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CASE REPORT

Can we consider ventriculo-gallbladder shunt a first-line treatment in selected patients? Case report of a successful management

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Abstract

Introduction Ventriculo-gallbladder shunt (VGS) has been recognized as a last-resort alternative to treat hydrocephalus when the peritoneum and/or other distal sites can no longer receive shunts. In some specific conditions, it may be conceded as a first-line treatment.

Case presentation We report the case of a 6-month-old girl with progressive post-hemorrhagic hydrocephalus who presented a concomitant chronic abdominal symptom. Specific investigations ruled out acute infection and led to the diagnosis of chronic appendicitis. Both problems were managed in a one-stage salvage procedure consisting of laparotomy sanctioning to treat the abdominal pathology and seize the opportunity to perform a VGS as a first option since the abdomen is prone to ventriculoperitoneal shunt (VPS) failure.

Conclusion Only few cases have reported the use of VGS as the first option to handle uncommon complex cases due to abdominal or cerebrospinal fluid (CSF) conditions. We wish to draw attention to VGS as an effective procedure not only in children with multiple shunt failures but also as first-line management in some selected cases.

Keywords Hydrocephalus · Shunt · Ventriculo-gallbladder · Case report

Introduction

The peritoneum remains the first receptacle choice for CSF diversion. However, in some instances, the VPS is rendered infeasible due to local complications. In these cases, the VGS, which is usually used as a last resort, becomes a viable option. Herein, we performed a VGS, on a 6-month-old girl, for progressive hydrocephalus secondary to neonatal intraventricular hemorrhage (IVH) as a first therapeutic option. Her peritoneum was found to be prone to VPS failure.

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Case presentation

We present the case of a 6-month-old girl with a history of preterm birth and neonatal intensive care unit (NICU) stay for respiratory distress, grade III IVH and necrotizing enterocolitis (NEC) handled conservatively. Furthermore, both her jugular veins were compromised by the implementation of Broviac lines. At follow-up, we noticed a progressive increase in head circumference. Computed tomography (CT) scan (Fig. 1) demonstrated significant communicating hydrocephalus indicating CSF diversion.

On admission, the patient was afebrile, alert with good activity. Physical examination findings were normal besides macro crania and wide tense anterior fontanel. However, her mother consistently mentioned episodic abdominal pain and mild fever. Complete blood count, C-reactive protein, and procalcitonin were normal. Stool test and urinalysis did not identify any potential pathogens. Abdominal CT scan and ultrasound (Fig. 2) unveiled thickening of the appendicular wall and mesenteric infiltration. Although rare, chronic appendicitis was the most likely diagnosis.

Considering that peritoneum might be hostile to VPS and her neonatal complications, we had few options for CSF.

