




Review Article

Ventriculoperitoneal shunt extrusion in pediatric patients, clinical patterns and therapeutic strategies: A scoping review

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ABSTRACT

Background: Ventriculoperitoneal shunts (VPSs) are frequently employed in neurosurgery to treat hydrocephalus, with a particular focus on pediatric patients. Although VPSs are commonly utilized, they are not exempt from difficulties, such as shunt extrusion. The main aim of this study is to enhance comprehension regarding the occurrence, causes contributing to, and consequences of VPS extrusion in pediatric patients.

Methods: A comprehensive search approach was implemented, including electronic databases, including PubMed, Google Scholar, and Scopus, to locate pertinent articles published between January 1950 and May 2023. The utilization of keywords such as “ventriculoperitoneal shunt” and “extrusion,” “ventriculoperitoneal shunt” and “migration,” and “ventriculoperitoneal shunt” and “perforation” was employed. Data on patient demographics, underlying diseases, origin of extrusion, presenting symptoms, treatment, and follow-up were gathered. Statistical studies were conducted to identify potential risk factors connected with the occurrence of shunt extrusion.

Results: A study analyzed 80 studies on 120 individuals with extruded VPS catheters. The majority of patients (55.8%) had symptoms such as cerebrospinal fluid leakage and irritation. Hydrocephalus was categorized into congenital (40%), obstructive (36.7%), and communicating (11.7%) groups. Catheter extrusion sites varied, with most from the anal or rectal site. Preoperative meningitis or peritonitis was present in 20% of patients. Treatments ranged from shunt removal to endoscopic third ventriculostomy, resulting in a 90% recovery rate, 1.7% mortality, and 5% follow-up loss.

Conclusion: Extrusion of the distal catheter in VPSs is a critical medical situation that necessitates urgent surgical intervention. The presence of an infection raises the likelihood of complications; hence, it is vital to promptly address the issue through the administration of antibiotics and the replacement of the shunt. Timely intervention enhances results.

Keywords: Child, Children, Extrusion, Hydrocephalus, Neurosurgery, Pediatric, Perforation, Ventriculoperitoneal shunt

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INTRODUCTION

Ventriculoperitoneal shunts (VPSs) have evolved into a crucial and widely used neurosurgical intervention for managing hydrocephalus, a neurological disorder characterized by an abnormal cerebrospinal fluid (CSF) accumulation within the cerebral ventricles.^[41,59,61,100] These implantable devices are a lifesaving solution for people suffering from hydrocephalus. They are composed of a proximal catheter that is positioned within the cerebral ventricles and a distal catheter that is routed beneath the patient's skin, frequently following a subcutaneous pathway down the neck, chest, and abdomen before ending within the peritoneal cavity.^[5,17,88]

The primary function of these components is to allow excess CSF to drain away from the cranial vault, reducing intracranial pressure (ICP) and alleviating associated neurological symptoms. This intricate system relies on pressure differentials to allow CSF to flow from the cerebral ventricles, through the proximal catheter, along the subcutaneous path, and finally into the peritoneal cavity, where the body's natural mechanisms reabsorb it. The regulation of CSF flow is further facilitated by a valve mechanism, often located along the distal catheter or within the shunt's programmable unit, which can be adjusted to optimize CSF diversion based on individual patient needs.^[17,34]

Although VPSs have significantly transformed the management of hydrocephalus, it is crucial to acknowledge that intricacies and potential consequences accompany their utilization.^[14,80] Shunt component extrusion is a significant complication of shunt elements' unintentional movement or protrusion through anatomical structures.^[2] This poses considerable clinical difficulty in the management of hydrocephalus.^[102]

This review aims to provide a comprehensive analysis of shunt extrusion, encompassing its occurrence, underlying pathophysiological causes, clinical symptoms, diagnostic techniques, and the range of therapeutic options currently utilized.

MATERIALS AND METHODS

The aims and objectives of the study were carefully considered, and a protocol was written to reflect that. Methods for conducting the search, extracting data, synthesizing data, determining which should be included, evaluating the quality of the studies, and ultimately screening them were all documented in the protocol. This review follows the guidelines established by Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA).

All of the studies reported in the literature in English with participants younger than 18 years were considered for

inclusion. Patients older than 18 years old, studies that reported only the migration of the VPS without extrusion, studies that provided an overview, and studies conducted in languages other than English were not considered.

We identified a database of studies on the extrusion of VPSs in the pediatric population through various orifices by conducting a systematic review and searching the relevant literature using PubMed, Google Scholar, and Scopus. The search terms "ventriculoperitoneal shunt" and "extrusion," "ventriculoperitoneal shunt" and "migration," and "ventriculoperitoneal shunt" and "perforation" were employed.

To find the articles that met the systematic review's inclusion criteria, we screened their titles and abstracts and then read them in their entirety. The studies that met the inclusion and exclusion criteria were subjected to a thorough critical review by two reviewers, O.A. and Y.A. We first looked at the titles with keywords, then at the abstracts, and finally at the full texts of those that seemed relevant. At the full-text screening stage, all studies were reviewed by an independent reviewer, A.B., to ensure eligibility. Articles selected or included differently by each of the three reviewers were briefly discussed until a consensus was reached [Figure 1].

The third review resolved any discrepancies that emerged following critical analysis, while the first two reviews independently determined the risk of bias and carried out data extraction using a designed protocol by the Cochrane Handbook.^[55]

Statistical analysis was carried out using Microsoft Excel and the statistical software package Statistical Package for the Social Sciences 26. Categorical data were compared and tested for significance using the Chi-square test. Statistical significance was defined as $P < 0.05$.

