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Intra-abdominal cystic lesions after ventriculoperitoneal shunting

Elif Başaran Gündoğdu, Esra Özçakır

[1]Department of Neurosurgery, University of Health Sciences, Bursa Yüksek İhtisas Training and Research Hospital, Bursa, Turkey; [2]Department of Pediatric Surgery, University of Health Sciences, Bursa Yüksek İhtisas Training and Research Hospital, Bursa, Turkey

ABSTRACT

Objectives: Definitive diagnosis is essential for the medical and surgical management of pediatric patients with ventriculoperitoneal (VP) shunt. In patients with a VP shunt, abdominal complications have been well described, among which abdominal pseudo cysts are uncommon. In this report, we present our experience in terms of the multi-disciplinary management of intra-abdominal cystic lesions associated with the VP shunt procedure.

Methods: From 2016 to 2021, 245 VP shunt procedures were performed in our institution. Intra-abdominal cystic lesions were recorded as intra-abdominal complications (abdominal pseudocyst, intestinal subserosal bowel cyst, and scrotal cyst) in 3 patients. For these patients we retrospectively collected data on medical history, complaints, diagnosis, treatment procedure, and postoperative results. The study was performed on 2 male and 1 female patients. The average patient age was 11.6 months (5 months to 1.5 years). The most common complaint was that of abdominal distention with ileus symptoms. The average time of admission after the catheterization of VP shunt was 1 month; laparotomy was performed for 2 patients in whom treatment was needed for high ligation.

Results: A VP shunt operation is followed by abdominal complications in about 5%-47% of all cases. These complications are manifested as ileus symptoms, such as vomiting, abdominal distension, and abdominal pain with intestinal obstruction.

Conclusions: In pediatric patients with VP shunts, a shunt catheter-induced abdominal cystic formation should always be considered a complication. Management of these cystic lesions requires the use of a multi-disciplinary approach with neurosurgery and pediatric surgery for treatment.

Keywords: Intra-abdominal cyst, ventriculoperitoneal shunt, hydrocephalus

Placement of a ventriculoperitoneal (VP) shunt often includes complications and is the most commonly applied treatment for hydrocephalus. Shunt complications are noted in 45%–59% of all patients undergoing VP shunting [1]. The most frequently observed complications are mechanical failure, dysfunction, and infection [2]. Mechanical complications are malposition, blockage, and fracture of the shunt [2]. Infection is the most common complication, comprising 8-12% of all shunt complications. It occurs mostly within the first 6 months [2]. Other complications may also be present, including overdrainage, underdrainage, subdural hematoma, hemorrhage, obstruction, displacement, and abdominal complications [2].

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Address for correspondence: *Elif Başaran Gündoğdu, MD., University of Health Sciences, Bursa Yüksek İhtisas Training and Research Hospital, Department of Neurosurgery, Mimarsinan Mah., Emniyet Cad., 16310 Yıldırım, Bursa, Turkey. E-mail: basaran.elif@hotmail.com, Phone: +90 224 295 50 50*

The peritoneum is the most common site for the placement of a VP shunt catheter. Complications include obstruction, peritoneal pseudocysts, ascites, bowel perforations, disconnection, infection and hernias [3]. Although these complications are rarely observed in patients (0.25%-10%), their symptoms and clinical manifestations (seizures, vomiting, abdominal pain, fewer and ileus) are very serious [3]. Under these conditions, a multi-disciplinary approach is required for managing the complications. As the management of intra-abdominal lesions for children is in the purview of the pediatric surgery department, with regard to the benefits in pediatric patients, the treatment protocol should be co-administrated by the neurosurgery and the pediatric surgery departments. The present study aims to critically discuss the management of intra-abdominal VP shunt complications in the context of the literature.

