International Surgery Journal
Ghritlaharey RK. Int Surg J. 2024 Jul;11(7):1168-1175
http://www.ijsurgery.com
pISSN 2349-3305 | eISSN 2349-2902

DOI: https://dx.doi.org/10.18203/2349-2902.isj20241614

Case Report

Cerebrospinal fluid shunt catheter extrusion through the mouth in a child: a case report with brief literature review

Rajendra K. Ghritlaharey*

Department of Pediatric Surgery, Gandhi Medical College and Associated, Kamla Nehru and Hamidia Hospitals, Bhopal, Madhya Pradesh, India

Received: 15 May 2024
Accepted: 07 June 2024

***Correspondence:**
Dr. Rajendra K. Ghritlaharey,
E-mail: drrajendrak1@rediffmail.com

ABSTRACT

Extrusion of the distal end of the cerebrospinal fluid (CSF) shunt catheter through the mouth is a rare complication of shunt surgery performed to treat hydrocephalus. A two-year-old girl had her first CSF shunt inserted when she was six months old to treat congenital hydrocephalus. The shunt became infected 2-months later and had to be removed. A few weeks later, a new CSF shunt was inserted on the left side. One year later, she presented with an extrusion of the distal end of the CSF shunt catheter through her mouth. However, she showed no symptoms or signs of peritonitis or meningitis. Her entire CSF shunt system on the left side required removal. A cranial magnetic resonance imaging (MRI) scan showed ventriculomegaly and a new CSF shunt catheter was implanted on the right side. She had an uneventful postoperative period and was doing well during the follow-up. Extrusion of the distal end of the CSF shunt catheter through the mouth is rare and more likely to occur in children, and clinicians need to be aware of such complications.

Keywords: Case report, Children, Complication, Extrusion, Gastric perforation, Hydrocephalus

INTRODUCTION

The procedure of diverting cerebrospinal fluid (CSF) from the ventricular system to the peritoneal cavity through a ventriculoperitoneal (VP) shunt catheter is widely accepted as a surgical treatment for hydrocephalus.[1,2] The VP shunt catheter insertion is frequently performed worldwide to treat the hydrocephalus caused by various reasons, across all age groups.[1-3] However, it has been found that children are more susceptible to VP shunt complications and requirements for shunt revisions than adults.[4-6] There have been reports of the migration of VP shunt catheters within hollow organs such as the intestine, urinary bladder, and uterus, which may or may not lead to the extrusion of the distal end through the natural orifice.[7] Peroral extrusion of the distal end of a peritoneal catheter is a rare complication. According to a literature review, there have been only 32 reported cases of this complication.[8] This report describes a new case of a two-year-old girl who presented with the extrusion of the distal end of her VP shunt catheter through her mouth. The successful management of this case is reported here, following the "CARE guidelines".[9]

CASE REPORT

A 2-year-old girl was admitted to the department of pediatric surgery with the chief complaint of extrusion of her implanted distal end of a CSF shunt catheter, accompanied by a brief episode of vomiting. Her history included that she was investigated for progressive head enlargement at the age of 6 months. A cranial computed tomography (CT) scan revealed communicating hydrocephalus (Figure 1) and a right-sided CSF shunt catheter was implanted. However, her CSF shunt got infected after 2-months and required removal. A new left-sided CSF shunt was inserted a few weeks later. One year later, she presented with an extrusion of the distal end of

the CSF shunt catheter through her mouth. She was well and conscious. Clinical examination detected that the distal end of her implanted CSF shunt catheter was protruding from her mouth (Figure 2). Her CSF shunt was functioning well and draining clear CSF. Clinical signs of peritonitis and meningitis were absent.