RESULTS

Following a comprehensive screening process, a selection of 80 studies was undertaken, employing rigorous criteria during the initial search across reputable databases, including PubMed, Scopus, and Google Scholar. This extensive study analyzed patient-level data derived from a defined cohort comprising 120 individuals, all grappling with the challenging clinical scenario of the extruded distal end of the VPS catheter. Our age distribution revealed that patients within our study were administered their first shunt at a median age of 3 months, while 75% of participants were below 18 months [Figure 2]. Furthermore, the median interval between the initial shunt implantation and their presentation with shunt extrusion was calculated at 10 months, when 75% of patients presented with the extrusion in <24 months from the first VPS.

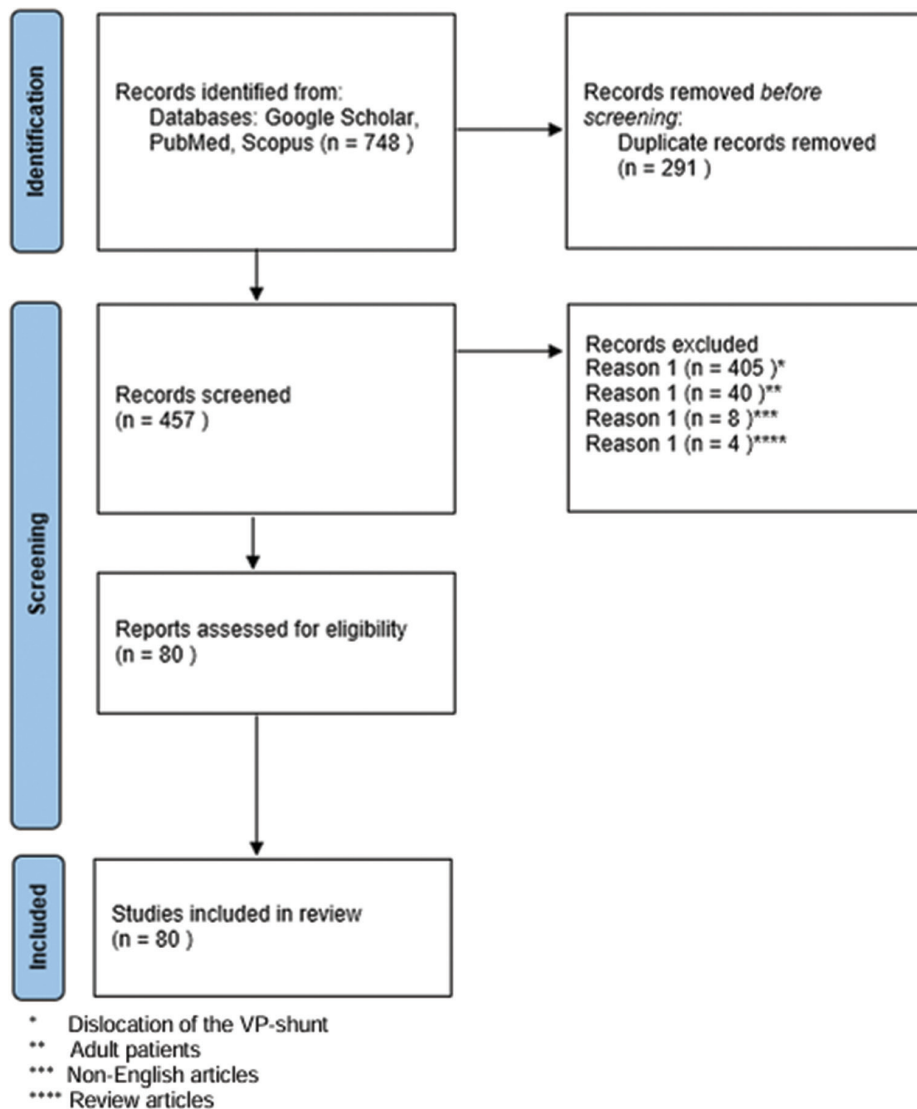


Figure 1: Flow diagram of articles included in the systematic review.

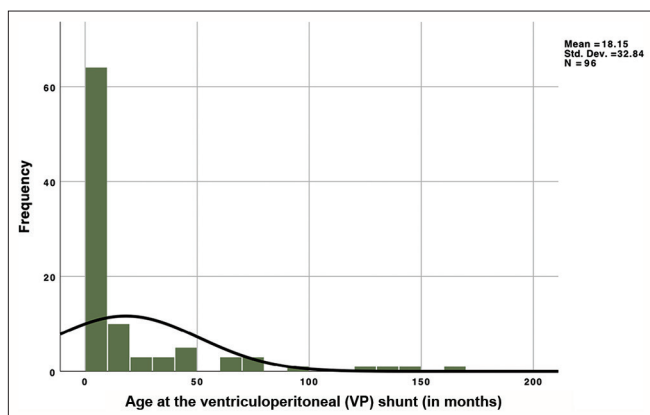


Figure 2: A histogram showing age by the first presentation to ventriculoperitoneal shunt surgery.

Gender analysis demonstrated that out of these individuals, 64 were unequivocally identified as male, constituting 53.3% of the total cohort. In contrast, 48 were unequivocally identified as females, accounting for 40% of the cohort, while the genders of the remaining 8 patients (6.7%) were unknown.

Remarkably, the gender of the remaining participants remained enigmatic, and our analysis disclosed no statistically significant gender preferences. It is essential to underscore that a substantial proportion of our cohort, precisely 67 patients, making up a significant 55.8%, presented with a spectrum of symptoms. These symptoms manifested diversely and were likely attributable to localized effects, including the leakage of CSF and irritation of the mucosal lining of the affected organ due to the infected

distal catheter. These localized effects often manifest as distressing symptoms such as vomiting, skin erosion, and purulent discharge. Conversely, some of the reported symptoms may have originated from systemic issues linked to VPS dysfunction or infection, resulting in altered mental status, fever, and sepsis. A comprehensive and thoughtful compilation of imaging findings for each individual was organized [Table 1].