METHODS

For our research, approval was obtained from Bursa Yüksek İhtisas Training and Research Hospital, Clinical Research and Ethics Committee. (Decision number: 2011-KAEK-25 2021/02-20). During the period from 2016 to 2021, 247 patients underwent VP shunt catheterization at our institution (Fig. 1). Among these,

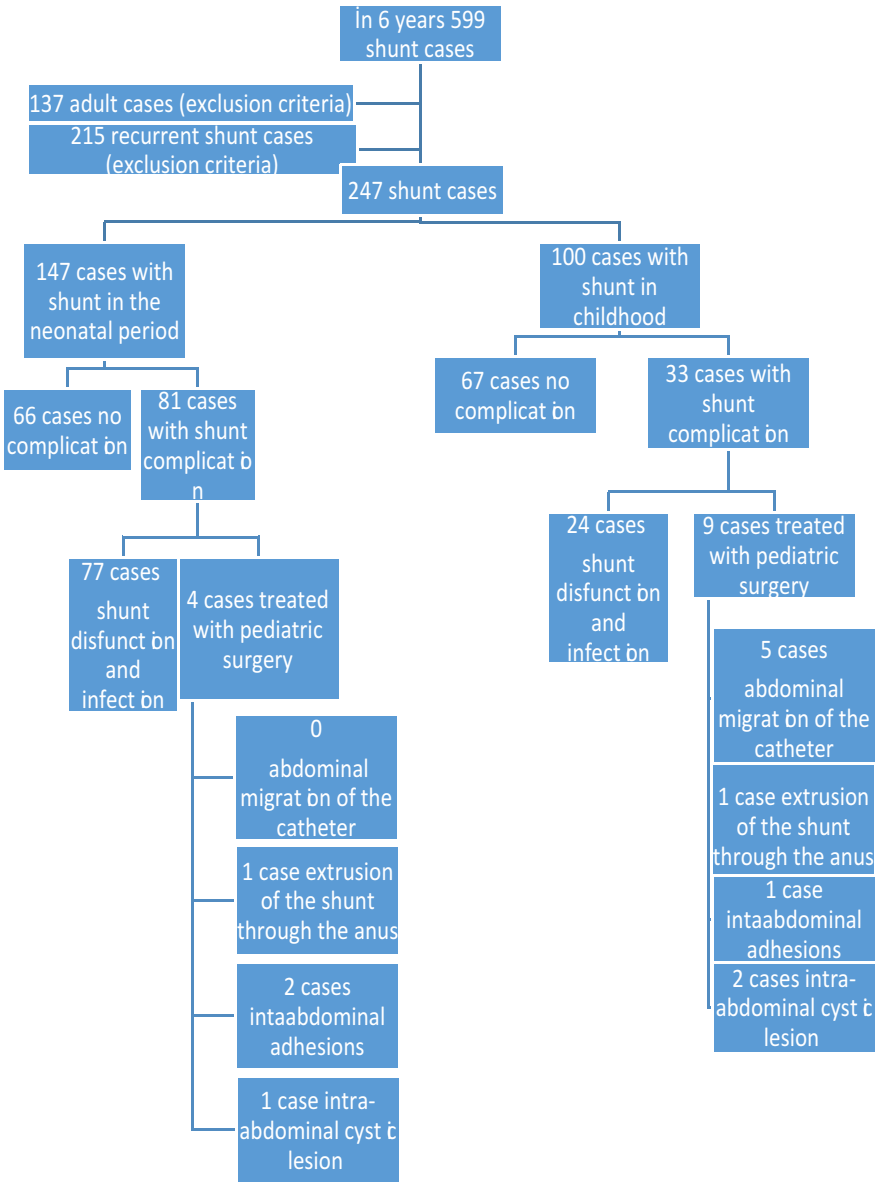

Fig. 1. During the period from 2016 to 2021, ventriculoperitoneal shunt catheterization at our institution.