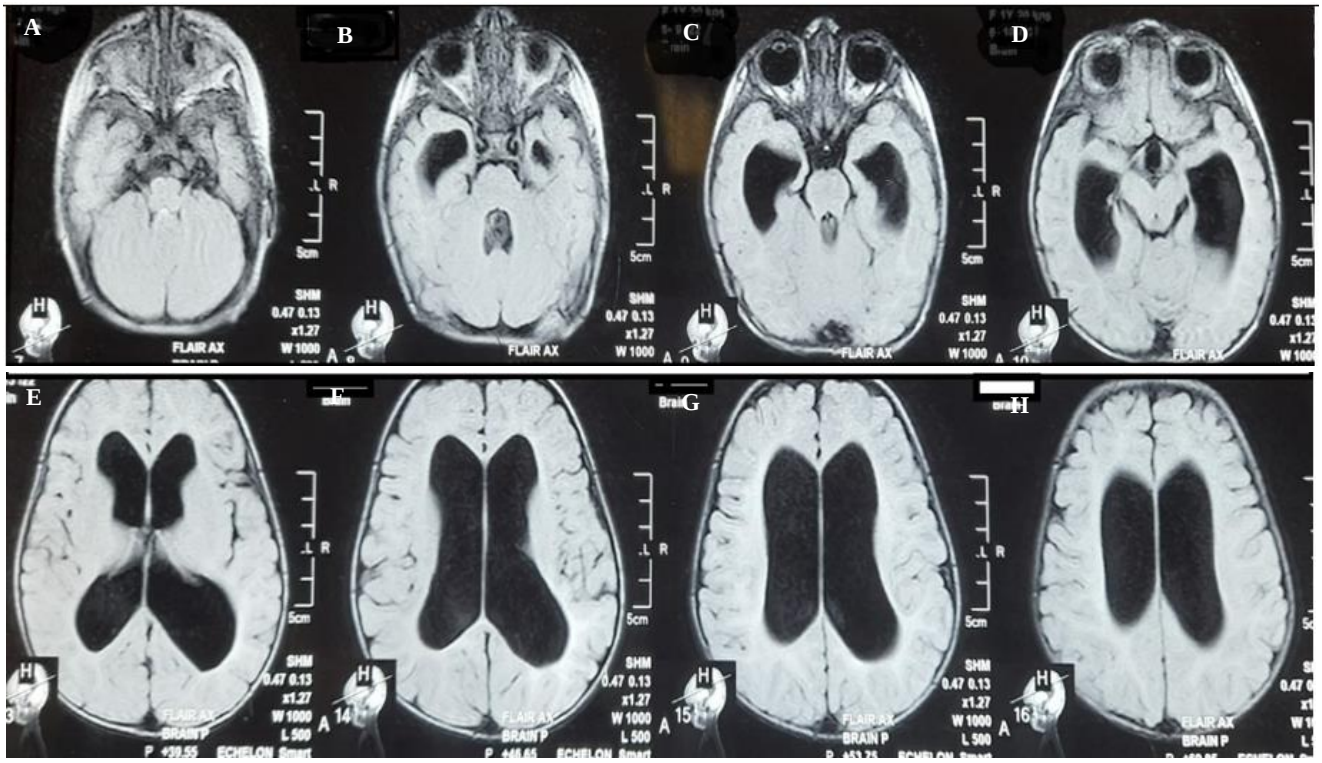

Figure 1 (A-H): Cranial computed tomography scan of the child showing ventriculomegaly (hydrocephalus).

Figure 2: Clinical photograph of the child showing the perorally extruded distal end of her cerebrospinal fluid shunt catheter.