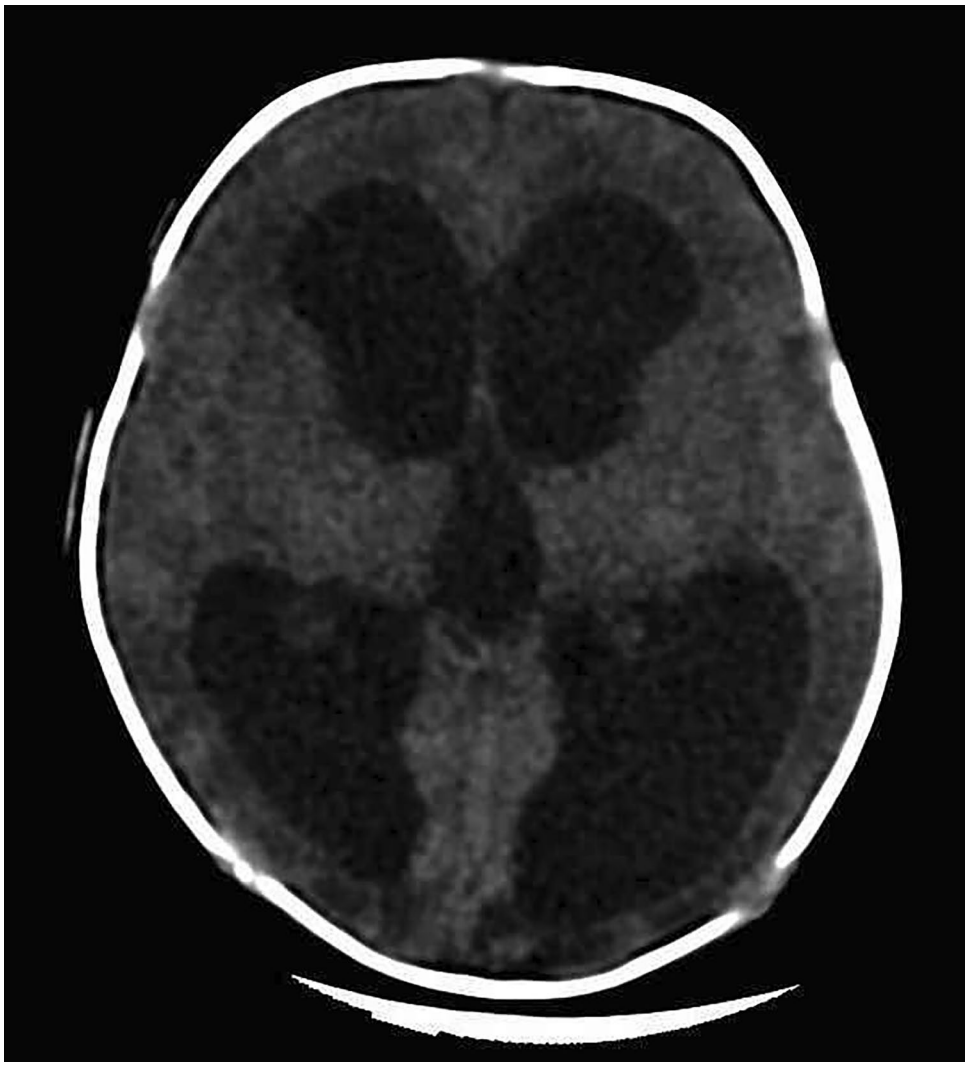

Fig. 1 Preoperative cranial CT scan showing communicating hydrocephalus

Since we had to open the abdomen for surgical exploration, the gallbladder seemed a simple accessible option for distal end placement. Surgery was planned with a pediatric surgeon. A coelioscopic first look showed extensive intraperitoneal adhesions; therefore, we converted to a median supraumbilical laparotomy. After adhesiolysis and mobilizing the caecum, the appendix appeared to be inflammatory with a cluster of mesenteric lymph nodes. An appendectomy was then performed. The pediatric surgeon presented the dome of the gallbladder into surgical view. The distal catheter was then embedded into the fundus throw a 5-mm incision and fixed with a purse-string suture around it. A 30-cm-loop of the distal catheter was left free in the abdominal cavity to fit with future growth. The infant was discharged home in stable condition 7 days after surgery. Histological findings confirmed the diagnosis of chronic appendicitis.

The current 60 months post-operatively follow-up finds normal neurological development and complete relief of abdominal symptoms. Moreover, control X-ray and sonographies (Fig. 3) were obtained, and no migration of the catheter's tip has been noted.

Discussion

Management of hydrocephalus, when the peritoneum is possibly hostile to CSF shunting, can be challenging. The gallbladder proved to be an effective and safe alternative and has been increasingly popular since first described by Smith et al. (1958) [1].

In previous studies, VGS has always been considered for cases in which VPS or ventriculo arterial shunt (VAS) are no longer feasible as a second or third-line salvage treatment [2–6].

To our knowledge, there are only five pediatric cases reporting VGS as a de novo procedure (Table 1). West et al. [6] described a patient with short bowel syndrome related to NEC, where VAS and VPS were not possible. Pal and