Table 1. Shunt complications

Shunt complications (n = 114)	Treated with pediatric surgery								Shunt disfunction and infection	
	Abdominal migration of the catheter		Extrusion of the shunt through the anus		Intra-abdominal adhesions		Intra-abdominal cystic lesion			
	n	%	n	%	n	%	n	%	n	%
Neonatal period	0	0	1	0.87	2	1.75	1	0.87	24	21.05
Childhood period	5	4.38	1	0.87	1	0.87	2	1.75	77	67.54

37 had a shunt infection, 60 had a shunt dysfunction, 4 had overdrainage, 5 had an abdominal migration of the catheter, 2 had an extrusion of the shunt through the anus, 3 had intra-abdominal adhesions, and 3 had an intra-abdominal cystic lesion (abdominal pseudocyst, subserosal bowel cyst and scrotal cyst) related to a VP shunt catheterization and were evaluated (Table 1). This descriptive study is based on the retrospective analysis of 3 patients who were treated at the department of neurology and pediatric surgery at the Bursa Health Sciences University (Figs. 2, 3 and 4). The required data were obtained from the electronic database of our institution. Information regarding the patients such as clinical features, diagnostic methods, surgical approaches and postoperative results was recorded. The intra-abdominal cysts were treated with both laparotomy and neurosurgical interventions. All the samples were subjected to histopathological examination.

Of these 3 cases, the first one was a 5-month-old female, the second one was a 1.5-year-old boy and the third case was a 1-year-old boy. These cases presented again with abdominal symptoms (2 cases after 1 month and 1 case after 2 months of the last revision of the shunt). 1 case presented with abdominal distention, vomiting, loss of appetite, and an inability to pass gas and stool, 1 case presented with abdominal distension, vomiting and constipation, and the last one showed tenderness in the right lower abdominal quadrant.

Radiologically, the first case control magnetic resonance imaging showed an abdominal cystic lesion extending from the subhepatic area to the right inguinal region, sized 10 × 5 × 10 cm. Her abdominal

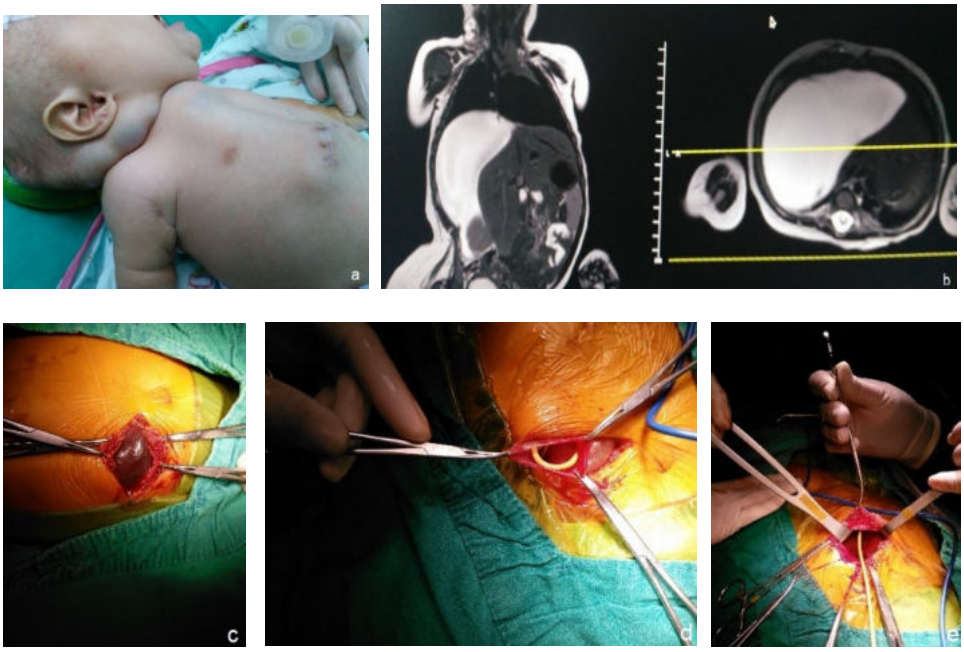

Fig. 2. (a) Preoperative view showed swelling along the shunt, (b) The MRI planning indicate intra-abdominal cystic lesion, (c) Laparatomy performed by median insicion and (d and e) VP shunt catheter, placed subserosal cystic lesion.