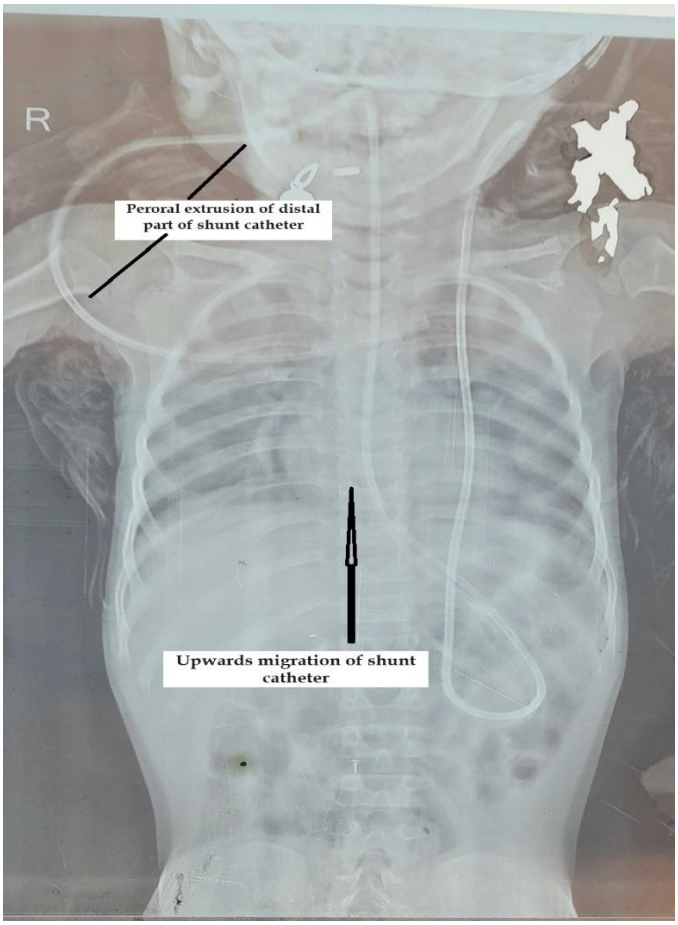

Figure 3: Plain X-ray of child's head, chest and abdomen showing upward migration and peroral extrusion of the distal cerebrospinal fluid shunt catheter.