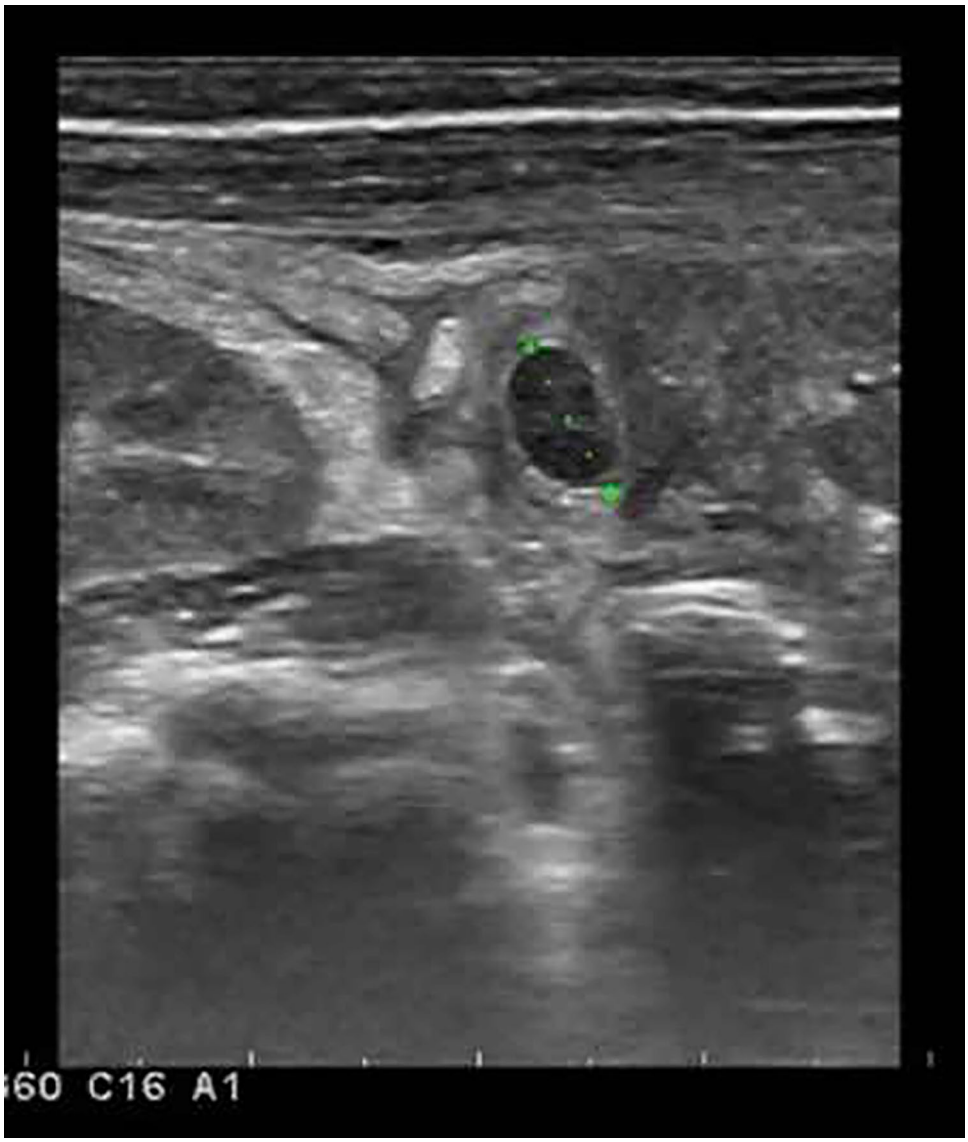

Fig. 2 Abdominal ultrasound demonstrating a 15*20 mm hypoechoic mass in the right lower quadrant (green discontinued line), suggesting appendicitis

Jindal [4] reported a similar case to ours with good clinical outcomes and revision-free until 3 years postoperatively. Aldana et al. [2] reported a case of an infant with no previous shunt but with multiple prior abdominal surgeries. Pancucci et al. [5] recorded a case of a 4-month infant presented with obstructive hydrocephalus caused by optic-chiasmal hypothalamic glioma. They stated that a high protein CSF rate predicts VPS failure since it may be responsible for ascites and non-resorptive peritoneum. Our child developed a progressive hydrocephalus and abdominal signs related to chronic appendicitis. Considering peritoneum hostility, poor venous access, and history of neonatal respiratory distress, we opted for VGS as a first-line and definitive treatment.

