Case Report

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Spontaneous Acute Subdural Hematoma due to Intracranial Hypotension Secondary to Lumboperitoneal Shunt: A Case Report and Review of the Literature

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ABSTRACT

Objective: To report a rare case with spontaneous intracranial acute subdural hematoma due to overdrainage of cerebrospinal fluid after lumboperitoneal shunting and to review the literature on this topic.

Case report: A 53-year-old lady with spontaneous acute subdural hematoma developing 3 years after lumboperitoneal shunting for treatment of benign intracranial hypertension is reported. She was treated with shunt removal and hematoma evacuation.

Material and Methods: We found 16 cases with intracranial bleeding developing spontaneously or after mild head injury after lumboperitoneal shunting. The characteristics of the patients were recorded, the outcome was given according to the Glasgow Outcome Scale, and a Glasgow Outcome Scale score from 1-3 was accepted as worse outcome. The factors affecting outcome were evaluated.

Results: There were 10 females and 7 males aged 59.7±15.1 years. In most cases, the primary disease treated by lumboperitoneal shunting was hydrocephalus or benign intracranial hypertension. In 12 of the cases, the bleeding happened into the subdural space and in 5 into other compartments (intracerebral or subarachnoid bleeding). Five of the cases died, and 62.5% had a worse outcome. The only factor affecting outcome was the time span from lumboperitoneal shunting to intracranial bleeding. This time was significantly shorter in the patients with worse outcome (3.7 versus 38.6 months).

Conclusions: Lumboperitoneal shunting may cause serious complications such as intracranial bleeding due to overdrainage of cerebrospinal fluid via shunt. Patients with lumboperitoneal shunting must be followed very closely for development of intracranial hypotension especially during the first few months after shunting.

Keywords: acute subdural hematoma, intracranial hemorrhages, intracranial hypotension, lumboperitoneal shunting

ÖZET

Lumboperitoneal şanta bağlı gelişen spontan akut subdural hematom: Olgu sunumu ve literatür taraması

Amaç: Lumboperitoneal şant uygulanması sonrası beyin omurilik sıvısının aşırı drenajına bağlı spontan kafaiçi akut subdural hematom gelişen nadir bir olgunun sunulması ve bu konuda literatür taraması yapılması.

Olgu Sunumu: Benign kafaiçi hipertansiyon tedavisi için lumboperitoneal şant uygulanmasından 3 yıl sonra spontan akut subdural hematom gelişen 53 yaşında kadın olgu sunuldu. Hasta şantın çıkarılması ve hematomun boşaltılması ile tedavi edildi.

Yöntem ve Gereçler: Literatürde lumboperitoneal şant takılması sonrası kendiliğinden ya da hafif kafa travmasıyla kafa içi kanama gelişen 16 olgu bulduk. Hastalıkların özellikleri kaydedildi, sondurum Glasgow sondurum skalasına göre verildi ve Glasgow sondurum skala derecesi 1-3 olması kötü sonuç olarak kabul edildi. Sondurumu etkileyen faktörler değerlendirildi.

Bulgular: Toplam 10 kadın ve 7 erkek olgu vardı, yaşları 59.7±15.1 idi. Lumboperitoneal şantla tedavi edilen asıl hastalık olguların çoğunda hidrosefali ya da benign kafaiçi hipertansiyondu. Olguların 12'sinde kanama subdural alana, 5'inde diğer kompartmanlaraydı (intraserebral ya da subaraknoid). Olguların 5'i ölmüştü ve kötü sondurum oranı %62.5 idi. Sondurumu anlamlı olarak etkileyen tek faktör lumboperitoneal şant takılmasından kafaiçi kanamaya kadar geçen süre idi. Bu süre kötü sonuçlanan olgularda anlamlı olarak kısaydı (3.7'ye 38.6 ay).

Sonuçlar: Lumboperitoneal şant uygulanması şanttan aşırı beyin omurilik sıvısı drenajına bağlı kafaiçi kanama gibi ciddi komplikasyonlara neden olabilir. Lumboperitoneal şantlı olgular özellikle şant takılmasından sonraki ilk birkaç ay olmak üzere çok yakından izlenmelidir.

