# **Systematic Review**

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# Extrusion of the distal ventriculoperitoneal shunt catheter through the umbilicus: a systematic literature review from 1973 to 2021

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# **ABSTRACT**

Trans-umbilical extrusion of the distal ventriculoperitoneal shunt catheter is a rare complication of the VPS insertion. The objectives of this review were to analyze various variables like the age, sex, indication for VPS insertion, clinical presentation, the operative therapy offered for the management, and outcome of the cases published/managed for the trans-umbilical extrusion of the distal VPS shunt catheter. In 1973, the maiden case of trans-umbilical extrusion of the distal VPS catheter was published. Literature/case reports were retrieved from 1973 to October 31, 2021. A total of n=24 (12 males, 12 females) cases were recruited for this systematic review. All of them were children below the age of 13-years. In four-fifths (n=19) of children, the initial VPS catheter was inserted during the first 6-months of life. The median age of children at the time of diagnosis of the above-described VPS complication was 7.5 (42.21 SD) months, and it ranged from 2 months to 13 years of age. The median interval from the initial VPS insertion/last VPS revision to the diagnosis of complication was 4 (24.77 SD) months, and it ranged from 3 weeks to 8 years. The main complaint was the extrusion of the distal VPS catheter through the umbilicus. Surgical procedures performed for the management were (a) removal of the entire VPS catheter n=15, (b) removal of distal/part of distal VPS catheter n=5, and (C) others n=2. For VPS catheter revision; delayed re-VPS insertion was performed in n=10, immediate revision of the distal VPS catheter in n=3, and others. Three (12.5%) children died during the postoperative and follow-up period. The distal VPS catheter extrusion through the umbilicus is a rare complication of VPS insertion, and it occurred exclusively in children. In 70% (n=17) of children, it was documented ≤6 months afterward of the VPS insertion. Delayed re-VPS catheter insertion was preferred for 40% of the children for VPS revision procedures.

**Keywords:** Children, Complication, Extrusion, Hydrocephalus, Infants, Umbilicus, Ventriculoperitoneal shunt, Ventriculoperitoneal shunt revision

# INTRODUCTION

Diversion of the cerebrospinal fluid (CSF) from the ventricular system to the peritoneal cavity through the VPS catheter is one of the most frequently performed surgical procedures for treatment of hydrocephalus.<sup>1-3</sup> VPS insertion is associated with a wide variety of complications. The extrusion of the distal VPS catheter through the natural orifices is more common.<sup>4-7</sup> Extrusion

of the distal VPS catheter through the body parts, other than natural orifices, has also been reported but less frequent. Bisla VPS catheter extrusion through the umbilicus has also been reported but less frequent, and only n=27 cases have been reported to date. The present manuscript is a systematic review of the literature of n=24 cases of trans-umbilical extrusion of the distal VPS catheter to highlight the demographics, clinical characteristics, surgical procedures offered for the management, and the outcome.

#### **METHODS**

PubMed, PubMed Central, Google Scholar, Google Images, ResearchGate, and a few other websites were searched for the retrieval of the relevant literature/case reports. The maiden case of trans-umbilical extrusion of the distal VPS catheter was published in 1973. Relevant literature and case reports were retrieved from 1973 to October 31, 2021. The variables included for the review were age, sex, indication for VPS insertion, interval, and operative therapy offered for management, and outcome. Cases with incomplete desired details were excluded from the review. Case reports presented with CSF umbilical fistula but without trans-umbilical extrusion of the VPS catheter were excluded from the review. For the present review, Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines are followed. Various keywords were employed during an electronic search for the literature. The keywords utilized for the online search were, "umbilical extrusion of peritoneal shunt catheter", "trans-umbilical extrusion of ventriculoperitoneal shunt catheter", "trans-umbilical extrusion of peritoneal shunt catheter", "extrusion of CSF shunt catheter through umbilicus", "extrusion of distal ventriculoperitoneal shunt catheter", and "rare/unusual complication of ventriculoperitoneal shunt catheter". The full text of the manuscripts was assessed for the final selection of the manuscripts. It is a single-author manuscript; search for the literature, screening and final selection of the articles, extraction of the desired variable from the manuscripts, manuscript writing, and all others were done by the author alone. This manuscript is a

systematic review of the already published literature/manuscripts, therefore approval from "the institutional ethics committee" was not required.

