

Systematic Review

Management of trans-vaginal extrusion of the distal ventriculoperitoneal shunt catheter: a systematic literature review from 1973 to 2021

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ABSTRACT

Trans-vaginal extrusion of the distal ventriculoperitoneal shunt (VPS) catheter is a rare complication of VPS insertion done for the treatment of hydrocephalus. The primary purpose was to review the demographics. The secondary objectives were to review the clinical presentation, operative intervention executed, and outcome of the cases published on trans-vaginal extrusion of the distal VPS catheter. The literature and case reports were retrieved from 1973 to September 30, 2021. This literature review included (n=19) cases of the above-mentioned VPS complication. The mean age of 15 children at the time of diagnosis of the complication was 18.13 months and ranged from 2 to 72 months. For the entire case, the interval from the initial VPS insertion/VPS revision to the clinical diagnosis of complication ranged from 1 to 36 months, with a mean of 8.16 months. In three-fourth of the cases, it was detected within 6-months of the VPS implantation. Extrusion of the distal VPS catheter through the vagina was the chief complaint. Surgical procedures were performed in the following order of frequency (A) removal of the entire VPS catheter (n=14), and (B) removal of the distal/peritoneal VPS catheter (n=3). During the postoperative period, one of the children died. Extrusion of the distal VPS catheter through the vaginal orifice is a rare complication of VPS insertion. It occurred across all the age groups but was more common during early childhood. Three-fourth of them were treated by removal of the entire VPS catheter. For VPS revision, delayed re-VPS insertion was the preferred option.

Keywords: Children, Complication, Extrusion, Hydrocephalus, Infants, Vagina, VPS, VPS revision

INTRODUCTION

Ventriculoperitoneal shunt (VPS) catheter insertion is one of the most frequently performed neurosurgical procedures, for the treatment of hydrocephalus.¹⁻⁴ A variety of complications are known to occur after the VPS insertion, and many of them require VPS revisions.⁵⁻⁸ Perforation of the abdominal hollow viscera and extrusion of the distal VPS catheter through the natural orifices is also a known complication. Extrusion of the

distal VPS catheter through the anal canal is the commonest, followed by the trans-urethra extrusion, trans-oral extrusion, and trans-vaginal extrusions.⁹⁻¹² Extrusion of the distal VPS catheter through the vaginal orifice has also been reported but less frequent, and twenty such cases have been reported to date.¹³⁻³¹ A systematic literature review was performed for 19 such cases to highlight demographics, clinical characteristics, surgical interventions executed, complications, and the outcome.¹³⁻³⁰

METHODS

Preferred reporting items for systematic reviews and meta-analyses (PRISMA) guidelines are followed for this literature review. An electronic database search was performed for the retrieval of literature, on the management of trans-vaginal extrusion of the distal VPS catheter. Various keywords were employed for the online search and were; “trans-vaginal extrusion of VPS catheter”, trans-vaginal extrusion of peritoneal shunt catheter”, “extrusion of shunt catheter through the vagina”, “extrusion of distal VPS catheter”, and “rare/unusual complication of VPS catheter”. The literature and case reports were- retrieved from 1973 to September 30, 2021.

One such case with incomplete desired details was excluded. The cases published on the perforation of the fallopian tube/uterus by the distal VPS catheter, but without per-vaginal extrusion of the same were excluded. The final selection of manuscripts for this review was done by assessing the full texts. The literature search, manuscripts selection, and retrieval of the desired

variables/information were done by the author alone. Institutional ethical committee approval was not required for this review.