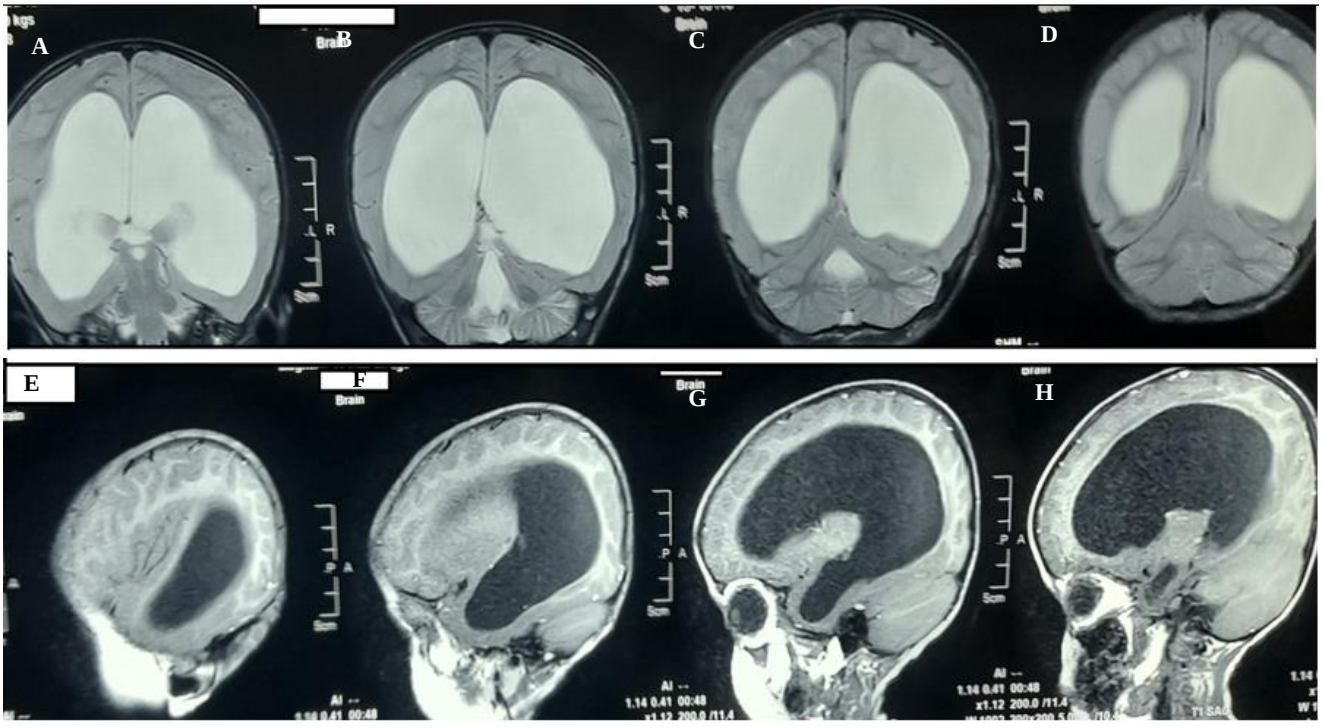

Figure 4 (A-H): Cranial MRI with the contrast of the child showing grossly dilated ventricular system with thinning of the brain parenchyma.

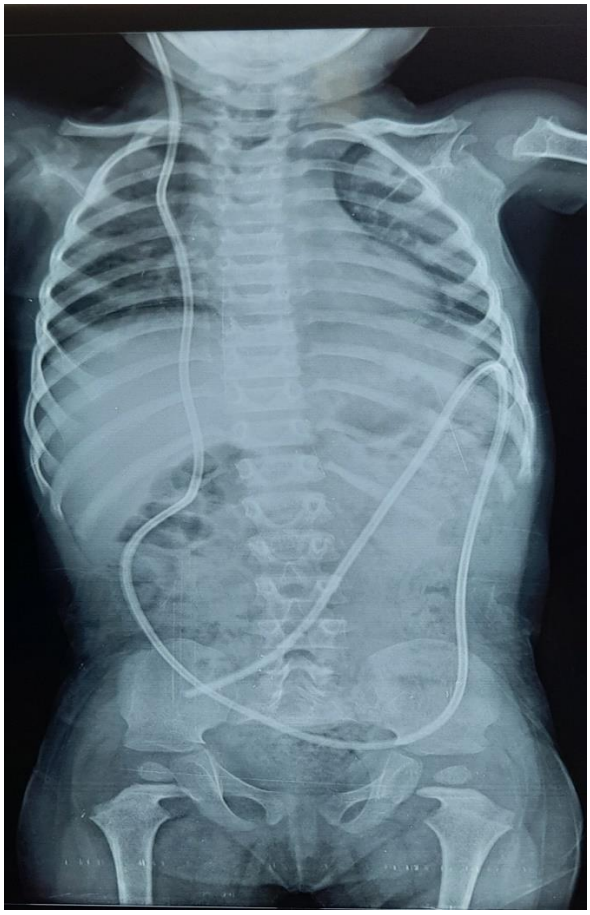

Figure 5: Follow-up X-ray of the neck, chest and abdomen showing the normally positioned shunt catheter and the distal end located in the right lower quadrant of the abdomen.

While managing the per-orally extruded distal shunt catheter X-rays of the chest and abdomen including the head were obtained. The X-ray showed continuity of the shunt system, upward migration probably after perforation of the stomach, and extrusion of the distal part of the peritoneal catheter per-orally (Figure 3). The clinical diagnosis was transoral extrusion of distal CSF shunt catheter, which is one of the rare complications of CSF diversion carried out to treat hydrocephalus. Her entire CSF shunt catheter required removal for the treatment of complication, mentioned above.

Before the right-sided CSF shunt was inserted, the patient underwent an abdominal ultrasonography which showed normal results. A plain abdominal X-ray was also taken, revealing no haziness or air-fluid levels. In addition, a cranial magnetic resonance imaging (MRI) scan was acquired before the shunt insertion, which showed significant ventriculomegaly and thinning of the brain parenchyma, as seen in Figure 4.