#### **RESULTS**

The literature search was performed for the selection of the manuscripts for the systematic review of the management of trans-umbilical extrusion of the distal VPS catheter, and is presented in the PRISMA flow chart in Figure 1. A total of n=27 cases related to the treatment of trans-umbilical extrusion of distal VPS were retrieved. 11-33 This review consisted of a total of n=24 cases (12 males, 12 females) of trans-umbilical extrusion of the distal VPS catheter, and all were children below the age of 13-years. 11-30 Nineteen manuscripts were published and available in the English language. 11,13-30 One of the manuscripts was published and available in a language other than English.<sup>12</sup> Fourteen cases of extrusion of the distal VPS catheter through the umbilicus were published as case reports. Three of them were parts of the two original articles, and the remaining six of the cases were parts of the four cases series. Three of the cases were part of the other article, and the desired details were not available and excluded from this review.<sup>31-33</sup> The demographics, clinical and operative details of n=24 children included in the present review of the management of extrusion of the distal VPS catheter through the umbilicus are provided in Table 1.11-30 Results of the present review on the management of trans-umbilical extrusion of the distal VPS catheter are summarized in Table 2.

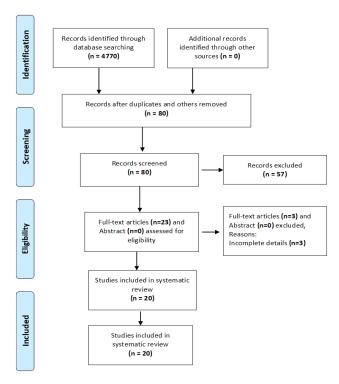


Figure 1: Preferred Reporting Items for Systematic Reviews and Meta-Analyses flow diagram for manuscript selection for the systematic review of the extrusion of the distal ventriculoperitoneal shunt catheter through the umbilicus from 1973 to 2021.

Table 1: Demographics, clinical and operative details of the cases published on the management of extrusion of the distal ventriculoperitoneal shunt catheter through the umbilicus from 1973 to 2021 (n=24).

Case No.	Author(s) Pub. Year Ref. Number	Indication for VPS insertion	Sex	Age VPS insertion (months)	Age umbilical extrusion (months)	Interval (months)	VPS (R)	Meningitis	Operative procedures performed	Remark
1.	Adeloye 1973 <sup>11</sup>	Cong hydroc	Female	9	12	3	No	No	(i) Removal of distal VPS catheter, (ii) Immediate revision of distal VPS catheter	Well
2.	Takahashi et al, 1988 <sup>12</sup>	Cong hydroc	Male	3	16	9	Yes	No	<ul><li>(i) Removal of part of distal VPS catheter,</li><li>(ii) Immediate revision of distal VPS catheter</li></ul>	Well
3.	Das et al, 1992 <sup>13</sup>	Cong hydroc	Male	5	7	3 weeks	Yes	No	Surgical procedure not done (consent not given by parents)	-
4.	Boroujeni 2002 <sup>14</sup>	Cong hydroc with NTD	Female	Neonate	4	4	No	No	<ul><li>(i) Removal of entire VPS catheter,</li><li>(ii) Ventriculo-atrial shunt insertion</li></ul>	Well
5.	Wani et al, 2002 <sup>15</sup>	Cong hydroc	Female	12	18	6	No	Yes	<ul><li>(i) Removal of entire VPS catheter,</li><li>(ii) Delayed re-VPS catheter insertion</li></ul>	Well
6.	Silav et al, 2002 <sup>16</sup>	Hydroc with NTD	Male	1	2	1	No	Yes?	(i) Removal of entire VPS catheter, (ii) EVD, (iii) Delayed re-VPS catheter insertion	Death
7.	de Aquino et al, 2006 <sup>17</sup>	Cong hydroc	Female	1 day	36	36	No	No	(i) Removal of entire VPS catheter?? (Operative details not available)	Well
8.	de Aquino, et al. 2006 <sup>17</sup>	Cong hydroc	Female	6	31	25	No	No	(i) Removal of entire VPS catheter?? (Operative details not available)	Well
9.	de Aquino et al, 2006 <sup>17</sup>	Hydroc Post infective	Female	1	19	NA	Yes	No	(i) Removal of entire VPS catheter?? (Operative details not available)	Death
10.	Eser et al, 2006 <sup>18</sup>	Cong hydroc with NTD	Male	1 day	3	3	No	Yes?	<ul><li>(i) Removal of entire VPS catheter,</li><li>(ii) Delayed re-VPS catheter insertion</li></ul>	Well
11.	Kanojia et al, 2008 <sup>19</sup>	Cong hydroc with NTD	Male	3	6	3	No	No	(i) Removal of entire VPS catheter, (ii) Delayed re-VPS catheter insertion	Well
12.	Kella et al, 2008 <sup>20</sup>	Cong hydroc	Female	1	18	17	No	No	<ul><li>(i) Removal of entire VPS catheter,</li><li>(ii) Delayed re-VPS catheter insertion</li></ul>	Well
13.	Sarkar et al, 2010 <sup>21</sup>	Cong hydroc	Male	5	9	4	No	No	Surgical procedure not done (consent not given by parents)	-