Anahtar kelimeler: akut subdural hematom, Kafaiçi kanama, kafaiçi hipotansiyon, lumboperitoneal şant uygulanması

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Introduction

Lumboperitoneal (LP) shunting is frequently used to treat certain disorders such as benign intracranial hypertension and communicating hydrocephalus. It may be performed percutaneously and it is a minimally invasive operation that is simpler than ventriculoperitoneal (VP) shunting because it does not required ventricle catheterization (1). However, some serious complications may be seen after insertion of an LP shunt, too, the most important one being the development of intracranial hypotension due to overdrainage of cerebrospinal fluid (CSF) (2). Actually, the rate of this complication is quite high in patients with LP shunt. In a review on idiopathic intracranial hypertension, Friedman and Jacobson (3) reported a rate of up to 25%. Fortunately, in most cases symptoms and signs of intracranial hypotension due to LP shunt are usually mild. However, this intervention may cause very serious complications such as development of intracranial bleeding. Very few cases with spontaneous intracranial bleeding secondary to LP shunting have been reported in the literature.

We report a woman with development of spontaneous supratentorial acute subdural hematoma (ASDH) due to overdrainage of CSF via LP shunt. In addition, we review the literature for development of intracranial bleeding spontaneously or after mild head injury in patients with LP shunt and we discuss the characteristics of those patients.

Case Report

A 53-year-old woman had been admitted with complaint of suddenly and spontaneously developing severe headache 2 days earlier. There was benign intracranial hypertension in her history, and she had been treated with LP shunting in another center 3 years previously (Figure 1). Her neurological examination revealed no findings. A thin ASDH located in the right temporoparietal region, 7 mm in thickness, was seen on brain computerized tomography (CT) (Figure 2). Because of possible intracranial hypotension due to her history of LP shunting, brain magnetic resonance imaging (MRI) with intravenous contrast enhancement was performed and prominent pachymeningeal enhancement and thickening was seen (Figure 3). It was thought that spontaneous ASDH had

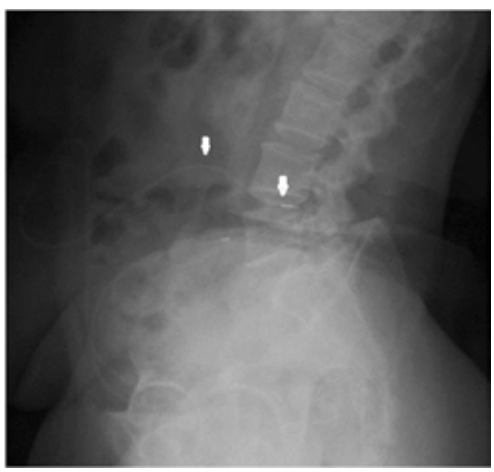

Figure 1: Lateral abdominal X-ray showing lumboperitoneal shunt (white arrows).

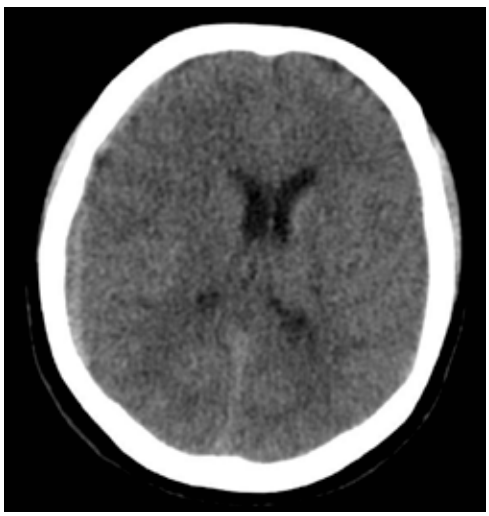

Figure 2: Brain CT showing thin subdural hematoma on the right side.

