reviewed. Surprisingly, from our review of the literature, no cases presented clinically with peritonitis, and 2 out of 21 (9.5%) presented with CSF infection, which led to a grave outcome as both patients are deceased. A proposed explanation for the low number of infection is a protective mechanism by the fibrous encasement of the catheter which prevented extension of infection from the bowel to the peritoneal cavity. Then again, many of the cases we reviewed have gaster as the site of perforation, which had a lower number of bacterial colonization compared to other sites of the bowel, such as the colon. Infection of the shunt tract itself may contribute through a mechanism which is similar to silicone allergy or mechanical irritation, eventually leading to local inflammation by means of foreign body reaction.[14,15] Stiff, hard tipped, sharp, long, or spring coiled type of catheter has been associated with increased bowel perforation risk.[1,5,17–19] However, the rigid and hard catheters are not the only cause. From the literature, it was observed only two (9.5%) cases used Raimondi coil spring catheter,[4,5] five (23.8%) cases (including the present case) used Chhabra-slit-spring silicone catheter,[5,7,10,17] and one (5%) case used Holter valve with soft tube.[6] The rest did not mention the type of shunt that was used. Longer distal catheter length is also mentioned as a risk factor.[14] In our case, there were no signs of abdominal infection or CNS infection. We also use a soft silicone shunt that is relatively safe against perforation of the abdominal organ. The time of the incident is also one year after surgery, so the surgical error factor can be eliminated.

Management

From the literature we have reviewed, our recommendation for the principles of treatment for shunt catheter perforation with peroral extrusion is: (1) emergency removal, (2) appropriate antibiotic therapy, (3) nil per oral, (4) reinsertion (if necessary). Emergency removal can be performed by open laparotomy, endoscopy, or pulling the catheter manually through the mouth. Earlier cases tend to choose open laparotomy as a method of removal, because laparotomy aided in visualization of any opening in the bowel caused by perforation that might need primary closure.[17] However, many authors suggested that invasive procedures such as laparotomy are not necessary because any opening in the bowel caused by perforation is small and should seal off spontaneously.[15] Before removal, the proximal and distal part of the catheter is divided, and then the proximal part is externalized or removed while the distal part is removed either through the extrusion site or proximally through the site of division. Disconnecting the ventricular from the peritoneal catheter further decreased the chance of infection, because there is no contact of the contaminated tube with neither the peritoneum nor the shunt tract.[1] Kothari[12] performed removal of the shunt through an incision behind the ear, which is not recommended, because of possibility of contamination from pulling the distal catheter through the peritoneal cavity.[16,18,20] Recently, minimally invasive laparoscopy has replaced laparotomy in cases of bowel perforation. Mandhan[15] did upper gastrointestinal endoscopy to assess entry point of the perforating catheter, then proceeded with laparoscopy to remove it. Pulling the distal catheter through the mouth has been the most frequent option of removal in the cases we reviewed. Pulling the catheter through the mouth is also the choice of removal management in our case. We postulated that the fibrous encasement surrounding the catheter play a role in sealing off the perforation site, which occurs when pull through is performed. In our opinion, an open laparotomy is indicated if there are signs of peritonitis, failed or was detained when pulling through or acute injury following improper operating procedures.

Infection control is an important part of the management of this complication. Our case emphasizes the importance of confirming early presence of infection. Broad spectrum antibiotics that cover the intestinal flora should be started at the time of admission. The risk of contamination during removal of the catheter should be minimized, by performing removal of the catheter with minimally invasive procedure and under antibiotic cover.[21] Postop antibiotics should also be given as prophylaxis.

Keeping the patient nil per oral is necessary for the healing process in bowel perforation cases. Recommendation for nil per orally management in our case is three days for recovery of the bowel. Several previous reports, recommendations for fasting after treatment vary greatly, mostly between 2 and 4 days.[1,9,10,13,16,21] In two cases, fasting lasted until 7 to 14 days.[5,15] All the literatures reported that no abdominal problems occurred posttreatment.

The patient might not need a replacement shunt because they might have become shunt independent, probably because their CSF pathway has already healed from the time of shunt placement or the primary cause has been corrected.[21] This is shown in our patient, because he showed signs of catheter obstruction but no sign of increased intracranial pressure. It can be concluded that he had become shunt independent. If the patient is asymptomatic after his shunt is removed, then he probably does not need shunt reinsertion.

Outcome

From the literature we get on the case of VP shunt complications with peroral extrusion; almost all the

outcome is good. Only two cases of deaths were reported as a result of complication of meningitis. With proper management, the case could provide an optimal outcome with minimal morbidity.

CONCLUSION

Upward migration and peroral extrusion of VP shunt is extremely rare. Clinicians should be aware of this complication with early diagnosis and proper management. The best management should be emergency shunt removal by pulling the distal catheter through the mouth and prevention of ascending infection. In most of the cases, the prognosis for good recovery is excellent, with only two death being reported in the literature. The mortality cases were associated with complication of meningitis. Complication of shunt extrusion is difficult to avoid, but knowing the risk factors, pathophysiology and proper management will decrease the morbidity and mortality of such cases.

Declaration of patient consent

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

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Conflicts of interest

There are no conflicts of interest.

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