We suggest classifying the predictor factors of VPS failure into two groups: CSF-related disorders and abdominal-related conditions. Indeed, CSF with a high protein concentration, specifically caused by optic-chiasmal hypothalamic glioma [5, 7–9], craniopharyngioma [10], plasminogene deficiency [3], and tuberculosis [5, 11], may be subject for proteinaceous ascites and therefore VPS failure. In these cases, the gallbladder is an efficient receptacle: CSF will be excreted and then absorbed in the intestinal tract. Nevertheless, the lytic action of bile may catabolize fibrinous adhesion around the distal shunt tip CSF [1, 5, 8, 11].

Abdominal-related conditions include multiple prior abdominal surgeries, peritoneal adhesions, ascites, pseudo-occlusion, and peritonitis [5, 12, 13]. Commonly, in these cases, surgeons use the gallbladder to drain CSF as last resort, electing the atrium or the pleural cavity for the second viable receptacle. This disinclination to VGS is not counselled by standard guidelines, but mainly as a consequence of potential complications (Table 2) and technical concerns [9, 11–21, 24–26]. From our perspective, the VGS placement technique, as detailed by Morosanu et al. [19], was a

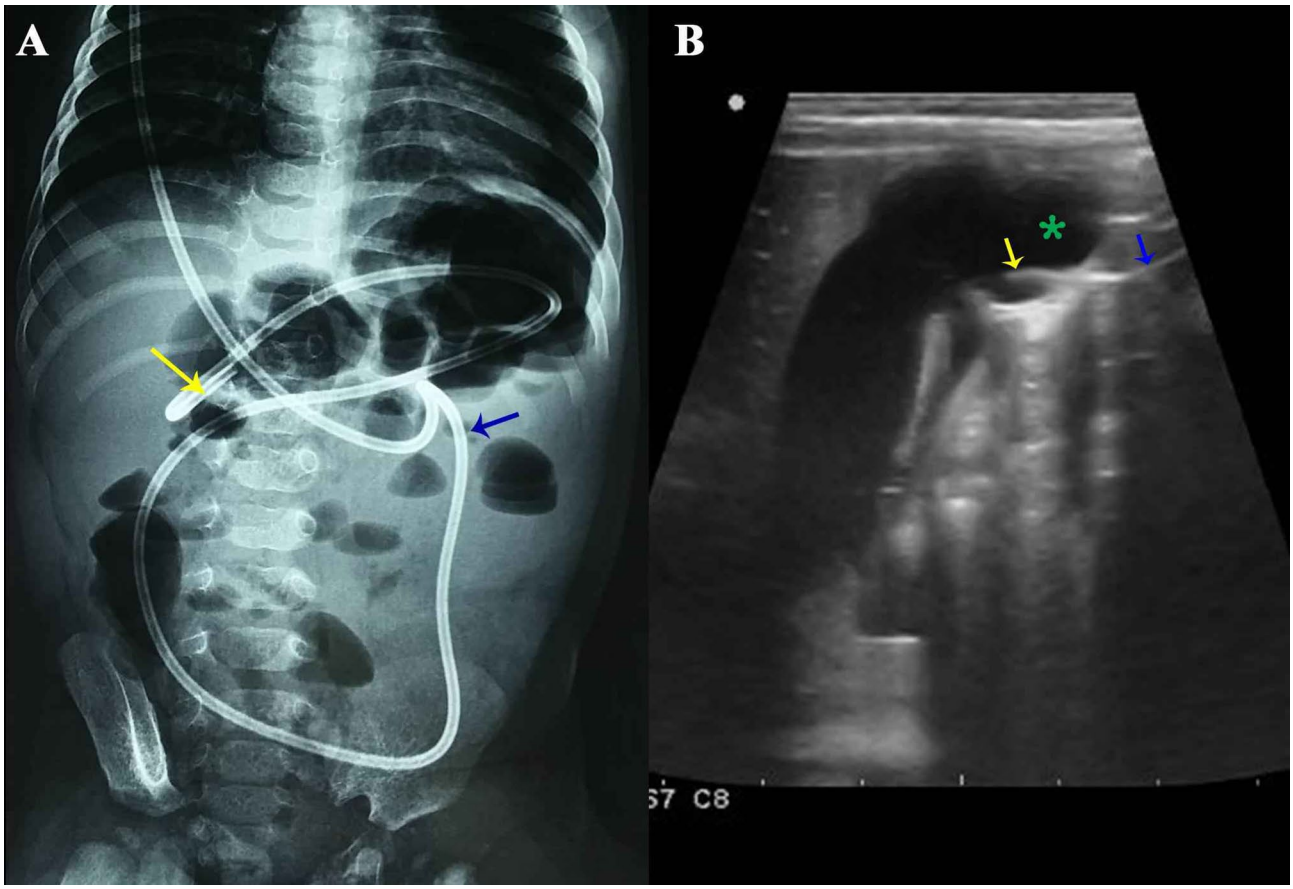

Fig. 3 Post-operative imaging X-ray **A** and ultrasound **B** showing the tip of the distal catheter (yellow arrow) embedded into the gallbladder (green asterisk) and the intraperitoneal catheter (blue arrow)

Table 1 VGS as a first-line treatment in pediatric hydrocephalus

Author	Cause of hydrocephalus	Cause of hydrocephalus	Predictor factor of VPS failure	Technique	Outcome/follow-up
West et al. (1987)	*	Myelomeningocele	Short bowel syndrome related to NEC and enterocutaneous fistula	Open laparotomy	*
Ketoff et al. (1997)	*	*	*	*	*
Pal K et al. (2007)	6 months	Post meningitic hydrocephalus	History of NEC and extensive intraperitoneal adhesions	Open laparotomy	Good outcome/3 years
Aldana et al. (2008)	8 months	*	Prior multiple intraabdominal operative procedures	Open laparotomy	*
Pancucci et al. (2019)	4 months	Obstructive hydrocephalus/ optic-chiasmal hypothalamic glioma	Extremely high protein CSF rate	Laparoscopy	Good clinical evolution/16 months
Our case	6 months	Post hemorrhagic hydrocephalus	History of NEC, concomitant appendicitis, and extensive intraperitoneal adhesions	Open laparotomy	No complication related shunt and good neurological evolution/16 months

*No data available

