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A 10-Year Experience in the Treatment of Intraabdominal Cerebrospinal Fluid Pseudocysts

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A B S T R A C T

The aim of this retrospective study was to assess a ten-year experience in the treatment of rare complications of ventriculoperitoneal shunting – intraabdominal cerebrospinal fluid pseudocysts. At this time there are no data about incidence, clinical course and treatment of these complications in Croatia. Cerebrospinal fluid (CSF) abdominal pseudocyst is an uncommon but important complication of ventriculoperitoneal shunts. Retrospective data were obtained from 5 children with abdominal CSF pseudocysts, treated between 1996 and 2007. The incidence of intraabdominal CSF pseudocysts in our study is 2.9%. All patients were girls ranged in age from 4 to 12 years old (mean 8.8 years). In most cases etiology of hydrocephalus was congenital, idiopathic. Abdominal pain and distension were the most frequent clinical finding (4/5). Although infection has been reported as responsible for pseudocyst formation, we did not found it in our series. Laparotomy with cyst wall excision and catheter replacement was performed in 2/5 cases, and only cyst fluid aspiration with catheter replacement in 3/5 cases. Recurrence of the abdominal cyst was observed in one girl who was in terminal stadium of anaplastic ependymoma. It is our opinion that only catheter replacement and cyst fluid evacuation, as one of the treatment modalities, may be successful, even in large CSF intraperitoneal pseudocysts.

***Key words**: hydrocephalus, CSF pseudocyst, ventriculoperitoneal shunt*

Introduction

Insertion of ventriculoperitoneal shunts is well known and very successful treatment of hydrocephalus, especially in the childhood. The abdominal intraperitoneal cerebrospinal fluid (CSF) pseudocysts are infrequent complications of ventriculoperitoneal shunts[1–3]. At this time there are no data about incidence, clinical course and treatment of these complications in Croatia. Furthermore, some neurosurgeons never observed this rare clinical condition. Although they are not life-threatening, the diagnosis and treatment of these complications can be difficult. In some studies, the incidence of this complication is the lowest of all CSF shunt complications (1.6–4.5%), with the equal incidence in children and in adults[3,4]. The etiology is not always clear, but the most suggested mechanisms of CSF pseudocysts formation are infections and multiple shunt revisions[2,4–6]. Ultrasonography is the method of choice for the diagnosis of this complication[2,7]. The treatment is not unique and involves removal of peritoneal catheter and placement of new catheter intraperitoneal in a different quadrant, with or without excision of pseudocyst wall by laparotomy or by laparoscopy, by performing ventriculo-atrial shunt or endoscopic third ventriculostomy[2,3,8–13]. The results are different and recurrences are possible[2,3,5]. This report presents our experience in the treatment of these complications over a ten-year period.

Subjects and Methods

Retrospective data were obtained from 5 children with cerebrospinal fluid abdominal pseudocysts treated

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in the period from 1997 to 2006. The incidence of CSF pseudocysts in our study, based upon the number of shunt complications, is 2.9%. All the patients were girls aged at the time of CSF cyst diagnosis between 4 and 12 years (mean: 8.8 years). Although the most suggested predisposing factors of CSF pseudocysts formation are multiple shunt revisions, all of our patients have had only initial shunt procedure, without any shunt revisions prior to occurrence of CSF pseudocyst. The reasons for V-P shunting were congenital hydrocephalus (3/5), one girl had postmeningitis obstructive hydrocephalus, and another one had obstructive hydrocephalus due to malignant ependymoma located in the posterior fossa. All of three congenital hydrocephaluses were treated in the first two months of girl's life, postmeningitic hydrocephalus in a nine months old girl, and the last one at a girl aged two years. At the time of CSF cyst diagnosis there were no previously noted shunt dysphunctions, infections, or shunt revisions. The main symptoms were abdominal pain with abdominal wall distension (4/5) without signs of valve malfunction. One girl had shunt dysphunction with clinical signs of elevated intracranial pressure (headache, nausea, drowsiness). No one of our patients had enlargement of ventricular size.

Diagnosis of CSF pseudocysts was made by ultrasonography and/or computed tomography. Although we sampled the cyst fluid for further microbiological and biochemical analysis we did not measure the volume of cyst fluid, except in one patient with the largest cyst (containing almost 2 litres of CSF fluid).

Figure 1 and 2 show a large intraabdominal pseudocyst, around the tip of the distal catheter, located in the left and part of the right hemiabdomen.

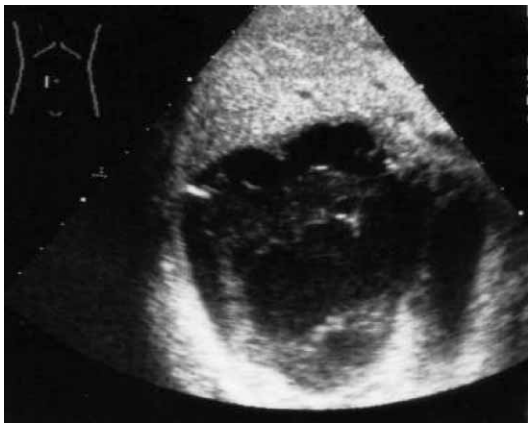

Fig. 1. Abdominal ultrasound showing large intraabdominal cyst.