The classification of hydrocephalus represents a pivotal facet of our study, and we categorized it into three principal groups for our statistical analysis: congenital ($n = 48$, 40%), obstructive ($n = 44$, 36.7%), and communicating ($n = 14$, 11.7%), while the type in the rest 14 patients (11.7%) was unknown. This deliberate categorization provides valuable insights into our study's broad spectrum of conditions.

The origin of catheter extrusion emerged as a significant aspect of our analysis. Our findings strongly suggest that in 44 patients (36.7%) of the cases, the catheter extruded from the anal or rectal site. Conversely, the remaining 17 patients (14.2%) experienced extrusion from the oral site, 14 patients (11.7%) extruded from the abdominal wall, 9 patients (7.5%) from the vagina, 8 patients (6.7%) from the urethra, 7 patients (5.8%) from the umbilicus, and 6 patients (5%) from an inguinal site. The remaining 12 patients (12.5%) exhibited extrusion from random sites [Figure 3].

An important variable in our study is the presence of preoperative meningitis or peritonitis in only 24 patients (20%). Our analysis found no statistically significant difference in peritonitis or meningitis presentation between patients presenting across the three primary extrusion sites (oral or rectal, abdominal wall, and urethra), with $P = 0.743$.

Irrespective of the presenting symptoms, all patients underwent invasive management procedures. The spectrum of treatments employed was diverse and included various approaches: 62 patients (51.7%) underwent the removal of the VPS with subsequent replacement during the same hospital admission; 32 patients (26.7%) had the VPS removed

exclusively; 11 patients (9.2%) underwent distal catheter revision alone; 4 patients (3.3%) underwent VPS removal followed by endoscopic third ventriculostomy (ETV); 3 patients (2.5%) underwent external ventricular drainage (EVD) without mentioning the further management; and 3 patients (2.5%) had the VPS removed and replaced with a ventriculoatrial (VA) shunt. The remaining 3 patients (2.5%) underwent alternative treatment management strategies. In addition, there was one patient for whom the therapy remained enigmatic [Figure 4]. After the management, as mentioned above, a noteworthy 108 patients (90%) made a complete recovery, while 2 patients (1.7%) regrettably succumbed to their conditions, and 6 patients (5%) were lost to follow-up.

DISCUSSION

The distal catheter of the VPS typically resides serenely within the peritoneal cavity, navigating its course amidst the labyrinthine folds of the intestines without causing any disruption. However, under certain predisposing conditions, it embarks on a transformative odyssey, migrating to atypical locations where it adheres and initiates an inflammatory cascade. Abdominal wall contractions can exert force on the catheter, propelling it into the encircling fibrous tract. This phenomenon may follow episodes of elevated intra-abdominal pressure or may result from anchoring to a calcified point along the catheter's path, thereby prompting its migration toward subcutaneous tissues.^[29,42,71,92] By adherence of the shunt distal end to the viscera or body wall, an inflammatory response will be provoked with the release of the cytokine interleukin-13,^[57] and chemotaxis of Th1 cells, CD4+ cells, macrophages, and dendritic cells leading to cytolysis and necrosis of the organ wall with gradual extrusion of the catheter.^[3] A firm type of catheter may provoke migration^[61] because using a softer catheter reduces the risk rate of migration.^[61] Poor immunity, wound dehiscence, and insufficient surgical technique can lead to extrusion^[25,32] [Figure 5].

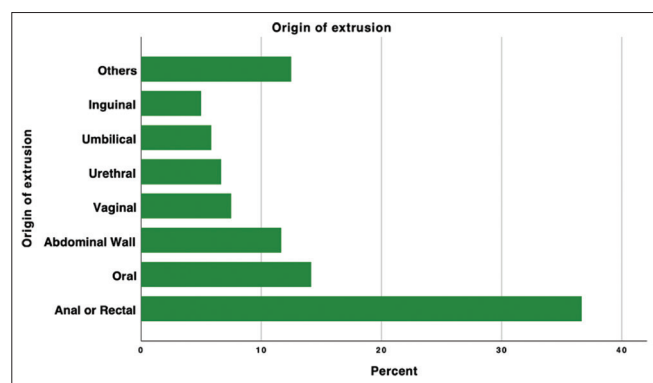


Figure 3: The distribution of the extrusion sites.

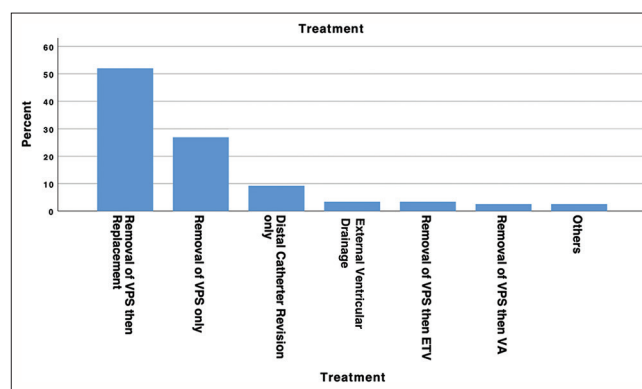


Figure 4: The treatment modalities.

Table 1: Patient characteristics of the related articles.