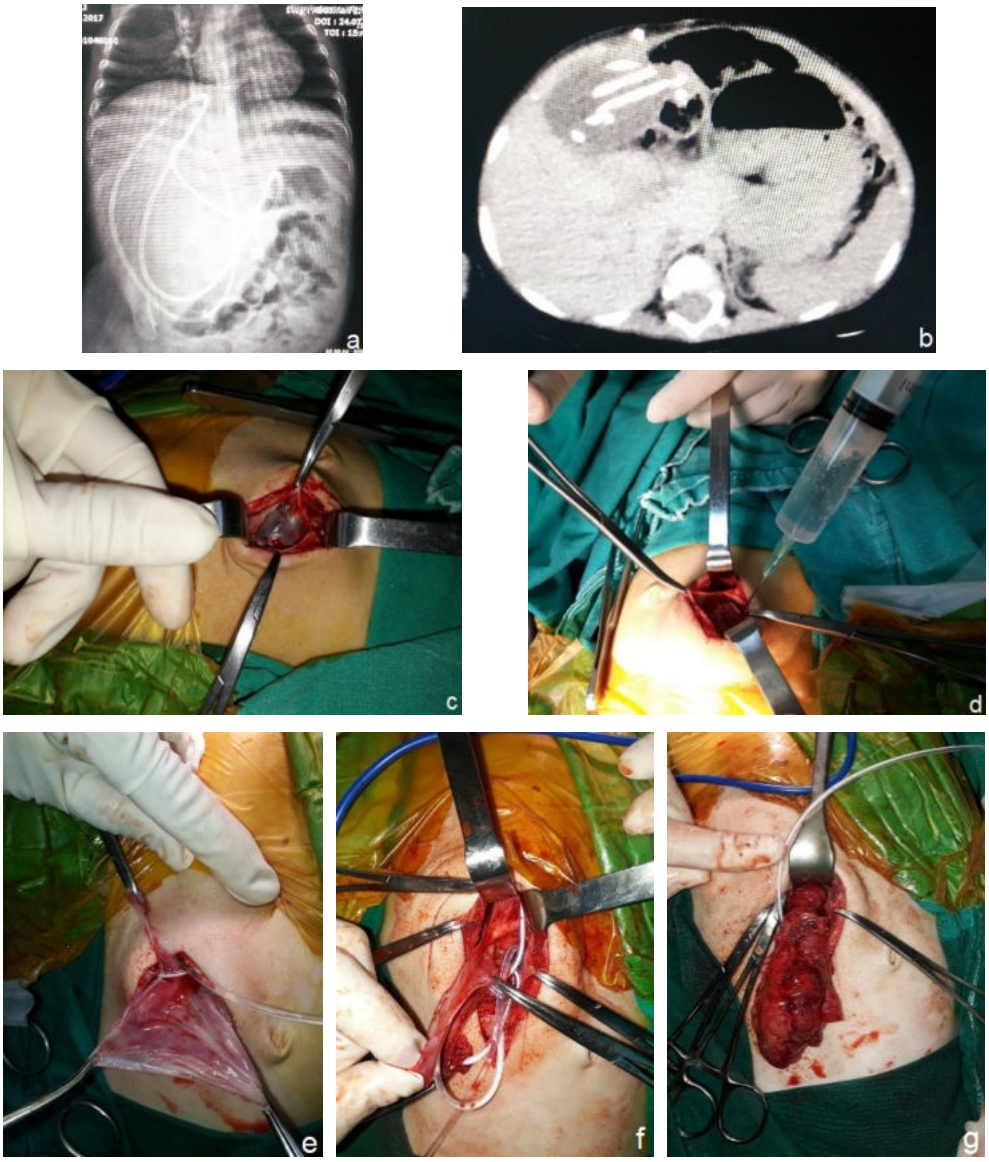

Fig. 3. (a and b) The VP shunt catheter was showed on X ray and CT sagittal image, (c) Laparatomy performed by median insicion and large pseudocyst seen, (d) cyst aspiration and (e, f, and g) cyst excision and adesiolysis performed.

radiography also confirmed ileus (Fig. 2b). In the second case, X-ray and computed tomography (CT) demonstrated a cystic mass in the abdominal cavity, measuring about 10 × 5 × 5 cm in size (Figs. 3a and 3b). In the third case, on X-ray, the size of the cyst was found to be 1 × 1.5 cm toward the shunt line to the scrotal area (Fig. 4a).