A summary/timeline of surgical procedures executed for the present case is detailed in Table 1. Her postoperative period was uneventful and she was doing well in the follow-up done 4 months after the VP shunt revision. An X-ray of the neck, chest, and abdomen was done in the follow-up period that was showing the normally positioned shunt catheter without fracture/break and the distal end located in the right lower quadrant of the abdomen (Figure 5). Recently a photograph and video of the child were provided by the parents, showing a dramatic improvement in her clinical condition. She was advised to have regular follow-ups in OPD.

Table 1: Summary (timeline) of surgical procedures executed for the present case.

Month/year	Age (months)	Surgical procedure executed	Institution	Operating surgeon
April 2022	6	Right-sided VP shunt catheter insertion	Private nursing home	Another surgeon
July 2022	9	Entire shunt system removed for shunt/shunt tract infection	Private nursing home	Another surgeon
September 2022	11	Left-sided VP shunt catheter insertion	Private nursing home	Another surgeon
November 2023	25	Removal of part of distal/per-orally extruded VP shunt catheter and proximal converted as EVD	Author's institution	Another surgeon
December 2023	26	Revision of peritoneal part/distal VP shunt catheter	Author's institution	Another surgeon
December 2023	26	Entire shunt system removal for shunt tract infection	Author's institution	Another surgeon
January 2024	27	Right-sided VP shunt catheter insertion	Author's institution	Corresponding author
May 2024	31	Doing well in follow-up period and advised for regular follow-ups in OPD	Author's institution	Corresponding author

EVD=external ventricular drainage; VP=ventriculoperitoneal

DISCUSSION

It is known that the distal VP shunt catheter can migrate into the intestine, urinary bladder, and uterus, with or without extrusion to the exterior through the natural orifices, which is a known and possible complication. The colon is the commonly perforated area followed by the urinary bladder and uterus.[10-12] Peroral extrusion of the distal end of a peritoneal catheter is a rare complication of CSF diversion used to treat hydrocephalus.[8] In 1980, Nishijima et al reported the first case of perforation of the stomach by a distal VP shunt catheter with migration of part of it within the gastric lumen without peroral extrusion.[13] Griffith et al reported the first historical case of migration of a VP shunt catheter into the stomach with extrusion of the distal end per-orally in 1987.[14]

Table 2: Summary of the literature review (peroral extrusion of the distal end of ventriculoperitoneal shunt catheter, n=35).

Description	Total (%)
Number of cases	35
Sex distribution	
Male	20 (57.14)
Female	15 (42.85)
Male to female ratio (20/15)	1.33:1
Age (at the time of transoral extrusion)	
Infants	10 (28.57)
13-24 m	8 (22.85)
25-36 m	2 (5.71)
37-60 m	6 (17.14)
61-120 m	3 (8.57)
11-15 years	3 (8.57)
16-years and above/adults/older age	3 (8.57)
Interval	
0-6 m	12 (34.28)
7-12 m	14 (40)
13-24 m	4 (11.42)
25-60 m	3 (8.57)
6-10 years	2 (5.71)
Chief complaints	
Trans-oral extrusion of distal catheter	35 (100)
Clinical signs	
Peritonitis	Nil
Meningitis	Nil
Radiological investigations	
X-rays series	35 (100)
Cranial CT/MRI scan	12 (34.28)
Abdominal CT/MRI scan	3 (8.57)
Surgical procedures executed (n=35)	
Removal of entire VPS without EVD	17 (48.57)
Removal of entire VPS with EVD	4 (11.42)
Removal of part of distal VPS and proximal catheter converted as EVD	10 (28.57)
Removal of part of distal VPS catheter and relocation in peritoneal cavity	2 (5.71)
Removal of distal VPS with others	2 (5.71)
Shunt revision (n=23)	
Delayed re-VPS catheter insertion	13
Delayed VPS revision	2
Delayed ventriculo atrial shunt	2
Immediate re-VPS/VPS revision	6
Result/outcome	
Recovered	33 (94.28)
Mortality	2 (5.71)