Reference Nr.	Investigator	Sex	Age at VPS Surgery (years)	Primary diagnosis	Interval to VPS Extrusion	(Preoperative) Meningitis Peritonitis	Previous One year Treatment	Presentation of Patient	Origin of extrusion	Treatment	Follow-up and outcome
1	Patel ^[85] 1973	F	At birth	Spina bifida-associated hydrocephalus	11 months	No	VPS	Vomiting, fever	Vaginal	Removal of VPS and then reinserted	Recovered successfully
2	Joubert ^[58] 1983	F	14 months	Communicating Hydrocephalous	1 year	No	VPS	No symptoms	Mid Lumbar region	Removal of VPS and reinserted in VA shunt	Recovered successfully
3	Aricó ^[13] 1985	F	2.5 year	Tri-ventricular Hydrocephalus	8 months	No	VPS	No symptoms	Anal	Removal of VPS and reinserted in VA shunt	Recovered successfully
4	Griffith ^[45] 1987	F	Unknown	Congenital hydrocephalus	Unknown	No	2 VPS	No symptoms	Oral	Removal of VPS and reinserted in VA shunt	Patient died due to acute brain stem herniation
5	González ^[43] 1987	Pt1: M Pt2: M	Pt1: 1 month Pt2: 10 years	Pt1: communicating hydrocephalus Pt2: aqueduct stenosis	Pt1: same time Pt2: 3 years	No (both)	VPS (both)	Pt1 and 2: protrusion through the anus	Pt1 and 2: anus	Pt1 and 2: the shunt was removed and replaced	Pt1 and 2: recovered successfully
6	Sridhar ^[96] 1988	F	2 months	Congenital hydrocephalous	6 months	Yes	NR	Repeated bouts of vomiting, no fever or irritability	Oral	Removal of shunt through a cranial incision	Satisfied follow-up progress
7	Prasad ^[87] 1995	M	6 years	Cerebellar medulloblastoma	2 years	No	VPS	Protrusion through the penis	Urethra	The shunt was removed	Recovered successfully
8	Ashpole ^[15] 1995	F	4 years	Aqueduct stenosis	3,5 years	No	VPS	Asymptomatic	Anal	Not mentioned	Recovered successfully
9	Fermin ^[87] 1996	F	8 months	Communicating hydrocephalus	6 months	No	VPS	Cough and respiratory distress	Oral passing through the trachea	The shunt was removed and reinserted	Recovered successfully
10	Naglic ^[76] 1996	F	49 days	Hydrocephalic malformation	5 months	No	VPS	Leaking of the CSF from the external genitalia	Between the right labia majus and minus	Resected and reinserted	Not mentioned
11	Digray ^[33] 2000	F	20 months	Congenital hydrocephalus	4 months	No	VPS	No symptoms	Anal	Removal of the shunt, external ventriculostomy, and antibiotics	Recovered successfully
12	Park ^[84] 2000	F	12 months	Congenital hydrocephalus	48 months	No	VPS	Abdominal pain, vomiting, regurgitation of the catheter during vomiting session	Oral	Removal of the peritoneal catheter after antibiotic courses	Went well for one year follow up
13	Gupta ^[50] 2004	M	5months	Congenital hydrocephalus	3 months	No	VPS	No symptoms	Anal	Reinsertion of VP shunt one month after peritoneal and ventricular catheter removal	Not reported
14	Ansari ^[12] 2005	M	20 months	Cervico-thoracic myelomeningocele	5.5 years	Meningitis	VPS	Fever, abdominal pain, frontal headache, 7dizziness, neck stiffness	Anus	External ventricular drain followed by shunt removal	Recovered successfully after follow-up for 6 months
15	Eser ^[36] 2006	M	First day	Congenital hydrocephalus	3 months	No	VPS	Abdominal distension and restlessness	Umbilical	Removal of VPS and then reinserted	Recovered successfully
16	Akcora ^[4] 2006	M	6 months ago	Obstructive hydrocephalus	6 years	No	VPS	Vomiting and abdominal pain	Anal	Removal of VPS	Discharge on the 10th postoperative day
17	Handa ^[52] 2007	Case1:F, case 2:M	18 months 5 year	Meningitis associated hydrocephalus Cong. hydrocephalus	Unknown	Frank infection No	VPS	Case1: High fever, an episode of convulsion, deterioration of neurological status Case1: No symptoms	Case1,2: Anal	Removal of VPS and then reinserted	Recovered successfully
18	Odebode ^[77] 2007	F	9 months	Hydrocephalus	6 months	No	VPS	Skin necrosis along its path behind the ear	Oral with jejunal perforation	Shunt was removed and replaced	Recovered successfully
19	Kanojia ^[61] 2008	Unknown	1 to 3 months	Spina bifida-associated hydrocephalus	3 to 6 months	No	VPS	One patient with meningitis and severe septicemia, others asymptomatic	The right lumbar region in 1 patient, cervical region in 2, umbilicus in 1 and anus in 1	Removal and replacement of shunts after 3-6 weeks	Recovered successfully
20	Borkar ^[25] 2008	F	132 months	Obstructive hydrocephalus	34 months	No	NR	Fever, headache	Anterior chest wall	extruded distal part removed	Recovered successfully
21	Kella ^[63] 2008	F	25 days	Congenital hydrocephalus	1.5 years	No	VPS	Purulent discharge from the umbilicus	Umbilicus	Shunt was removed and replaced	Recovered successfully
22	Berhouma ^[23] 2008	M	9 months	Spina bifida-associated hydrocephalus	15 months	Yes	VPS	Vomiting	Oral	The original ventricular catheter was kept after the peritoneal catheter was withdrawn and the shunt was exteriorized in the neck and attached to a ventricular drain.	Patient died of sepsis within 15 days
23	Mohta ^[78] 2009	F	9 days	Meningitis and hydrocephalus	3 months	No	VPS	Excessive crying and protruding shunt from the anus	Anal	Colon was closed and peritoneal lavage was performed. The shunt revision was done after 3 weeks	Recovered successfully