Continued.

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Case No.	Author(s) Pub. Year Ref. Number	Indication for VPS insertion	Sex	Age VPS insertion (months)	Age umbilical extrusion (months)	Interval (months)	VPS (R)	Meningitis	Operative procedures performed	Remark
14.	Kumar et al, 2010 <sup>22</sup>	Cong hydroc	Male	Neonate	3	3	No	No	<ul><li>(i) Removal of entire VPS catheter,</li><li>(ii) Delayed re-VPS catheter insertion</li></ul>	Well
15.	Ardalan et al, 2011 <sup>23</sup>	Cong hydroc with NTD	Female	Neonate?	4	3	No	No	<ul><li>(i) Removal of entire VPS catheter,</li><li>(ii) Delayed re-VPS catheter insertion</li></ul>	Death
16.	Ghritlaharey et al, 2012 <sup>24</sup>	Cong hydroc	Male	5	8	3	No	No	<ul><li>(i) Removal of distal VPS catheter,</li><li>(ii) Immediate revision of distal VPS catheter</li></ul>	Well
17.	Kundal et al, 2012 <sup>25</sup>	Cong hydroc	Male	1	6	5	No	No	<ul><li>(i) Removal of entire VPS catheter,</li><li>(ii) Delayed re-VPS catheter insertion</li></ul>	Well
18.	Ogungbo et al, 2016 <sup>26</sup>	Cong hydroc	Female	2	5	3	No	No	<ul><li>(i) Removal of entire VPS catheter,</li><li>(ii) Delayed re-VPS catheter insertion</li></ul>	Well
19.	Gadjradj et al, 2020 <sup>27</sup>	Hydroc Arterio- venous fistula	Male	NA	12 years	8 years	Yes	No	(i) Removal of entire VPS catheter (re-VPS catheter insertion not required)	Well
20.	Xia et al, 2020 <sup>28</sup>	Hydroc Postoperative	Male	7.6 years	8 years	6	No	No	<ul><li>(i) Extruded distal VPS catheter as EVD,</li><li>(ii) Delayed removal of extruded distal VPS,</li><li>(iii) Repositioning of remaining distal VPS</li></ul>	Well
21.	Calgaro et al, 2020 <sup>29</sup>	Cong hydroc	Female	Neonate?	5	4?	No	No	(i) Removal of entire VPS catheter, (ii) Immediate re-VPS catheter insertion	Well
22.	Pant et al, 2021 <sup>30</sup>	Cong hydroc with NTD	Female	≤6 weeks	6 (median)	5 (median)	No	No	<ul><li>(i) Removal of part of distal VPS catheter,</li><li>(ii) EVD,</li><li>(ii) Delayed re-VPS catheter insertion</li></ul>	Well
23.	Pant et al, 2021 <sup>30</sup>	Cong hydroc with NTD	Male?	≤6 weeks	6 (median)	5 (median)	No	No	(i) Removal of extruded distal VPS catheter, (ii) Repositioning of remaining distal VPS catheter	Well
24.	Ghritlaharey (Present Case) 2021 <sup>p</sup>	Hydroc Post infective	Female	6 years	13 years	7 years	No	No	(A) Extruded distal VPS catheter as EVD (B) Delayed revision of distal VPS catheter	Well

Cong = Congenital, EVD= External ventricular drainage, Hydroc= Hydrocephalus, NA= Not available, NTD= Neural tube defect, Pub=Publication, Ref=Reference, VPS= Ventriculoperitoneal shunt, R=Revision

## Present case report

A 13-years-old girl presented with dull, periumbilical pain for 2-months. There was a thin purulent discharge through the umbilicus for the past 10-days. Her distal VPS catheter was extruded out through the umbilicus for 5 days.