Regarding surgical intervention, in the first two cases of abdominal cyst, laparotomy and excision of the cyst and adhesiolysis were performed, then the shunt was repositioned in the right inguinal space (Figs. 2c, 2d and 2e) (Figs. 3c, 3d, 3e, 3f and 3g). In all 3 cases, a right inguinal incision was performed to approach the cystic lesion for hernia repair and high ligation procedure was performed for the cystic hernia sac; the left upper quadrant of the peritoneum was chosen for placing the new shunt catheter with laparotomy (Figs. 4b, 4c, 4d and 4e). There was no problem in the 2-year follow-up for all 3 cases.

RESULTS

A total of 3 intra-abdominal cystic formation repairs were performed in 247 patients. The patient age ranged from 5 months to 1.5 years (median, 11.6 month). One neurosurgeon and 1 pediatric surgeon individually performed all the procedures. All the cases

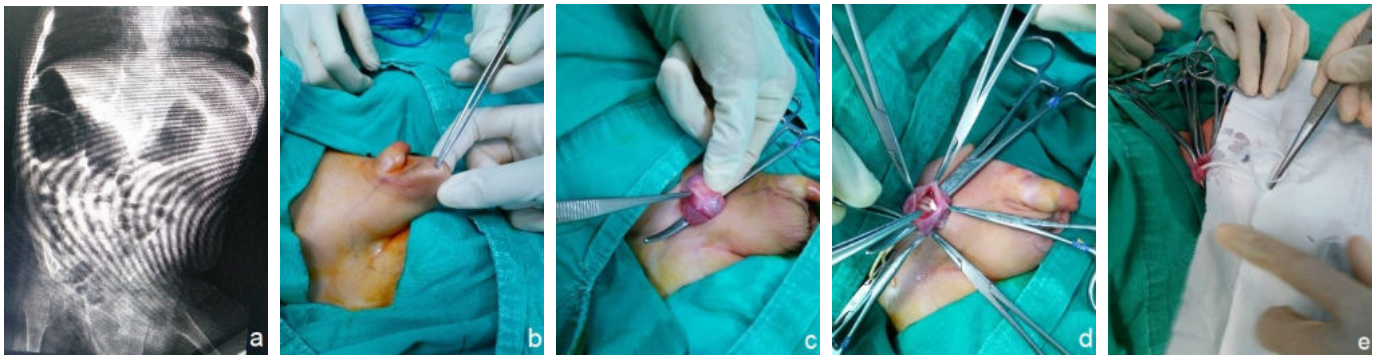

Fig. 4. (a) The VP shunt catheter was showed on X ray in the scrotal area, (b) Right inguinal incision was performed, (c) The cystic hernia sac dissected from spermatic cord and (d and e) The shunt catheter was seen and removed.

were complicated with bowel obstruction. The patients presented with increased intracranial pressure, abdominal distention and ileus symptoms. All the patients initial evaluation was performed with abdominal X-ray. After ileus was detected, cystic lesions were first determined by ultrasound. CT was performed on 1 patient to assess an identified cystic mass and make a treatment decision. A unilateral high inguinal approach was used on 1 patient, and laparotomy was performed on 2 patients for cystic lesion treatment. The histopathological evaluation revealed a mesothelial cyst in 2 patients and a simple cyst in 1 patient.

DISCUSSION

In our clinic, a VP shunt is commonly used for hydrocephalic patients who are ineligible for a third ventriculostomy. VP shunts' complications are common in pediatric and adult patients with a reported incidence rate of 45% and 59%, respectively [1]. The complication rate seen in our cases was 46%. Several predisposing factors, including infection, multiple shunt revisions, obstruction or dislodgement and a peritoneal foreign body reaction have been suggested; however, the pathophysiology remains unclear [4].