24	Vuyyuru ^[106] 2009	M	6 years	rontoparietal oligodendroglioma	4 months	No	VPS	Protrusion of the shunt from the anus	Anal	External ventricular drainage followed by removal of shunt	Recovered successfully
25	Low ^[71] 2010	M	18 hours	Congenital hydrocephalus	12 months	Yes	VPS	Vomiting	Oral	VP shunt removal	Recovered successfully
26	Silva Neto ^[94] 2011	M	At birth	Congenital hydrocephalus with agenesis of corpus collosum	5 years	No	VPS	Lumbar pain with swelling and hyperemia	Retropertoneal and sacral bone	External ventricular drainage followed by VPS	Recovered successfully
27	Chiang ^[28] 2010	F	7months	Congenital hydrocephalus	3 years & 5months	Yes	VPS	signs of peritonitis	Anal	Distal catheter was removed	Recovered successfully
28	Kumar ^[67] 2010	2F 1M	Pt 1: 15 days Pt 2: At birth Pt 3: One moth	Congenital hydrocephalus	Pt 1: 1.5 months Pt 2: 2 months Pt 3: 35 months	No	VPS	Protrusion of the shunt	Pt 1: Vagina Pt2: Umbilicus Pt3: Rectum	Shunt was removed and replaced on the contralateral side	Recovered successfully
29	Dua ^[35] 2011	M	20 days	Congenital hydrocephalus	8 months	No	VPS	Vomiting	Oral	proximal end of the shunt externalized, and the distal end detached at the level of the neck.	Recovered successfully
30	de Aguiar ^[51] 2011	F	144 months	Congenital Hydrocephalus	3 months	No	VPS	Altered consciousness	Urethra	Removal of VPS	Patient died
31	Agrawal ^[1] 2011	M	1 year	Congenital hydrocephalus	6 months	No	VPS	Vomiting	Oral	Removal of VPS and then reinserted	Recovered successfully
32	Pohlman ^[86] 2011	M	14 years	Spastic quadriparetic cerebral palsy and severe hydrocephalous	11 years	No	VPS	No symptoms	Urethral	Removal of VPS	Recovered successfully
33	Panigrahi ^[82] 2012	1F, 1M	case 1: 4 months case2: 6 years	Posttraumatic hydrocephalus Pilocytic astrocytoma associated hydrocephalus	3 months 96 months	No	VPS	Case 1: Painless, small blister, and erythema on the upper abdominal wall Case2: No symptoms	Abdominal wall	Peritoneal end removed through abdomen, shunt revision	Recovered successfully
34	Teegala ^[98] 2012	2F, 1M	Case 1: 1.5 year F Case 2: 5 years, M Case 3: 4 months F	Case 1: tubercular meningitis associated hydrocephalus Case 2: middle cranial fossa tumor associated hydrocephalus Case 3: Dandy Walkers malformation associated hydrocephalus	Case 1: 18 months Case 2: 12 months Case 3: 2 months	No	VPS	Case 1: Fever neck rigidity, refusal to food Case 2 : No symptoms Case 3: High-grade fever, neck rigidity, food refusal	Case 1: Anal Case 2 : Anal Case 3: Vaginal	Removal of VP shunt following ETV	Recovered successfully
35	Altas ^[11] 2012	F	At the time of birth	Congenital Hydrocephalus	14 months	No	VPS	No symptoms	Vaginal	Removal of VP shunt and external ventricular drainage and reinsertion of shunt	no follow-up abnormalities
36	Gupta ^[48] 2012	Unknown	6 months	Congenital hydrocephalus	3.5 years	No	VPS	One episode of vomiting followed by fracture of distal end of VPS	Oral	The shunt was removed	Recovered successfully after 12 months follow-up
37	Kataria ^[62] 2013	M	1 month	Congenital hydrocephalus	17 months	Yes	VPS	No symptoms	Penis	The whole shunt was removed through division	Recovered successfully
38	Sharifian ^[93] 2013	Unknown	6 months	Hydrocephalus, cerebral palsy	2 years	No	VPS	Protrusion of the mass from anus, restless, mild abdominal distension	Anal	Extrusion of the catheter from the anus	Patient lost to follow up
39	Gupta ^[49] 2014	M	1 year	Congenital hydrocephalus	11 years	No	VPS	Vomiting, headache, fever	Oral	Removal of VPS No need for shunt more	Recovered successfully
40	Voronovich ^[105] 2014	M	1 year	Post-infectious hydrocephalus, malnutrition, anemia	10 months	No	VPS	Protrusion through the skin of the right frontal region with enterocutaneous fistula above the right clavicle with bilious drainage. Frequent vomiting	Skin	Shunt was removed	Recovered successfully after 9 months of follow-up
41	Mandhan ^[72] 2015	F	NR	Spina bifida-associated hydrocephalus	NR	No	VPS	No symptoms	Oral	VPSC Removal through division	No follow-up abnormalities
42	Oktay ^[79] 2015	M	1months	Spina bifida-associated hydrocephalus	2 years	No	VPS	No symptoms	Right lumbar region	Removal of VPS and then reinserted	Recovered successfully
43	Bansal ^[20] 2015	F	12 months	Congenital hydrocephalus	6 months	No	NR	CSF discharge, normal neurological and physical exam	Anal	Removal of VPS and then reinserted	Recovered successfully
44	Thiong'o ^[99] 2015	4M, 2F	3months to 8 years	Congenital hydrocephalus	Case 1: NR Case 2: 15 months 18 Case 3: 3 months Case 4: 12 months Case 5: NR Case 6: NR	No	VPS	Case1 and 2: No symptoms Case 3: Irritability, retching, and vomiting In case 4,5,6 No symptoms	Case1 and 2: Anal Case 3: Oral Case 4: Midsternal region Case5:Subclavicular region. Case 6: Exposed on the scalp	Case 1 and 2: after shunt division, the distal end was removed through the rectum Case 3: whole shunt was removed through a scalp incision Case 4,5,6 shunt removed on its entirety	Recovered successfully