RESULTS

The result of an electronic database search of literature/manuscript for the management of the trans-vaginal extrusion of the distal VPS catheter is provided as PRISMA flow chart in Figure 1. A total 20 cases on the management of the above-mentioned complication were retrieved.¹³⁻³¹ A case was excluded from the review, due to the incomplete desired details.³¹ A total of 19 cases were recruited for the systematic review and retrieved from the 18 manuscripts.¹³⁻³⁰ Fifteen manuscripts are available in the English language.^{13,14,17-21,23-30} Three of the manuscripts are available in a language other than English.^{15,16,22} The demographics and other clinical details of the 19 cases of trans-vaginal extrusion of the distal VPS catheter are provided in Table 1. The summary of the result, obtained from the systematic review of the 19 cases of trans-vaginal extrusion of the distal VPS catheter are provided in Table 2.

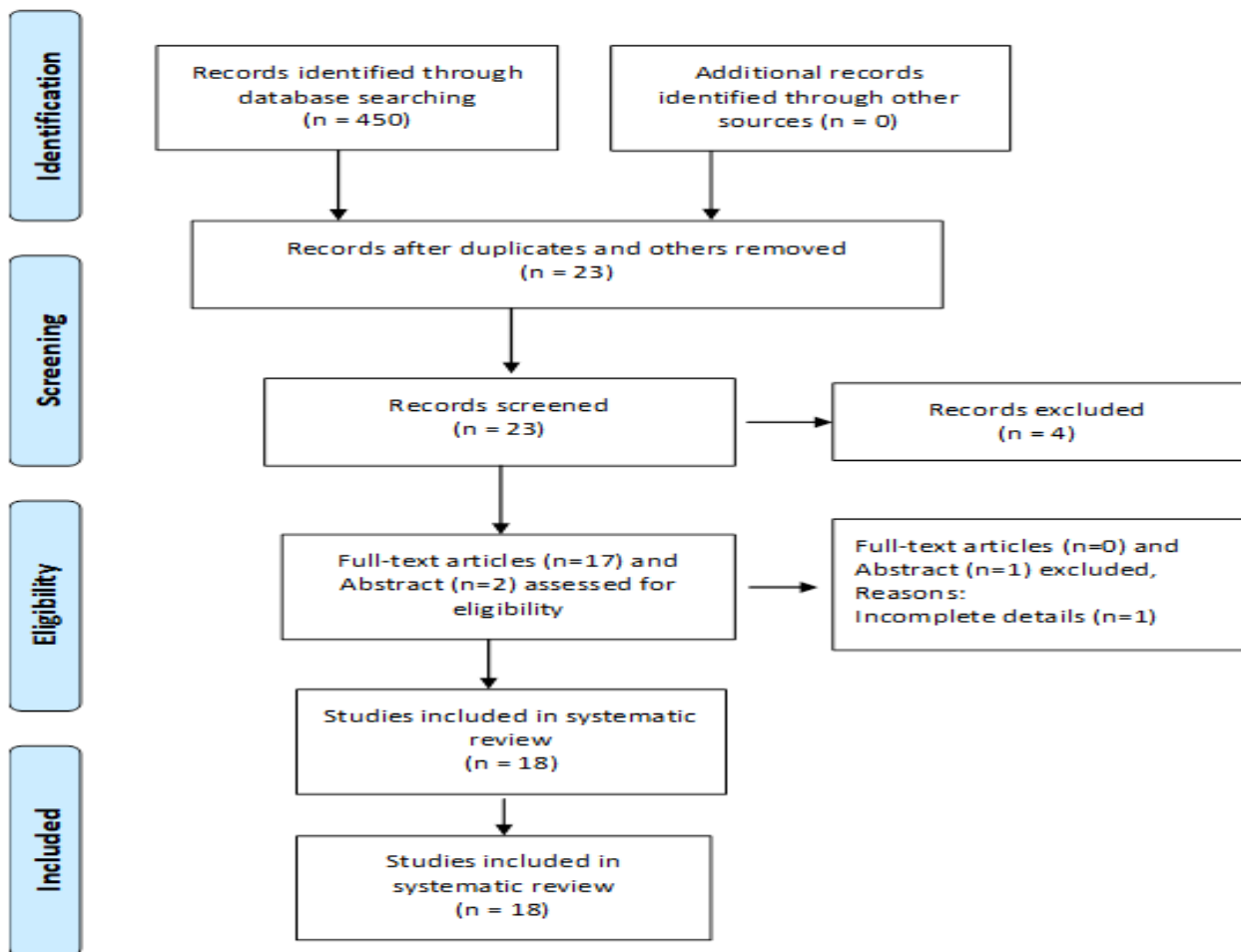


Figure 1: Preferred reporting items for systematic reviews and meta-analyses flow diagram for manuscript selection for systematic literature review of the management of trans-vaginal extrusion of the distal VPS catheter from 1973 to 2021.

Table 1: Demographics, clinical and operative details of cases published on the management of trans-vaginal extrusion of the distal VPS catheter from 1973 to 2021, (n=19).

| Author(s), Publication year and reference numbers | Indication for VPS insertion | Age VPS insertion (months) | Age per-vaginal extrusion (months) | Interval (months) | VPS (R) | Meningitis Peritonitis | Operation procedures executed | Complication | Remark |