Table 2 Potential complications specific to VGS system

Complication	Author
Biliary ventriculitis	Barami et al. [15]
Bile peritonitis	Kulwin et al. [18]
Obstruction of the common bile duct	Parikh et al. [26]
Cholangitis	Scaife et al. [20]
Cholelithiasis	Fountas et al. [16], Alraee et al. [14], Surfield and Klein [21]
Gallbladder atony	West et al. [9]
Bile dilution/lipids. Malabsorbtion	Morosanu et al. [19]
Post-prandial headaches	Frim et al. [11]
Shunt failure due to high CSF output	Henderson et al. [17]

little different compared to VPS. It could be planned with pediatric surgery teams. Furthermore, laparoscopic access makes this approach even more valuable [5, 10]. Neonatal history of NEC may be recognized as another abdominal-related cause predicting VPS failure by leading to an inflammatory peritoneal disease and a higher incidence of distal shunt obstructions [22].

Reisner et al. [23] suggest that prior to deciding on VGB, an extensive evaluation should be conducted including a liver function test, ultrasound imaging of the gallbladder, or preferably a CT scan of the abdomen. Bile culture and CSF volume assessment is not mandatory. When a CSF external drainage is previously placed, a surgeon should be aware that a high CSF flow may overwhelm the gallbladder [17]. Up to date, our child remained complication-free. If a revision is ever needed, a surgeon must be aware that retrieval of the distal end should be under visual control in order to prevent bile leakage.

Conclusion

VGS placement requires a conventional preoperative assessment and rigorous surgical technique. There have been five documented pediatric cases that have undergone VGS as de novo shunt system. Our case adds to the existing literature a detailed illustration and long outcome data. If future reports support the current results, surgical teams may legitimately resort to VGS earlier in selected cases.

Author contribution MH: conceptualization. ZS: writing—original draft and editing. MB: review and editing. SM: review of literature. AB: methodology. MD: supervision. All authors read and approved the final manuscript.

Availability of data and material Not applicable.

Declarations

Ethics approval and consent to participate Not applicable.

Consent for publication Informed consent was obtained from a relative for the publication of this case report and accompanying images.

Conflict of interest The authors declare that they have no competing interests.

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