(Contd...)

Reference Nr.	Investigator	Sex	Age at VPS Surgery (years)	Primary diagnosis	Interval to VPS Extrusion	(Preoperative) Meningitis Peritonitis	Previous One year Treatment	Presentation of Patient	Origin of extrusion	Treatment	Follow-up and outcome
45	Mutlu ^[75] 2015	F	6 day	Arnold Chiari II Malformation associated hydrocephalus	Unknown	No	VPS	No symptoms	Urethra	Removal of VPS and then reinserted	Recovered successfully
46	Lee ^[68] 2015	M	9 months	Traumatic subarachnoid haemorrhage associated hydrocephalus	3 years	No	VPS	Abdominal pain	Anal	Removal of VPS and then reinserted	Recovered successfully
47	Moghul ^[74] 2016	F	48 months	Congenital Hydrocephalus	12 months	Yes	VPS	Excessive irritability, crying, and anorexia for a month	Anal	both proximal and distal part removed	Recovered successfully
48	Sarkari ^[91] 2016	Unknown	14 months	Chiari II Malformation associated Hydrocephalus	Unknown	No	VPS	No symptoms	Anal	Entire removal of VPS No more VPS	Recovered successfully
49	Bodeliwala ^[24] 2016	M	6 months	Congenital Hydrocephalus	4 months	No	VPS	No symptoms	Anal	Removal of VPS and then reinserted	Recovered successfully
50	Gatta LA ^[43] 2016	M	Age of 1 month	Congenital hydrocephalus	7 months	No	VPS	No symptoms	Anal	Removal of VPS and then reinserted	Recovered successfully
51	Lotfinia ^[70] 2017	F	3months	Congenital hydrocephalus	21 months	No	VPS	Vomiting confusion, malfunction of VPS	Vaginal	The peritoneal part was removed from the vagina and reinserted	2 years of follow-up, full recovery
52	Al Fauzi ^[51] 2017	M	2 months	Congenital hydrocephalus	48 months	No	VPS	Abdominal discomfort, severe headache,	Anal	VPSC Removal through division	No follow-up abnormalities
53	Vankipuram ^[104] 2017	M	6 months	Tuberculous meningitis and hydrocephalus	4.5 years	No	VPS	Watery discharge from the umbilical region for 15 days	Umbilical	The shunt was removed	Recovered successfully after 3 months of follow-up
54	Gan ^[60] 2017	M	Unknown	Congenital hydrocephalus	8months	No	VPS	No symptoms	Urethra	VP shunt removal	Discharge on the 6th postoperative day
55	Indra ^[47] 2017	F	6 months	Congenital hydrocephalus	5 months	No	VPS	Protrusion through the anus	Anal	The shunt was removed	Patient lost to follow-up
56	Badri ^[16] 2018	M	48 months	Obstructive hydrocephalus	1 month	No	NR	Vomiting, mild headache, altered consciousness, abdominal pain	Oral	Peritoneal catheter removed and new connection inserted	Recovered successfully
57	Kouitcheu ^[89] 2018	M	2 years	Tetraventricular hydrocephalus	10 months	Yes	No	Vomiting, fever, convulsion	Oral	Removal of VPS and then reinserted	Recovered successfully
58	Chugh ^[90] 2018	5M, 3F	6 months to 7 years	Communicating and non-communicating hydrocephalus	Average, 4.25 months	Yes	3 VP Shunts, 2 ETV	No symptoms	3 abdominal, 3 anal, 2 vaginal	Patients were reinserted with VPS after the removal of extruded ones	One of them expired Others improved
59	Hasan ^[53] 2018	2M	Case 1: 18 months Case 2: 2.5 Years	Case 1: tubercular meningitis associated hydrocephalus Case 2: congenital hydrocephalus	Case 1: 6 months Case 2: 6 months	Yes	VPS	Case 1,2: No symptoms	Case 1: Anal Case 2: Anal	Case 1: removal of shunt Case2: reinsertion after removal of shunt	no follow-up abnormalities
60	Guthe ^[51] 2018	M	48 months	Congenital hydrocephalus	24 months	No	VPS	No symptoms	Anal	Removal of VPS	Recovered successfully
61	Ibebuike ^[56] 2018	M	8 days	Sacral spinal bifida	4 months	No	VPS	Painful Scrotal swelling	Inguinal	a connector was used to reconnect the extruded distal shunt tube to the proximal portion of the shunt tube outside the chest. VPS was removed	Follow-up was lost after 7 weeks
62	Marino ^[73] 2019	F	2 Months	Congenital hydrocephalus	Unknown	Yes	VPS	No symptoms	Anal	Removal of VPS	Recovered successfully
63	Turkis ^[101] 2019	F	3 days	Congenial hydrocephalus	10 months	No	VPS	Drowsy and hypoactive	anal	Removed and reinserted	No complications after 2 years of follow-up
64	Ghritlaharey ^[44] 2019	M	3 months	Malresorptive hydrocephalus	84 months	No	VPS	Multiple infected ulcers around the VPS catheter	Anterior chest wall	Entire removal of VPS	Patient lost to follow up
65	Korulmaz ^[65] 2019	F	At birth	Congenital Hydrocephalus	10 months	No	VPS, colostomy	Vomiting	Vaginal	Removal of VP shunt and external ventricular drainage was performed	Recovered successfully
66	Xia ^[107] 2020	M	90 months	Communicating hydrocephalus	3 months	No	VPS, Hemispherect-omy for intractable epilepsy	Fever, abdominal pain, muscular cramps	Umbilical, 25cm	Extruded part removed intra-abdominal part was repositioned on the diaphragm-facing surface.	No follow-up abnormalities
67	Bakshi ^[18] 2020	M	8 months	Congenital Hydrocephalus	3 months	No	VPS	No symptoms	Anal	extruded part removed VPS reinserted on the other side	Patient lost to follow up
68	Cardinale ^[27] 2020	F	3 week	Intraventricular hemorrhage associated hydrocephalus	8 weeks	No	VPS	No symptoms	Anal	Removal of VPS and then reinserted	Recovered successfully
69	Alhassan ^[8] 2020	M	5 years	Congenital hydrocephalus	1 year	No	VPS	No symptoms	Anal	Removal of VPS No need for shunt more	Recovered successfully
70	Tamlikha ^[97] 2020	M	7 months	Congenital hydrocephalus	2 years	No	VPS	No symptoms	Oral	Removal of VPS and then reinserted	Recovered successfully
71	Alolyani ^[10] 2020	M	At birth	congenital hydrocephalous, cerebral palsy, and seizure	11 years	No	VPS	right abdominal wall patchy redness with localized swelling and drops of fluid coming out of it	Right side of the abdominal wall	Abdominal externalization followed by reinsertion	No complications after 4 years of follow-up
72	Pant ^[83] 2021	11M 6F	6 weeks	lumbosacral or lumbar meningomyelocele	11 pts: before 7 months 3 pts: 12-48 months 3 pts: 60-86 months	2 pts had meningitis	VPS	4 cases presented with fever, 2 of them had seizure 2: intestinal obstruction 1: gastric perforation and per-oral presentation of the shunt 2:rectal perforation with per-anum extrusion of the shunt 1: per-urethral extrusion following vesical perforation 2: umbilical extrusion 5: inguinal hernia	4: skin extrusion 6: hollow viscus perforation (1: gastric/2: rectal/1: urethral) 2: umbilical 5: inguinal	2 skin extrusion: rotational flap (Z-plasty) 2 (long segment inflammation): shunt was removed and contralateral shunt is placed 4: ESD 2: ESD followed by shunt replacement 5: VP shunt was replaced in the peritoneum 2: shunt was removed	Functioning shunt in all patients. 1 case presented with CSF leak in the peritoneum after one month. The shunt
73	Bal'afit ^[19] 2021	F	1 month	Congenital hydrocephalus	12 months	Yes	VPS	No symptoms	Anal	Removal of VPS	Recovered successfully
74	Bosy ^[26] 2021	M	1month	Congenital hydrocephalus	180 months	Yes	NR	Headache, intermittent vomiting	Anal	extruded part removed	Recovered successfully
75	Hidayat ^[54] 2021	F	36 months	Malresorptive hydrocephalus	4 months	No	NR	White watery discharge, intermittent pain in supra pubic area	Vaginal	Distal catheter revision	Recovered successfully
76	Arnaout ^[14] 2021	F	1month	Obstructive hydrocephalus	6months	No	VPS	No symptoms	Urethra	both proximal and distal parts removed	Recovered successfully
77	Guimarães ^[46] 2022	M	48 months	Congenital hydrocephalus	2 months	No	VPS	Long-term dysuria, intermittent incontinence, and chronic abdominal pain	Urethra	The distal portion of VPS removed	Recovered successfully
78	Alalula ^[6] 2022	F	Right at birth, left at 2 months of age	Obstructive hydrocephalus	5 months	No	VPS	No symptoms	Urethra	Removal of the extruded catheter and insertion of new VP SHUNT	Recovered successfully
79	Şahin ^[90] 2023	M	At the age 2 day	Spina bifida-associated hydrocephalus	30 months	No	VPS	Vomiting, fever	Anal	Removal of VPS and then reinserted	Recovered successfully
80	Nazwar ^[7] 2023	M	Unknown	Congenital hydrocephalus	2 years	No	VPS	No symptoms	Anal	Removal of VPS	Recovered successfully