There were no other symptoms relating to the gastrointestinal tract, central nervous system, or others. A VPS catheter was implanted (not by the author) seven years back for post-infectious hydrocephalus. Clinically, her general and systemic examinations were within

normal. Part of her distal VPS catheter was extruded out through the umbilicus. Her VPS catheter was functional and draining clear CSF from the distal end (Figure 2A). Thin purulent discharge through the umbilicus was present (Figure 2B). Mild, peri-umbilical tenderness was present. An organomegaly, abdominal lump, or ascites were not detected. X-rays (AP and lateral view) of the head were done that showed the ventricular catheter, chamber of the shunt system, and upper part of the distal VPS catheter in continuity (Figure 2C). Her x-ray of the chest and abdomen showed continuity of the distal VPS catheter without any kinking or break (Figure 2D). Ultrasound of the abdomen was also reported as normal.

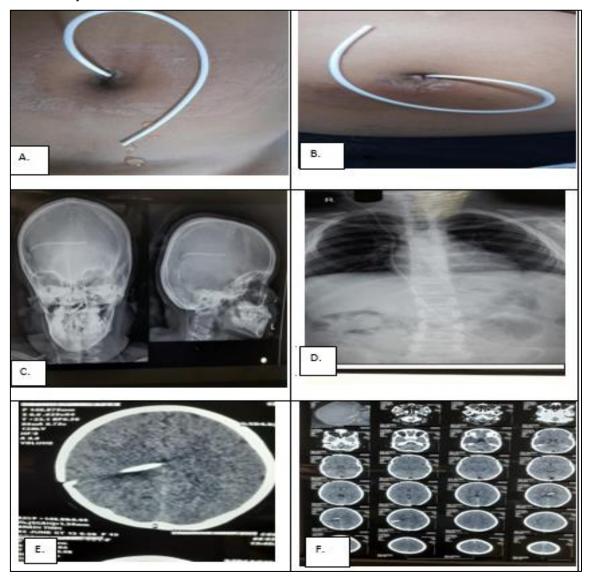


Figure 2 (A) Clinical photograph showing trans-umbilical extruded distal VPS catheter, CSF drops also seen at the tip of the distal VPS catheter. (B) Clinical photograph showing trans-umbilical extruded distal VPS catheter, pus discharge also seen at the umbilicus. (C) X-ray of the head (AP and lateral view) showing ventricular catheter and shunt chamber in place. (D) X-ray of the chest and abdomen showing distal VPS catheter without break/kinking. (E) Cranial computed tomography scan showing ventricular catheter within the brain parenchyma. (F) Cranial computed tomography scans showing ventricular catheter. Ventricular dilatation (hydrocephalus) not seen in any of the scans.

Table 2: Summary of results obtained for the systematic review of the cases of extrusion of the distal ventriculoperitoneal shunt catheter through the umbilicus.

Variables	N				
Total number of cases	n=24 (males=12, females=12)				
Indication for VPS insertion	Cong hydroc=13 (54.16%), Cong hydroc with NTD=7 (29.13%)				
Age: VPS insertion	Median=2 (22.31 SD) months, ranged from 1 day to 7.6 years, Four-fifths (n=19) of children were $\leq$ 6 months of age				
History of VPS revision	Yes, n=4				
Age: Trans-umbilical extrusion	Median=7.5 (42.21 SD) months, ranged from 2 months to 13 years, Four-fifths (n=19) of children were $\leq$ 24 months of age				
Interval	Median =4 (24.77 SD) months, ranged from 3 weeks to 8 years, In seventy percent (n=17) children, the interval was $\leq$ 6 months.				
Chief complaint	Extrusion of the distal VPS catheter through umbilicus=24				
Additional complaints	Discharges through the umbilicus=6, Abdominal pain=7 Symptoms relating to the central nervous system=2				
Meningitis/CSF infection	Clinical meningitis=2, CSF infection=7, peritonitis=nil				
VPS function	Distal end of VPS catheter was draining CSF=17				
Radiological investigations	Plain x-rays=9, Ultrasonography of the abdomen=8, Ultrasonography of the head=2, Computed tomography scan of the head=1, Computed tomography scan of the abdomen=1,				
Operative procedures done	(A) Removal of the entire VPS catheter=15 (62.5%), (B) Removal of distal/part of distal VPS catheter=5 (20.8%), (C) Distal VPS catheter as external ventricular drainage=2 (8.3%), (D) Surgical procedures not done =2 (8.3%)				
VPS revision done	(A) Delayed re-VPS insertion=10 (41.6%), (B) Immediate revision of distal VPS catheter=3 (12.5%), (C) Immediate re-VPS insertion=1 (4.1%), (D) Delayed revision of distal VPS catheter=1 (4.1%), (E) Conversion to ventriculo-atrial shunt=1 (4.1%), (F) Repositioning of the distal VPS catheter=2 (8.3%), (G) Surgical procedures not done=2 (8.3%), (H) re-VPS insertion not required=1 (4.1%), (I) Details not available =3 (12.5%)				
Postoperative mortality	n=3 (12.5%)				