|---|-------------------------------|----------------------------|------------------------------------|-------------------|---------|------------------------|---|--------------|--------|
| Patel et al (1973)¹³ | Cong hydroc with NTD | 8 | 11 | 4 | Nil | Meningitis | Removal of entire VPS catheter, Delayed re-VPS catheter insertion | Nil | Well |
| Mozingo et al 1974¹⁴ | Hydroc brain tumor | 49 years | 50 years | 3 | Nil | Nil | Removal of entire VPS catheter, Delayed re-VPS catheter insertion | Nil | Well |
| Pianetti et al 1991¹⁵ | Cong hydroc with NTD | 2 | 13 | 8 | Yes | Meningitis | Removal of entire VPS catheter, External ventricular drainage? | Yes | Death |
| Pereira et al 1999¹⁶ | Cong hydroc | 1 | 6 | 5 | Nil | Nil | Removal of entire VPS catheter, Delayed re-VPS catheter insertion | Nil | Well |
| Farrokhi et al 2007¹⁷ | Cong hydroc with NTD | 4 | 16 | 6 | Yes | Nil | Operative details not provided | NA | NA |
| Kumar et al 2010¹⁸ | Cong hydroc | 1 | 2 | 1 | Nil | Nil | Removal of entire VPS catheter, Delayed re-VPS catheter insertion | Nil | Well |
| Altas et al 2012¹⁹ | Cong hydroc | 3 | 14 | 2 | Yes | Nil | Removal of entire VPS catheter, External ventricular drainage Delayed re-VPS catheter insertion | Nil | Well |
| Teegala et al 2012²⁰ | Cong hydroc | 4 | 6 | 2 | Nil | Meningitis | Removal of entire VPS catheter third ventriculostomy | Nil | Well |
| Bonfield et al 2015²¹ | Hydroc (pseudo-tumor cerebri) | NA/adult | 45 years | 13 | Yes | Nil | Removal of entire VPS catheter, External ventricular drainage, Delayed ventriculo-pleural shunt | Nil | Well |

Continued.