The most common intra-abdominal complications with VP shunt are infection, malfunction, disconnection and catheter migration. Infection is the most common complication, comprising 8-12% of all shunt complications. It occurs mostly within the first 6 months [2]. Infection was observed in 37 of our 247 cases (this includes cases of shunt infection referred from other institutions). The most common agent was Staphylococcus epidermidis, which is consistent with the literature.

In addition, complications related to equipment failure, such as extraperitoneal retraction of the shunt catheter, subcutaneous collection of CSF and peritoneal pseudocyst formation, are noted in patients with VP shunt [5, 6]. Intestinal perforation, CSF ascites, inguinal hernia and intestinal volvulus are rare complications that are associated with a high morbidity rate [5]. Of our 247 cases, 147 shunts were inserted in the neonatal period and 100 in the post-neonatal period. In 13 (5.2%) of 247 cases, abdominal complications that required the intervention of pediatric surgery developed, and no additional complications developed after the intervention. Of 247 cases, 5 had an abdominal migration of the catheter (in cases of subduroperitoneal shunt), 2 had extrusion of the shunt through the anus, 3 had intra-abdominal adhesions, and 3 had an intra-abdominal cystic lesion. The rate of intra-abdominal complications, that recovered with the combined effort of neurosurgery and pediatric surgery, was 5%.

An abdominal pseudo cyst is an uncommon manifestation of VP shunt catheterization [6]. Causes of VP shunt-related intestinal obstruction vary from intestinal perforation to mechanical obstruction caused by the mass effect of the cystic structure. Intestinal volvulus is the most common cause in the pediatric population with a VP shunt, a mechanical obstruction due to the twisting of the shunt catheter is the second most common cause. In some cases, obstruction occurs when a loop of the shunt catheter is tightened around a bowel loop during removal [7-9]. Peritoneal CSF pseudo cysts are a rare but significant complication of VP shunt surgery, with a reported incidence rate ranging from 10% to 0.25% [3]. Based on the data of our patients, the annual incidence rate of pseudocysts

is 1.21%.

Dabdoub *et al*. [10] reported that 295 of their 393 patients presented with abdominal pseudocysts related to the VP shunt procedure. Further, 33% of these patients were < 10 year-old, and the recurrence ratio of pseudocyst formation due to VP shunt was 19.8%. In our patients, no recurrence was observed at the 2-year follow-up.

In abdominal pseudocyst formation, the time till the occurrence of complications and symptoms ranges from 3 weeks to 5 years [5]. However, as per some case reports on pseudocyst formation of CSF, the pseudocyst developed between 5 days to 25 months after the VP shunt procedure. The average duration between VP shunt operation and abdominopelvic CT was 11 months (range, 1 week to 115 months) and the average number of VP shunt procedures was 1.4 (range, 1-6) [5]. The period till the development of the pseudocyst was 1 month in 2 of our patients and 2 months in the third patient.

CSF pseudocysts have a variable appearance and may impair CSF absorption [11]. Bowel obstruction is a rare complication. Larger CSF pseudocysts tend to be noninfectious, while smaller cysts or multiloculated cysts may cause an infection [11]. Pathi *et al*. [12] reported no significant relationship between pseudocyst size and infection. As per Ersahin *et al*. [13], Staphylococcus epidermidis may be present in pseudocysts that develop in the first year. Our patients had no infective process, and they each had a single cyst.

From a clinical perspective, concerning patients with a VP shunt, pediatric patients mainly present with symptoms of elevated intracranial pressure, such as a headache and nausea, while adults predominantly exhibit local abdominal symptoms [14]. Clinical symptoms are similar to those in acute abdomen, such as abdominal pain with/without a palpable mass, abdominal distension with/without tenderness, nausea and/or vomiting. In addition, decreased appetites, constipation, fever, and signs of shunt malfunction, such as lethargy and headache may be present [6]. In keeping with previous findings, our patients exhibited the symptoms of abdominal distention; vomiting; appetite loss and ileus symptoms, such as the inability to pass gas and stool.