CT=Computed tomography, EVD=external ventricular drainage, MRI=magnetic resonance imaging, m=months, VPS=ventriculo-peritoneal shunt

According to a recent systematic literature review, there have been a total of n=34 cases of trans-oral extrusion of the distal VP shunt catheter with two more cases published recently.[8,15,16] During the literature search six more cases of transoral extrusion of VP shunt were identified but were excluded from the present literature review for various reasons, such as incomplete details.[17-21] Two children had the distal end of the shunt perforate the diaphragm, ascend upwards through the trachea, and protrude out per-orally and were excluded from the review.[17,18] Four cases of trans-oral extrusion of the distal VP shunt catheter were also excluded due to incomplete case details.[19-21]

A systematic literature review published in 2023 confirmed that the peroral extrusion of VP shunt catheters was more frequently found in males than females.[8] In addition to the present case, there were 20 males and 15 females resulting in a male-to-female ratio of 1.33:1.[8,15,16] A summary of a brief literature review is provided in Table 2. The summary provided in Table 2 is an extract of my work, a systematic literature review to which three new cases were added including the present case.[8] In Table 2, the results obtained and the details are interpreted in numbers and percentages. Including the present case, out of 35 cases, 32 were children.[8,15,16] At the time of clinical presentation and diagnosis, more than half of them (n=18; 51.42%) were children aged two years or below. At the time of clinical presentation and diagnosis, more than a quarter of the cases (n=10; 28.57%) were infants. Upon further review, it was discovered that trans-oral extrusion of the distal CSF shunt catheter is less frequent in adults/older age. Only three such cases were reported in adults/older age.[8]

The literature review revealed that more than four-fifths (n=30; 85.71%) of cases showed symptoms within 24 months of VP shunt insertion or revision. Three-fourths (n=26; 74.28%) of cases were diagnosed within 12 months of VP shunt insertion or revision. The main symptom observed was the transoral extrusion of the VP shunt catheter's terminal end which was accompanied by vomiting in most cases. However, there were no clinical signs supporting meningitis or peritonitis. The diagnosis in the aforementioned/reviewed cases was confirmed by the presence of a distal VP shunt catheter that was extruded through the mouth. Various radiological investigations were ordered including plain X-rays, USG abdomen, and CT/MRI scans of the head and abdomen. Plain X-rays were the primary radiological investigation ordered for all cases, while cranial CT/MRI scans were also advised/obtained as supported by a literature review in 12 cases (34.28%). CT/MRI scans of the abdomen were less frequently advised for the management of per-orally extruded VP shunt catheters.

The surgical therapy offered for cases of per-orally extruded VP shunt catheter includes: the removal of the entire catheter with or without external ventricular drainage (EVD) and the removal of part of the distal VP shunt while converting the proximal part into an EVD. Sixty percent (n=21) of the cases were treated with the removal of the entire VP shunt catheter. Out of these, n=17 of them were treated without the insertion of an EVD while the remaining n=4 cases were managed with the insertion of an EVD. In n=10 cases, the surgical therapy offered was the removal of part of the distal VP shunt while converting the proximal part into an EVD. On reviewing the literature, it was found that repair of perforation of a hollow viscus including the stomach was not required in most cases and heals spontaneously after the removal of the shunt catheter.[8,15,16] The authors repaired the perforation site only for three cases. In the cases of trans-oral extrusion of the distal VP shunt catheter, subcutaneous surgical procedures were used to manage more than three-fourths (n=27; 77.14%) of cases. Exploratory laparotomy was only carried out in one-fifth (n=7) of the cases.