Surgical technique

In two girls pseudocysts were treated by laparotomy, with complete or partial cyst wall excision and transposition of the catheter. In 3 out of 5 patients the cyst fluid was aspirated first through the distal part of ventriculoperitoneal shunt. Catheter was than replaced in another abdominal quadrant via a small abdominal wall incision, thus avoiding a conventional laparotomy and the consequent risk of further complications. Cyst aspiration or cyst fluid sampling during laparotomy permitted further microbiological and biochemical analysis. There were no microbiological signs of bacterial or parasitic infections of cyst fluid, and biochemical analysis confirmed cerebrospinal fluid presence in the cysts. In our case 1 we sampled almost 2 liters of cyst fluid.

TABLE 1
PATIENT DATA

Patient	Gender	Initial diagnosis	Age at V-P shunting	Age at CSF cyst diagnosis	Symptoms
1	female	congenital hydrocephalus	1 month	12 years	abdominal pain and distension
2	female	congenital hydrocephalus	2 months	8 years	abdominal pain and distension
3	female	congenital hydrocephalus	1 month	11 years	abdominal pain and distension
4	female	postmeningitis hydrocephalus	9 months	9 years	abdominal pain and distension
5	female	infratentorial anaplastic ependymoma	2 years	4 years	elevated intracranial pressure

TABLE 2
TREATMENT AND RECURRENCES OF CEREBROSPINAL FLUID PSEUDOCYSTS

Patient	Laparotomy with cyst wall excision and catheter transposition	Cyst aspiration with catheter transposition	Recurrences
1	+	–	–
2	–	+	–
3	–	+	–
4	–	+	–
5	+	–	+

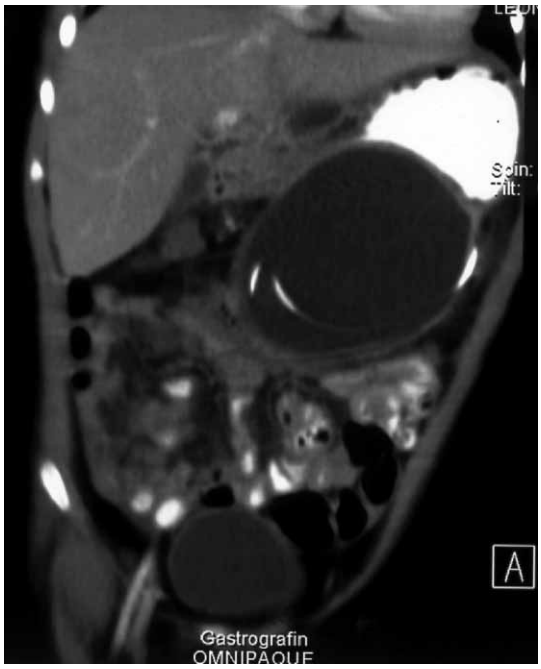

Fig. 2. Parasagital abdominal CT scan showing cyst around the tip of the distal part of V-P shunt.

Results

In for patients postoperative ultrasound showed good regression of the cyst volume, sometimes with some residual cyst fluid. After the treatment all symptoms disappeared promptly, without cyst recurrences. These patients are still followed, and there are no neurological or abdominal symptoms at present time. Patients without conventional laparotomy had much faster postoperative recovery. A patient treated for anaplastic intracranial ependymoma quickly developed recurrent intra-abdominal cyst after initial laparatomy. She required another laparotomy with cyst wall excision. Furthermore**,** postoperative course was complicated with paralytic ilues. Three months after the last shunt revision the girl died. This is the only patient in our study with predictable cause of cyst formation.

Discussion

Ventriculoperitoneal shunting of cerebrospinal fluid is the standard therapy for the management of hydrocephalus[14]. This method has a lower morbidity than ventriculo-atrial shunts and other methods of CSF diversion, and severe complications are uncommon[1,15].

Abdominal complications of VP shunts are now rare events; however, their frequency varies in different studies from 5 to 47%[12]. They are various and include shunt migration and obstruction, infections, intestinal, bladder and vaginal perforations, perforations of viscera, bowel obstruction, development of inguinal hernia or hydrocele, CSF ascites, volvulus[1,10,12,15–19]. As opposed to these references, some authors found abdominal shunt revisions frequently necessary[9].

The abdominal intraperitoneal CSF pseudocysts are also known, but infrequent complications of VP shunts[1–3]. The incidence of CSF pseudocysts is the lowest of all CSF shunt complications; it varies in different studies from 0.7 to 4.5%[3,4].