CSF: Cerebrospinal fluid, ETV: Endoscopic third ventriculostomy, EVD: External ventricular drainage, VPSs: Ventriculoperitoneal shunts, VA: Ventriculoatrial

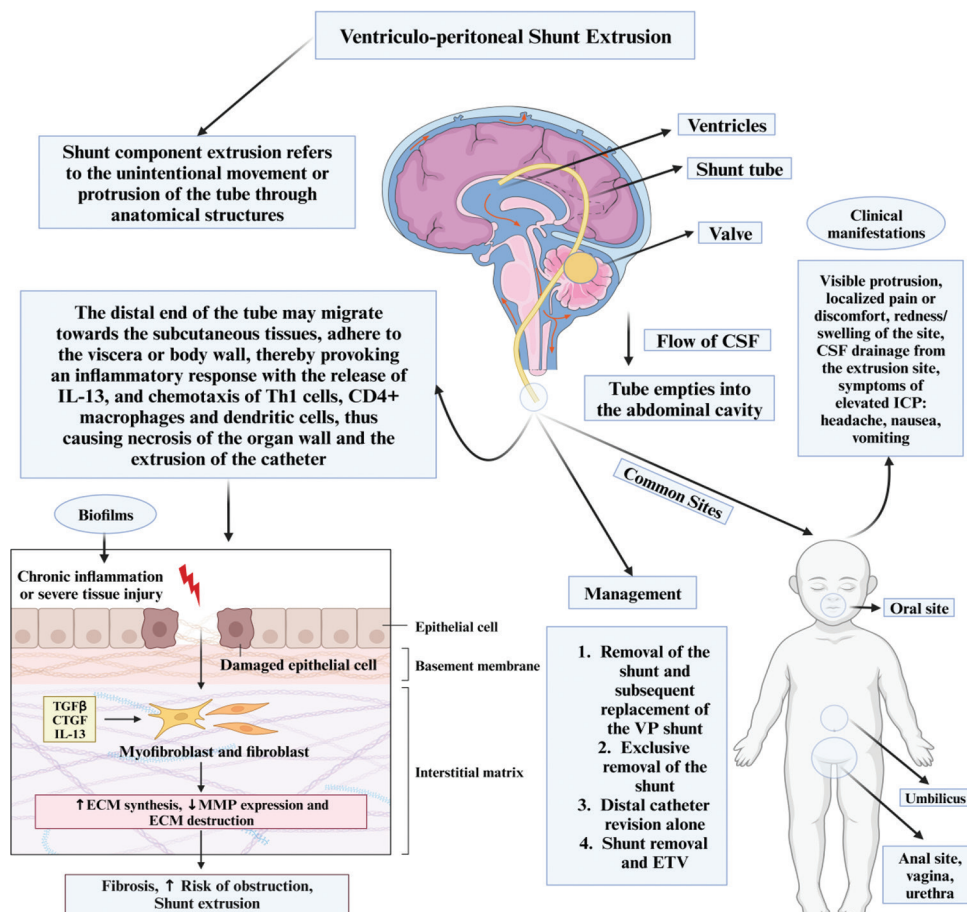


Figure 5: Overview of the ventriculoperitoneal shunt extrusion. IL-13: Interleukin 13, TGF: Transforming Growth Factor, CTGF: Connective Tissue Growth Factor, ECM: Extracellular Matrix, MMP: Matrix Metalloproteinase, ICP: Intracranial Pressure, ETV: External Third Ventriculostomy, VP Shunt: Ventriculoperitoneal shunt