Her cranial CT scan showed a ventricular catheter well in place and ventricular dilatation (hydrocephalus) not seen in any of the films (Figures 2E and 2F). Her previous CT scan of the head was not available for review. CSF was positive for Methicillin-resistant Staphylococcus aureus. Her extruded VPS catheter was connected to the sterile collecting bag. The daily CSF drainage was 400 ml to 600 ml, and the CSF was clear. As there was no hydrocephalus on her cranial CT scan, and therefore her VPS catheter was clamped a few times, but she experienced a headache after that. She was VPS dependent, and after two weeks of injectable antibiotic therapy, her distal VPS catheter was revised through mini-laparotomy. Her postoperative recovery was excellent and, doing well on telephonic follow-up was done three months after the VPS revision. The present case is added as case-24 in table 1 and cited as "p" in the text. Consent from the parent was obtained for the publication/presentation of the clinical/radiological photographs of the girl.

# **DISCUSSION**

Perforation of the hollow viscera, followed by the distal VPS catheter extrusion through the natural orifices, is one of the known complications of VPS insertion.<sup>2,4-7,34-38</sup>

Harischandra et al, in a comprehensive literature review for the VPS migration, and found that approximately 14% of the migration occurred through the abdominal wall.<sup>9</sup> They documented that majority of the migration was through the anterior abdominal wall. They also identified n=10 cases of trans-umbilical migration/ extrusion of the distal VPS catheter, and the least was the migration through the posterior abdominal wall.<sup>9</sup> In 1973, Adeloye first reported the spontaneous trans-umbilical extrusion of the distal VPS catheter in a child.11 The present systematic review of the literature revealed a total of n=27 cases of extrusion of the distal VPS catheter through the umbilicus. 11-33 Due to the incomplete details, three of the cases/children were excluded from the present review.31-33 In 1975, Antunes et al first reported spontaneous CSF umbilical fistula in two cases, detected after the VPS insertion.<sup>39</sup> CSF umbilical fistula is more frequent than that of the trans-umbilical extrusion of the distal VPS catheter.40-43

# Summary of evidence

A review of the reported/published cases of transumbilical extrusion of the distal VPS catheter revealed that it occurred exclusively in children and occurred equally in girls and boys. It was detected earliest at the age of two months and ranged up to 13 years. Four-fifths (n=19) of children were ≤24 months of age at the time of diagnosis of the umbilical extrusion of the distal VPS catheter. The median age of children at the time of diagnosis of the above-described VPS complication was 7.5 (42.21 SD) months. It was less frequent, 36-months after the VPS insertion/revision, and occurred only in n=2 of children.<sup>27</sup>

Initial VPS catheter was implanted for congenital hydrocephalus in (n=13) 54% of children. 11-13,15,17,20,22,24-26,29 In 30% (n=7) of children, the indication for initial VPS catheter insertion was hydrocephalus associated with NTD/developed after repair of NTDs. 14,16,18,19,23,30 Initial VPS catheter was implanted for post-infectious hydrocephalus in n=2 (8.3%). 17 In two (8.3%) of children, the indication for VPS insertion was others. 27,28 The age of children at the time of initial VPS insertion ranged from 1 day to 7.6 years with a median of 2 (22.31 SD) months. In four-fifths (n=19) of children, the initial VPS catheter was inserted during the first 6-months of life.

The median interval from the initial VPS insertion/last VPS revision to the diagnosis of the above-described VPS complication was 4 (24.77 SD) months, and it ranged from 3 weeks to 8 years. In seventy percent (n=17) children, the interval was ≤6 months. This complication was less frequent after 36-months of VPS insertion and n=2 cases were identified. For the cases with a prior history of VPS shunt revisions, the interval was calculated from the last VPS revision to the diagnosis of the extrusion of the distal VPS catheter.