| Author(s), Publication year and reference numbers | Indication for VPS insertion | Age VPS insertion (months) | Age per-vaginal extrusion (months) | Interval (months) | VPS (R) | Meningitis Peritonitis | Operation procedures executed | Complication | Remark |
|---|------------------------------|----------------------------|------------------------------------|-------------------|---------|------------------------|---|--------------|--------|
| Tavallae 2015²² | Cong hydroc | 6 | 36 | 30 | Nil | Nil | Removal of entire VPS catheter, External ventricular drainage Delayed re-VPS catheter insertion | Nil | Well |
| Lotfinia et al 2017²³ | Cong hydroc | 3 | 24 | 6 | Yes | Nil | Removal of distal VPS catheter, Immediate revision of distal VPS | Nil | Well |
| Houten et al 2017²⁴ | Cong hydroc | NA/child | 38 years | NA | NA | Nil | Removal of part of distal VPS catheter Immediate ventriculo-pleural shunt | Nil | Well |
| Chugh et al 2018²⁵ | Cong hydroc | 4 | 6 | 2 | Nil | Nil | Removal of entire VPS catheter, Delayed third ventriculostomy | Nil | Well |
| Chugh et al 2018²⁵ | Cong hydroc | 5 | 8 | 3 | Nil | Nil | Removal of entire VPS catheter, Delayed re-VPS catheter insertion | Nil | Well |
| Soufiany et al 2018²⁶ | Cong hydroc with NTD | 8 | 12 | 4 | Nil | Meningitis | Removal of entire VPS catheter, Delayed re-VPS catheter insertion | Nil | Well |
| Korulmaz et al 2019²⁷ | Cong hydroc | 1 (Neonate) | 10 | 6 | Yes | Nil | Removal of entire VPS catheter, External ventricular drainage Delayed re-VPS catheter insertion | Nil | Well |
| Sahinturk et al 2020²⁸ | Hydroc | 24 years | 34 years | 12 | Yes | Nil | Active surgical therapy not needed (distal VPS catheter already passed) | Nil | Well |
| Lord et al 2020²⁹ | Cong hydroc | 36 | 72 | 36 | Nil | Nil | Removal of entire VPS catheter, External ventricular drainage, Delayed re-VPS catheter insertion | Nil | Well |
| Hidayat et al 2021³⁰ | Hydroc (meningitis) | 32 | 36 | 4 | Nil | Nil | Removal of distal VPS catheter, Immediate revision of distal VPS | Nil | Well |

Cong=Congenital, Hydroc=Hydrocephalus, NA=Not available, NTD=Neural tube defect, VPS=Ventriculoperitoneal shunt, (R)=revision.

Table 2: Summary of the result of the cases of management of trans-vaginal extrusion of the distal VPS catheter, (n=19).

| Variables | N |
|---|---|
| Indication for VPS insertion | Cong hydroc=11 |
| | Hydroc with neural tube defects=4 |
| | Brain tumor=2 |
| | Post-infective hydroc=1 |
| | Others=1 |
| Age: (months), VPS insertion | Mean=7.86 for children, ranged from 1 to 36 |
| | Mean=36.5 years for adults, ranged from 24 to 49 years |
| VPS revision | Yes=7 |
| Age: (months), trans-vaginal extrusion | Mean=18.13 for children, ranged from 2 to 72 |
| | Mean=41.75 years for adults, ranged from 34 to 50 years |
| Interval for entire cases (months) | Mean=8.16, ranged from 1 to 36 |
| Chief complaint | Extrusion of distal VPS catheter through the vagina=19 |
| Additional complaints | Watery discharge through the vagina=10 |
| | Symptoms relating to the central nervous system=5 |
| | Lower abdominal pain/discomfort=2 |
| | |
| Meningitis | Clinical meningitis=4 |
| Peritonitis | Nil |
| Functional status of VPS | CSF draining at the distal end of VPS catheter=13 |
| CSF infection | CSF positive for pathogens on culture test =5 |
| Radiological investigations | Plain x-rays=19 |
| | Ultrasonography of abdomen=2 |
| | Computed tomography scan of head=8, |
| | Computed tomography scan of abdomen=4 |
| | |
| Operative procedures done | (A) Removal of the entire VPS catheter=14 |
| | (B) Removal of the distal VPS catheter=3 |
| | (C) Surgical therapy not needed =1 |
| | (D) Details not available=1 |
| VPS revision | Delayed re-VPS insertion=10 |
| | Immediate revision of distal VPS catheter=2 |
| | Conversion to ventriculo-pleural shunt=2 |
| | Endoscopic third ventriculostomy=2 |
| | Re-VPS insertion not needed=1 |
| | Details not available=1 |
| Complication and mortality | N=1 (5.26%) |