This journey through uncharted territories triggers intricate foreign body tissue responses – inflammatory cells, collagen deposition, and fibrous tissue encapsulation – that are intricately linked to the risk of catheter extrusion, ultimately culminating in one of the most prevalent complications – obstruction, a frequent impetus for revision surgeries.^[3,60] The formation of granulation tissue and fibrotic encapsulation catalyzes the migration and then extrusion of the peritoneal terminus of the VPS, contributing substantively to the constellation of shunt-related complications and constituting approximately 10.81% of the total shunt complications encountered in clinical practice.^[81] We posit that this process of distal catheter migration represents the initial phase in the intricate narrative of catheter extrusion. Importantly, this pathophysiological sequence aligns harmoniously with extant findings in the scientific literature, some of which have been substantiated through investigative studies employing canine models.^[21]

When delving into the realm of other potential predisposing factors, infection emerges as a prominent contender when localized around the shunt components. This infectious

component holds the potential to exert a considerable influence on the likelihood of shunt extrusion. This revelation harmonizes seamlessly with the prevailing body of neurosurgical literature, which underscores the pivotal role of infection in the landscape of shunt-related complications.^[34,66,95] Shunt infection increases the risk of extrusion due to the formation of biofilms on catheter surfaces.^[22] Biofilms can serve as a nidus for chronic inflammation and tissue reactions, ultimately causing more weakness in tissue integrity and increasing the likelihood of catheter migration.^[22]

The clinical presentation of distal catheter extrusion in a VPS can vary depending on the extent of catheter displacement and associated complications. Common clinical signs and symptoms associated with distal catheter extrusion may include visible protrusion of the catheter from the abdomen, usually at the site where it was initially placed;^[5,39] patients may experience localized pain or discomfort at the extrusion site or redness and swelling in the area around the extruded catheter.^[9] There may be drainage of CSF or clear fluid from the extrusion site, which can indicate shunt malfunction.^[39]

The extrusion site can become a potential entry point for infection, leading to symptoms such as fever, increased pain, or redness.^[39,103] Depending on the extent of CSF diversion disruption, patients may experience symptoms related to increased ICP or shunt malfunction, such as headaches, nausea, vomiting, or changes in mental status.^[38,39]

When it comes to the management of extrusion of the distal catheter from a VPS through the abdominal wall, the prompt administration of prophylactic antibiotics is essential, followed by complete replacement of the entire shunt system.^[25,32] Shunt revision in the first 6 months after implantation constitutes around 24% of the total shunt complications.^[81] Pulling the extruded distal end distally avoids the potential spread of infection; cases of in-existent peritonitis, laparotomy, and peritoneal lavage are unneeded.^[64] In the event of peritoneal inflammation or catheter attachment to peritoneal contents, the necessity arises for a laparotomy. Once the existing shunt system has been completely removed, reinserting a fresh shunt system is dependent on the CSF analysis results. When they are negative, it may be contemplated within the same surgical session; however, in scenarios where an infection is suspected, the standard procedure entails installing an EVD system accompanied by initiating intravenous antibiotic treatment.^[82,94] Following this, sequential CSF sampling is carried out until two consecutive negative culture results are obtained. At this juncture, the option to reintroduce a new shunt system becomes a feasible consideration.

Some studies investigated the outcomes of pediatric patients who underwent VPS revision for various complications, including extrusion, and found that early intervention and meticulous surgical techniques were associated with improved outcomes and reduced recurrence rates.^[1,81]

In recurring infections and complications associated with VPSs, a potential solution may involve transitioning to a VA shunt. This approach, as reported in some studies, has been linked to enhanced clinical outcomes and a reduction in ventricular size,^[108] which was done in three cases in our review with two recoveries and one death.^[45] ETV – with postoperative EVD as a detector for ETV failure – is an alternative intervention for patients experiencing shunt malfunction, revealing that 68.4% of cases achieved shunt independence,^[69] which is done in four cases in our review with a 100% recovery rate. The statistically quantifiable success rate supports VPS removal and replacement efficacy with a new VPS. Alternative drainage replacement approaches, such as ETV and VA shunt, should be reserved.

CONCLUSION

Distal catheter extrusion in VPSs involves the unintended protrusion of the catheter from its intended exit site, causing

clinical symptoms such as visible protrusion, localized pain, redness, swelling, CSF drainage, and signs of infection. Prompt management is crucial, typically involving prophylactic antibiotics and complete shunt system replacement, with the timing of reinsertion determined by CSF analysis results, indicating feasibility for immediate reinsertion.

Extrusion sites vary, including the anal or rectal area, abdominal wall, urethra, vagina, umbilicus, and inguinal region, each presenting unique management challenges. Distal catheter extrusion is a medical emergency, necessitating immediate medical attention, surgical intervention, and shunt revision to prevent complications such as infection or intracranial hypertension. Alternative drainage options, such as VASs and ETV with postoperative EVD, may be considered based on the clinical scenario and patient characteristics. Managing distal catheter extrusion has diverse outcomes, with early intervention, meticulous surgical techniques, and appropriate strategies associated with improved outcomes and reduced recurrence rates. Continued research is essential to refine treatment approaches for the varied distribution of extrusion sites in VPSs.

Ethical approval

The Institutional Review Board approval is not required.

Declaration of patient consent

Patient's consent was not required as there are no patients in this study.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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