All the children clinically presented with the chief complaint of extrusion of the distal VPS catheter through the umbilicus. Associated additional symptoms were also documented and that were: discharges through the umbilicus (n=7) abdominal pain (n=6) and symptoms related to the central nervous system (n=2). 13,15,16,18,20,23,26-<sup>28</sup> Signs of clinical meningitis were evidenced in n=2 of the children. 15,18 CSF was positive for pathogens in n=7 cases. 12,15,16,18,23,25,27 Peritonitis was not evidenced in any of the cases. In seventy percent (n=17), the VPS system was functional, and it was draining CSF from the distal end of the catheter. 11-13,18-28,30 The clinical diagnosis was possible in all the cases due to the finding of an extruded VPS catheter through the umbilicus. The radiological investigations ordered during the management, and were plain x-rays (n=9), USG of the abdomen (n=8) cranial CT scan (n=1) p and CT scan of the abdomen (n=1). 15,20,23-

Surgical interventions were carried out in children for the treatment of trans-umbilical extrusion of the distal VPS catheter. The surgical therapy offered in the order of frequency were as- removal of the entire VPS catheter n=15.  $^{14\text{-}20,22,23,25\text{-}27,29}$  Removal of the distal/part of distal VPS catheter n=5.  $^{11,12,24,30}$  Distal VPS catheter converted as external ventricular drainage n=2.  $^{28}$  Surgical procedures not done in n=2 of children, as their parents refused surgical therapy.  $^{13,\,21}$ 

VPS revision is also an integral part of the management of cases of extruded distal VPS catheter, whether it was extruded through one of the natural orifices or extruded through the other body parts. Revision of the VPS catheter was also carried out in n=16 children. Delayed re-VPS insertion was the most preferred option and performed in n=10 children. 15,16,18-20,22,23,25,26,30 The immediate revision of the distal/peritoneal part of the VPS catheter was preferred for n=3 children. 11,12,24 Immediate re-VPS insertion was done for one of the children. 29 Conversion to the ventriculoatrial shunt was done for n=1 of children. 4 One of the children did not require re-VPS insertion. 27

This review revealed three (12.5%) deaths observed during postoperative and follow-up periods. <sup>16,17,23</sup> Deaths were not directly related to the therapy offered for transumbilical extrusion of the distal VPS catheter. Deaths were related to the disease itself and the complications related to the VPS insertion. One of the children died due to ventriculitis/meningitis. <sup>17</sup> Another child died 10-months after the VPS revision due to unknown reasons, and another one died 2-months after the VPS revision due to respiratory failure. <sup>16,23</sup>

The exact cause for the trans-umbilical extrusion of the distal VPS catheter is not known. In the anterior abdominal wall, umbilicus is a relatively weak point. 44,45 Probably various factors are involved in the occurrence of the trans-umbilical extrusion of the distal VPS catheter. They are weakness of the umbilicus, CSF infection/shunt infection, peristaltic movements, omental activity, abdominal adhesion formation, wall contractions, intra abdominal pressure, and tissue reaction for the shunt catheter. Probably, infection is one of the important factors and it was documented in nine of the cases. 12,15,16,18,23,27,28 In two of the umbilical/peri-umbilical abscess was also present. 16,18 Extrusion of the VPS catheter through the persistent of the part of the urachus towards the umbilicus was also documented by one of the authors.<sup>20</sup>

## Limitations

One of the limitations of this systematic review is that the cases on the management of umbilical extrusion of the VPS catheter were limited. Three such cases were also not included in the present manuscript because of the unavailability of the desired details. The standard guideline is not available for the management of extrusion of the distal VPS catheter through the umbilicus, most probably because of the fewer number of reported cases. The authors used various procedures, especially for the VPS revisions.

## **CONCLUSION**

Extrusion of the distal VPS catheter through the umbilicus is a rare complication of VPS insertion. Transumbilical extrusion of the distal VPS catheter occurred exclusively in children. In 70% of children, it was

documented ≤6 months of the VPS insertion. Sixty-two percent of children were treated by removal of the entire VPS catheter. Another one-fifths of children were treated by removal of the distal/part of the distal VPS catheter. Delayed re-VPS catheter insertion was preferred for 40% of the children for VPS revision.

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