A diagnosis of intra-abdominal shunt complication is based on clinical findings and imaging diagnostic studies (abdominal radiography, US and CT). Abdominal radiographs help rule out other causes of acute abdomen and enable a decision regarding the continuity of the catheter tube. Although the results of these modalities may be normal, ileus should be considered in the differential diagnosis [15].

Although abdominal radiography is a first-line radiological tool that is useful for evaluating patients with a VP shunt, US is important for the initial diagnosis of abdominal CSF pseudocyst [6]. US easily reveals cystic lesions in terms of the distances, localization and surrounding tissues. US may also help detect the distal end of the VP shunt catheter, in a similar way to radiography [15-17].

CT is the best noninvasive imaging method that enables the treatment decision procedure of APC by visualizing complete abdominal anatomy [18]. Thus, it provides diagnostic information about pseudocyst localization and may help identify the type of cystic lesion [6, 18]. In the present study, the initial evaluation for all the patients was performed using abdominal radiography and US. Thereafter, all cystic structures were confirmed on abdominal CT because of the treatment procedure.

As stated by Hamid *et al*., patients with intra-abdominal cysts after VP shunt catheterization should be managed individually [14]. The treatment options include percutaneous drainage and both surgical approaches, laparotomy and laparoscopy [3].

Percutaneous drainage of the cyst initially resolves the obstruction; however, this involves a high recurrence risk [3]. In particular, in patients with ileus, an urgent surgical procedure must be initially considered for both VP shunt removal and cyst excision. In general practice, it is not uncommon to encounter an abundance of adhesions between cystic formation and abdominal viscera and/or between the viscera themselves due to VP shunt catheterization. Thus, in most cases, the laparoscopic approach described by Kim *et al*. [19] for the management of a CSF pseudocyst is not routinely preferred, and depends on the experience of the surgeon and adhesions.

Considering visceral damage or perforation during adhesiolysis, laparotomy is safer than laparoscopy. Laparotomy provides either repositioning or exteriorization of a VP shunt catheter simultaneously during operation and adhesiolysis can be performed with minimal damage [14]. Thus, cyst excision was per-

formed using laparotomy in our cases. In addition, the shunts of all of our patients were repositioned in the peritoneal cavity with ease during laparotomy.

The authors believe that these complications are attributable to the fact that peritoneal catheters are longer than needed; however, previous literature shows contradictory findings. To our knowledge, this is the first study to demonstrate that the use of 90-cm-long peritoneal catheters in neonates and infants is safe and effective. It does not increase the incidence of abdominal complications and prevents the need for revision for insufficient length of the peritoneal catheter [20].

The cysts of CSF are surrounded by a wall of non-epithelial tissue, such as intestinal serosa, peritoneum, or intra-abdominal organ surfaces [14]. Histopathological evidence demonstrates the presence of inflamed serosal lining, fibrous tissue, inflammatory cells and granulomatous tissue consisting of fibroblasts and collagen with inflammatory cells [14]. Our histopathological results were consistent with previous findings in the literature.

CONCLUSION

Abdominal cysts are rare complications of VP shunt. In 5 years, 3 of our 247 patients developed abdominal complications, including an abdominal pseudocyst, subserosal bowel cyst and a scrotal cyst. There was no relevant infection. Abdominal complications should always be considered when a patient with a VP shunt presents with abdominal symptoms.

Ethical Approval

The study was initiated after obtaining approval from the Bursa Yüksek İhtisas Training and Research Hospital local ethics committee (Decision number:2011-KAEK-25 2021/02-20). All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Informed consent was obtained from all individual participants in the study.

Authors' Contribution

Study Conception: EBG, EÖ; Study Design: EBG, EÖ; Supervision: EBG, EÖ; Funding: N/A; Materials: N/A; Data Collection and/or Processing: EBG; Statistical Analysis and/or Data Interpretation: EBG, EÖ; Literature Review: EBG; Manuscript Preparation: EBG, EÖ and Critical Review: EBG, EÖ.

Conflict of interest

The authors disclosed no conflict of interest during the preparation or publication of this manuscript.

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