Shunt revision/re-VP shunt insertion, or conversion to a ventriculo-atrial (VA) shunt is a significant part of managing cases where the shunt catheter extrudes. Details of shunt revision, re-VP shunt insertion, or conversion to VA shunt were available for n=23 (65.71%) cases, while details were not available for n=4 (11.42%) cases. The literature review suggests that delayed re-VP shunt insertion was preferred over immediate revision or insertion during the postoperative period. In one-fourth of the cases, shunt revision/re-VP shunt insertion was not required either during the immediate postoperative period or after the late follow-up period.[8,16]

Surgical intervention usually results in improvement and mortality is rare, although it has been reported in the literature.[14,22] According to a literature review, there were 2 deaths (5.71%) during the management of per-orally extruded distal VP shunt catheters.[14,22]

Possible reasons for the intragastric migration, upward migration, and peroral extrusion of the distal end of the implanted CSF shunt catheter in present child include: during the first shunt revision, a shunt catheter was implanted on the left side, a plain X-ray provided by the parents showed that probably a whole length of the CSF shunt catheter was implanted, most probably, during the insertion of the peritoneal catheter within the peritoneal cavity, the most distal part of the peritoneal catheter was inserted in the close vicinity of the stomach, due to continuous pressure of the distal end of the shunt catheter (friction effect), a gastric perforation occurred, the intragastric migration of the distal part of the peritoneal catheter was the combined effect of the large intragastric space, and intraabdominal pressure, and once it was within the stomach, it was probably pushed upwards due to the reaction of the stomach for the peritoneal catheter, as a natural reaction to expel any intra-gastric foreign body.

The following are the predictors for this CSF shunt complication to happen: implantation of the CSF shunt catheter to the left side, although right-sided implanted shunt cases are not immune to this complication, use of mini-laparotomy or trocar for insertion of peritoneal

catheter within the peritoneal cavity, recurrent left upper abdominal discomfort, few months after the CSF shunt catheter implantation, and presence of distal end/distal part of the CSF shunt catheter in the left upper quadrant of the abdomen on radiological investigations.

The following are some of the suggestions to reduce the chances of the above-described CSF shunt catheter complication, especially in children: insertion of a shorter length of peritoneal catheter within the peritoneal cavity and use of laparoscopic technique for the insertion of peritoneal part within the peritoneal cavity. Regular and frequent follow-ups are a must as the literature supports that more than two-thirds of the CSF shunt complications especially extrusion of the distal end through the natural orifices occur within the first 12 months after the shunt insertion.[8,10-12]

CONCLUSION

Peroral extrusion of the distal end of a peritoneal catheter is a rare complication of cerebrospinal fluid diversion performed to treat hydrocephalus, mostly occurring in children and clinicians need to be aware of such complications. Cranial computed tomography or magnetic resonance scans can aid in decision-making about the shunt revision/re-VP shunt placement. Early surgical intervention is crucial for such cases, for achieving the best outcome.

ACKNOWLEDGEMENTS

Authors would like to thank the child's parents for providing clinical and few radiological photographs.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