CSF=Cerebrospinal fluid, Cong=Congenital, Hydroc=Hydrocephalus, VPS=Ventriculoperitoneal shunt

DISCUSSION

The extrusion of the distal VPS catheter through the natural orifices is a known complication of the VPS insertion.⁹⁻¹² The most common amongst the above is the trans-anal extrusion of the distal VPS catheter. It was first reported by Sells et al in 1973, and since then, approximately 275 such cases have been reported. Trans-anal extrusion of distal VPS catheter has been reported in 0.2 to 2.5% of the VPS insertion done for the treatment of hydrocephalus.^{5,9,11,32-34} Trans-urethral extrusion of the distal VPS catheter has been reported in approximately 30 cases to date.^{10,11,33-35} Trans-oral extrusion of the distal VPS catheter has also been reported in approximately 30 cases to date.^{11,12,33,36,37} The extrusion of the distal VPS catheter through the vaginal orifice has also been reported but is less frequent than all the above extrusions. In 1973,

Patel et al first-time reported trans-vaginal extrusion of the distal VPS catheter in a child, and since then, only 20 such cases have been reported to date.¹³⁻³¹ An infant with extrusion of the distal VPS catheter in between the labium majus and the labium minus has also been reported and retrieved during the search. The distal VPS catheter was not extruded through the vagina as such, and the same was not included for review.³⁸ During an online literature search, one case of hydrosalpinx, one case of fallopian tube perforation and, one case of uterine perforation by the distal VPS catheter were also retrieved but were without the extrusion through the vagina and were excluded from the review. The above three cases presented with vaginal CSF discharges.³⁹⁻⁴¹

This systematic review revealed that the trans-vaginal extrusion of the distal VPS catheter occurred across all

age groups. The mean age of the 15 children at the time of diagnosis of trans-vaginal extrusion of distal VPS catheter was 18.13 (17.47 SD) months, and ranged from 2 to 72 months. Four-fifth (n=15) of the cases were children below the age of 6 years, and four were adults. In 8 (42.1%) children, the complication was detected within 12 months of age. In 6 (31.57%) children, it was diagnosed during 13 to 36 months of life, and 4 (21.05%) of the cases were adults.

The indications for the initial/primary placement of the VPS catheter in the order of frequency were, (a) Hydrocephalus of congenital origin (congenital hydrocephalus) 11 (57.89%), (b) Hydrocephalus associated with neural tube defect (NTD) or hydrocephalus developed after the repair of the NTD, 4 (21.05%), (c) Hydrocephalus due to brain tumor 2 (10.52%), (d) post-infectious hydrocephalus (n=1), and (e) Other (n=1). In two-third (n=13) children, the primary VPS insertion was done during infancy. It was done during the 25 to 36 months of age in n=2 children, and in 3 of the cases were adults. The mean age at the primary VPS insertion for 15 children was 7.86 (10.49 SD) months, and ranged from 1 to 36 months.

For the entire cases, the interval from the initial VPS insertion/VPS revision to the diagnosis of trans-vaginal extrusion of the distal VPS catheter ranged from 1 to 36 months, with a mean of 8.16 (9.38 SD) months. For the cases with a prior history of VPS revision(s), the interval was calculated from the last VPS revision to the diagnosis of trans-vaginal extrusion of the distal VPS catheter. In three-fourth (n=15) of the cases, said complication was detected within 12 months of the VPS implantation, and two-third (n=13) of them were evidenced during the first 6-months following the VPS insertion.