The etiology is not clearly understood, but the most suggested predisposing factors of CSF pseudocysts formation are multiple shunt revision (the number of shunt revisions prior to formation of pseudocyst is important, but not always), infections, peritoneal adhesions and malabsorption of cerebrospinal fluid[5,6]. It is evident that an inflammatory process is a more strongly predictive factor[2,4]. The cause of infections may be microbiologically diagnosed (S. epidermidis, other gram positive or gram negative microorganisms, meningeal or peritoneal tuberculosis, neurocysticercosis, Ascaris lumbricoides), but in many cases the infection is caused by an organism which is difficult to culture[4,7,20]. It seams that abdominal shunt failure and pseudocyst formation can also be caused by allergic response to the shunt materials or by the treatment that the shunts receive prior to implantation (ethylene oxide)[20,21]. The important predisposing factors are also malposition of the shunt and central nervous system tumors[2].

The most common clinical presentation of CSF abdominal pseudocysts are digestive symptoms (abdominal pain and distension, palpable abdominal mass) which can produce confusion with other forms of abdominal cysts and symptoms of elevated intracranial pressure[3,20,22]. There is often no evidence of shunt malfunction[2,17].

The diagnosis is mainly made by ultrasonography, which is today the method of choice for the diagnosis of abdominal CSF pseudocysts, computed tomography and plain x-ray which can visualize the continuity and position of distal part of ventriculoperitoneal shunt[2,7].

The treatment of abdominal CSF pseudocysts is not unique. Some authors propose replacement of the shunt from the peritoneal cavity and performing a ventriculo-atrial shunting, and then to proceed to a laparotomy, drain the fluid and resect the cyst wall[3,10]. Other authors contended that the cysts reabsorbed spontaneously without excision of cyst wall or aspiration of cyst fluid. In the latter cases only replacement of a new catheter in a different abdominal quadrant was sufficient, once the cyst had reabsorbed[2]. Simple paracenthesis with catheter replacement and aspiration of cyst contents via distal catheter followed by ventriculo-atrial shunting are described also as sufficient[4,13,23,24].

Recently, laparoscopic technique is effective in catheter replacement or cyst wall excision within the abdominal cavity in many cases, thus avoiding a conventional laparotomy and the consequent risk of adhesions, which could cause further complications[8,9].

Some authors had higher rate of recurrences when performing only relocation of the shunt outside the pseu-

docyst and intraabdominally repositioning[5,23]. Others had no problems with cyst recurrence despite the intraperitoneal shunt replacement[2,3].

It is our opinion that only cyst fluid aspiration and catheter replacement, as one of the treatment modalities, may be successful, even in large CSF intraperitoneal pseudocysts. Some residual cyst volume can persist without any symptoms. If it is necessary, laparotomy with cyst wall excision, and (or) ventriculoatrial shunting can always be done.

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DESETOGODIŠNJE ISKUSTVO U LIJEČENJU LIKVORSKIH INTRAABDOMINALNIH PSEUDOCISTI

SAŽETAK

Cilj ove studije bila je procjena učestalosti i uspješnosti liječenja likvorskih intraabdominalnih pseudocisti, rijetke komplikacije liječenja hidrocefalusa ventrikuloperitonealnim likvorskim derivacijama, u desetogodišnjem razdoblju. U Hrvatskoj do sada nije bilo studija ovih rijetkih komplikacija. Retrospektivno je analizirano petero bolesnika (incidencija je 2,9%) od kojih su sve bile djevojčice, pri postavljanju dijagnoze stare između 4 i 12 godina (prosjek 8,8 godina). Inicijalna dijagnoza zbog koje je učinjena ventrikuloperitoneostomija bila je u tri djevojčice kongenitalni hidrocefalus i njima je likvorska derivacija postavljena u prva dva mjeseca života. Jedna je djevojčica u devetom mjesecu života liječena radi postmeninitičkog hidrocefalusa, a jedna u drugoj godini života radi obstruktivnog hidrocefalusa uzrokovanog anaplastičkim ependimomom stražnje lubanjske jame. Vodeći je klinički simptom u većine pacijanata bila abdominalna bol s distenzijom trbuha, a samo u jednog disfunkcija derivacijskog sistema sa znakovima povišenog intrakranijskog tlaka. Ciste su u dvije pacijentice tretirane klasičnom laparotomijom s ekscizijom stijenke ciste i transpozicijom katetera. U tri je pacijentice učinjena samo aspiracija sadržaja ciste kroz distalni kateter s transpozicijom katetera. Recidivna cista zamijećena je i tretirana kod pacijentice s primarnom dijagnozom anaplastičkog ependimoma. Iako se u literaturi kao relativno česti uzrok nastanka likvorskih intraabdominalnih pseudocisti spominje infekcija, u ovoj je studiji analizom sadržaja cisti nismo dokazali. Prema našim iskustvima, jednostavna aspiracija sadržaja ciste kroz kateter, s transpozicijom katetera, efektna je i poštedna metoda liječenja ovih komplikacija, a klasična laparotomija ostaje kao metoda izbora u liječenju eventualnih recidiva cista.