REFERENCES

1. Greuter L, Ruf L, Guzman R, Soleman J. Open vs laparoscopic ventriculoperitoneal shunt placement in children: a systematic review and meta-analysis. Childs Nerv Syst. 2023;39(7):1895-902.
2. Naftel RP, Argo JL, Shannon CN, Taylor TH, Tubbs RS, Clements RH, et al. Laparoscopic vs open insertion of the peritoneal catheter in ventriculoperitoneal shunt placement: review of 810 consecutive cases. J Neurosurg. 2011;115(1):151-8.
3. Sanderfer VC, Arnold MR, Mulvaney GG, Wang H, McLanahan CS, Wait SD, et al. Outcomes of laparoscopic and open ventriculoperitoneal shunt placement. Am J Surg. 2024;227:123-6.
4. Paff M, Alexandru-Abrams D, Muhonen M, Loudon W. Ventriculoperitoneal shunt complications: a review. Interdisciplinary Neurosurg. 2018;13:66-70.
5. Busse LC, Dubinski D, Gessler F, Dinc N, Konczalla J, Czabanka M, et al. Retrospective comparison of long-term functionality and revision rate of two different shunt valves in pediatric and adult patients. Acta Neurochir (Wien). 2023;165(9):2541-9.
6. Merkler AE, Ch'ang J, Parker WE, Murthy SB, Kamel H. The Rate of Complications after Ventriculoperitoneal Shunt Surgery. World Neurosurg. 2017;98:654-8.
7. Harischandra LS, Sharma A, Chatterjee S. Shunt migration in ventriculoperitoneal shunting: A comprehensive review of literature. Neurol India. 2019;67(1):85-99.
8. Ghritlaharey RK. Migration of the distal ventriculoperitoneal shunt catheter into the stomach with or without trans-oral extrusion: A systematic literature review and meta-analysis. World J Clin Pediatr. 2023;12(5):331-49.
9. Riley DS, Barber MS, Kienle GS, Aronson JK, von Schoen-Angerer T, Tugwell P, et al. CARE guidelines for case reports: explanation and elaboration document. J Clin Epidemiol. 2017;89:218-35.
10. Ghritlaharey RK. Systematic literature review of the management of transanal extrusion of distal ventriculoperitoneal shunt catheter: 1966–2020. Med J DY Patil Vidyapeeth. 2022;15(5):629-59.
11. Ghritlaharey RK. Systematic review of migration of distal ventriculoperitoneal shunt catheter into the urinary bladder with or without per-urethral extrusion. Med J DY Patil Vidyapeeth. 2022;15(6):840-53.
12. Ghritlaharey RK. Management of trans-vaginal extrusion of the distal ventriculoperitoneal shunt catheter: a systematic literature review from 1973 to 2021. Int J Res Med Sci. 2021;9(11):3416-23.
13. Nishijima M, Ohyama H, Higuchi H. Gastric perforation in ventriculo-peritoneal shunt--a case report. No Shinkei Geka. 1980;8(7):679-83.
14. Griffith JA, DeFeo D. Peroral extrusion of a ventriculoperitoneal shunt catheter. Neurosurgery. 1987;21(2):259-61.
15. Chowdhury SI, Shirodkar A, Bala A, Shankar KB, Tanwar I. Peroral extrusion of ventriculoperitoneal shunt: an unusual complication with literature review. Int J Scientific Res. 2023;12(9):33-4.
16. Abdul Rahim H. Zwayed, Sreenivas AV, Shabana AM, Rraziek YA, Nasser A, Refaat BF, et al. Abnormal migration and extrusion of lower end of ventriculoperitoneal shunt into the mouth. MAR Neurol Neurosurg Psychol. 2023;7:4.
17. Fermin S, Fernández-Guerra RA, Sureda PJ. Extrusion of peritoneal catheter through the mouth. Childs Nerv Syst. 1996;12(9):553-5.
18. Tamlikha A, Shukri F, Zahari Z. Transoral migration of ventriculoperitoneal shunt: A rare presentation. GMJ. 2021;32:131-4.
19. Pant N, Singh S, Singh G, Kumar A, Rai RK, Rawat J, Wakhlu A. The wandering ventriculoperitoneal shunt and the scope of its salvage. Childs Nerv Syst. 2021;37(8):2613-8.
20. Faheem M, Jaiswal M, Prajapati HP, Kumar R, Saba Nu, Ojha BK. Unusual complications of

ventriculoperitoneal shunt surgery: Tertiary care experience. J Pediatr Neurosci. 2022;17(1):30-5.
21. Sikandar M, Sridhar R. A prospective study of ventriculoperitoneal shunt complications and identification of risk factors. Int J Acad Med Pharm. 2023;5(6):290-5.
22. Berhouma M, Messerer M, Houissa S, Khaldi M. Transoral protrusion of a peritoneal catheter: a rare complication of ventriculoperitoneal shunt. Pediatr Neurosurg. 2008;44(2):169-71.

Cite this article as: Ghritlaharey RK. Cerebrospinal fluid shunt catheter extrusion through the mouth in a child: a case report with brief literature review. Int Surg J 2024;11:1168-75.