Extrusion of the distal VPS catheter through the vagina was the chief complaint about the entire case. Additional symptoms were watery vaginal discharge (n=10), symptoms relating to the central nervous system (n=5), and lower abdominal pain/discomfort (n=2). Clinical examination revealed signs of meningitis in four of the cases. None of the cases had the features of peritonitis. In two-third of the case (n=13) VPS catheter was functional, and it was draining CSF. Clinical diagnosis was obvious in all the cases due to the findings of trans-vaginal extruded distal VPS catheter. The radiological investigations were carried out and were plain x-rays (n=19), cranial CT scan (n=8), and CT scan of the abdomen (n=4). CSF obtained during the therapy were positive for pathogens in 5 of the cases.^{13,15,21,26,29}

Surgical procedures offered for the trans-vaginal extrusion of the distal VPS catheter were (A) removal of the entire VPS catheter (n=14), and (B) removal of the distal/peritoneal VPS catheter (n=3). Surgical intervention was not needed/not required for one of the

cases. The management details for one of the cases were not available/not provided by the author.

The site of perforation by the distal VPS catheter was reported as (a) vaginal wall (n=4), (b) vaginal cuff (n=3), (c) fallopian tube (n=2), (d) Cul-de-sac (n=1), and details not available/not mentioned by the authors (n=9). In 16 cases, perforation was not repaired and that healed spontaneously after removal of the migrated VPS catheter.^{13-15,18-20,22-30} The perforation was repaired in two of the cases only.^{16,21} Three-fourth (n=14) of the cases were managed by using percutaneous surgical procedures, for the removal of the extruded VPS catheter.^{13-15,18-27} Two of the authors managed their cases by doing laparotomy.^{16,30} One author managed his case by doing laparoscopic procedures.²⁹ At one occasion, active surgical therapy was not required, as the distal VPS catheter spontaneously passed per-vaginum.²⁸ One of authors did not mention the management part of case.¹⁷

Revision of the VPS catheter was executed in 17 of the cases. Delayed re-VPS insertion was preferred by the authors and was carried out in 10 cases. The immediate revision of the distal/peritoneal part of the VPS catheter was done for 2 cases. Conversion to a ventriculo-pleural shunt was carried out in two of the cases. The third ventriculostomy was carried out in two of the cases. Re-VPS insertion was not required for one of the cases, and details were not available for another case.

The postoperative complication was documented in one case, and the same child died during management. She had clinical meningitis, and her CSF was also positive for bacterial growth. She died of the meningitis; a complication of the VPS insertion done for the treatment of hydrocephalus.¹⁵

The exact cause for the occurrence of the trans-vaginal extrusion of the distal VPS catheter is not known. Amongst 19 cases included for review, 15 were children below the age of 6-years and four were adults. Seven of the cases had a prior history of the VPS revisions.^{15,17,19,21,23,27,28} Three adults had a prior history of hysterectomy, and the site of perforation was the vaginal cuff.^{14,21,24} The extrusion of the distal VPS catheter through the natural orifices, including trans-vaginal extrusion especially, in children, is a cumulative effect of the small peritoneal cavity, insertion of the extended length of the distal VPS catheter, the thinness of the visceral wall, an inflammatory reaction caused by the prior VPS revision/abdominal procedures, inflammatory reaction and mechanical pressure effect generated by the CSF, tissue reaction for the VPS catheter, and intra-abdominal pressure.

Limitations

The major limitation of the present review is that the cases were limited. A case was excluded from the present review due to the unavailability of full-text/desired

information in the manuscript. An infant with extrusion of the distal VPS catheter in between the labium majus and the labium minus was also excluded from review. Three cases were also not included for the review, as all of them presented with vaginal discharges, but without the trans-vaginal extrusion of the distal VPS catheter.

CONCLUSION

Extrusion of the distal VPS catheter through the vaginal orifice is a rare complication of VPS insertion done for the treatment of hydrocephalus. Trans-vaginal extrusion of the distal VPS catheter occurred across all the age groups but was more common during early childhood. It occurred within 6-months of the VPS insertion, in three-fourth of the cases. Three-fourth of the cases were managed by the removal of the entire VPS catheter. For VPS revision, delayed re-VPS insertion was the preferred option. This review finding also suggests that repair of the perforation site is not mandatory, and after removal of the extruded/migrated VPS catheter, it would heal spontaneously. Invasive surgical procedures are also not advisable for the management of such cases.

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