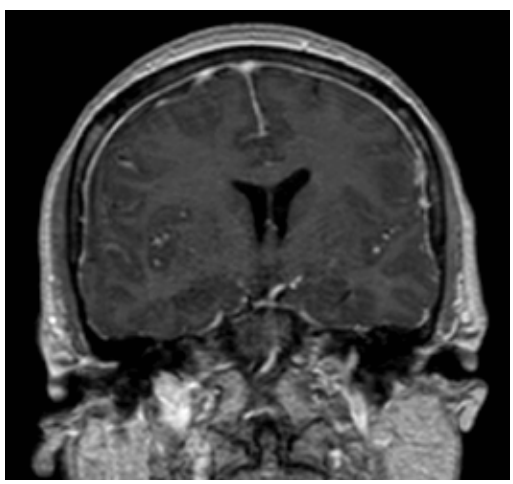

Figure 3: Coronary T1-weighted section of the brain MRI with intravenous gadolinium showing diffuse and thick pachymeningeal enhancement.

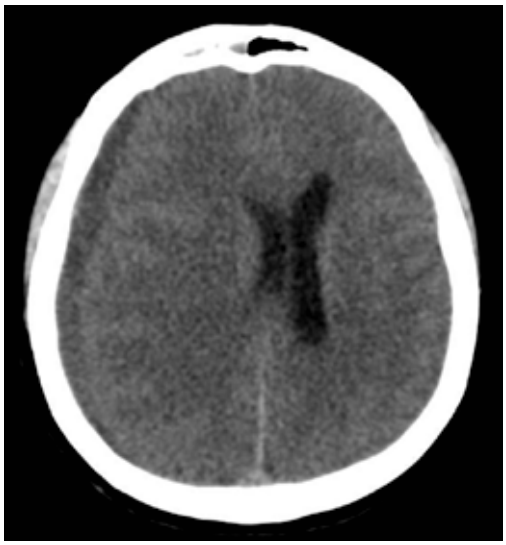

Figure 4: Brain CT 10 days after shunt removal showing chronic subdural hematoma.

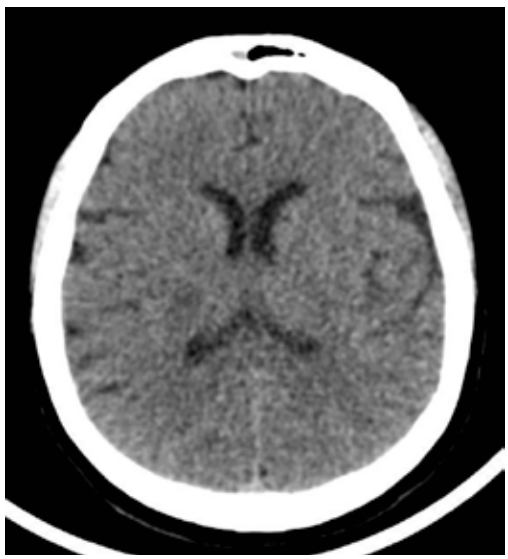

Figure 5: Normal brain CT 2 years later.

developed due to overdrainage of CSF via LP shunt. The LP shunt was removed and its entrance point to the lumbar dura was repaired with a piece of muscle and tissue glue. It was a valveless T tube catheter. The severe headache resolved after removal of the shunt. On control CTs, the hematoma increased in size and changed to chronic subdural hematoma (Figure 4). Subsequently, it was drained via a burr hole 10 days after the removal of the LP shunt. Neither subdural hematoma recurred, nor benign intracranial hypertension developed during 2 years' follow-up (Figure 5).

Material and Methods

Review of the Literature

We reviewed the literature for development of intracranial bleeding spontaneously or after mild head injury in patients with LP shunt. Jones (4) reported 63 cases with spinoperitoneal shunt performed for treatment of communicating hydrocephalus in 1966, and there was one case dying due to subdural hematoma developing after lumbar arachnoid-Fallopian tube shunt. However, this case was not included in our study because the article provided no data about the patient and the shunt. Selman et al. (5) also reported a case with traumatic acute subdural hematoma 48 hours after LP shunting with a distal slit valve shunt system. However, this case was also not included in the present study because of inadequate data given in the article. We found 16 cases with adequate data in the literature (6-14). Age, gender, primary indication for LP shunting, LP shunt type and pressure, time span between LP shunting and development of intracranial bleeding, type of bleeding, presence or absence of head trauma causing intracranial bleeding, treatment modalities, and outcome of the 17 patients (including the 16 cases from the literature and our one) were recorded. Outcomes of the patients were evaluated according to the Glasgow Outcome Scale (GOS) (15); a GOS score of 1-3 was accepted as worse outcome and GOS score of 4-5 as good outcome. We also evaluated the factors affecting outcome of the patients.

Statistical Evaluations

Chi-square or Fisher's exact tests were used to compare nominal variables according to their subject numbers. F test was used to compare the distribution of the series and Student's t test was used to compare numerical variables according to the results of the F test. Results were accepted as significant if the p value was <0.05.

Results

All characteristics of the 17 cases are shown in Table 1. There were 10 females and 7 males at 59.7±15.1 (average± standard deviation-SD-, age range 40-88) years of age. The primary disease treated with LP shunting was hydrocephalus in 11 cases, benign intracranial hypertension (BIH) in 5, and development of pseudomeningocele after posterior fossa epidermoid cyst surgery in one.

Table 1: Characteristics of cases with LP shunting and intracranial bleeding occurring spontaneously or after mild trauma.

No	Study	Age/ Gender	Bleeding	Primary disease	Head injury	Time span	Neurological condition	Treatment	Shunt type	CSDH	Outcome
1	Aoki (6)	44/F	ASDH	Pseudo-meningocele	yes	23 days	Comatose	HE	MP valve	yes	GOS 3
2	Aoki (6)	59/E	ASDH	NPH	yes	22 months	Comatose	HE	MP valve	yes	GOS 5
3	Aoki (6)	59/F	ASDH	NPH	yes	50 months	Stupor	HE	MP valve	Scalp depression	GOS 5
4	Aoki (6)	79/F	ASDH into CSDH	NPH	yes	7 months	Comatose	HE	MP valve	yes	GOS 1 (pneumonia)
5	Ayvalik (7)	52/M	Cerebellar ICH	BIH	no	1 day	Comatose	HE/SL	Distal slit valve	no	GOS 1 (ICH)
6	Barash (8)	42/M	ASDH into CSDH	H (Cryptococcal meningitis)	no	2 weeks	Alert	SL	Valveless	yes	GOS 1 (HIV)
7	Kamiryo (9)	77/F	ASDH	NPH	yes	10 months	Alert	SL	MP valve	yes	GOS 3
8	Kamiryo (9)	81/M	ASDH	NPH	yes	7 years	Alert	SL	MP valve	no	GOS 4
9	Kamiryo (9)	58/M	ASDH	NPH	yes	11 months	Comatose	HE/SL	MP valve	yes	GOS 2
10	Kamiryo (9)	66/F	ASDH	NPH	yes	1 month	Stupor	HE/SL	MP valve	no	GOS 1 (intractable seizure)
11	Turkoglu (10)	44/F	Parietal ICH	BIH	no	A few hrs	Comatose	Conservative	Distal slit valve	no	GOS 1 (pneumonia)
12	Suri (11)	42/F	SAH, frontal ICH	BIH	no	40 hrs	Comatose	HE/SL	Distal slit valve	no	GOS 5
13	Castillo (12)	40/F	ICH, SAH	BIH	no	In a few hrs	NA	Conservative	NA	no	NA
14	Hoya (13)	88/F	ASDH	NPH	yes	NA	Stupor	HE first, then SL	PV/LP (level 0,5)	NA	GOS 3
15	Hoya (13)	64/M	ASDH	NPH	yes	NA	Stupor	SL first, then HE (chronic)	PV/ LP (level 1)	NA	GOS 3
16	Er (14)	67/M	Cerebellar ICH	NPH	no	4 days	Alert	Conservative	PV/MP	no	GOS 5
17	Our patient	53/F	ASDH	BIH	no	3 years	Alert	SL first, then HE (chronic)	Valveless	no	GOS 5

ASDH: Acute subdural hematoma, BIH: Benign intracranial hypertension, CSDH: Chronic subdural hematoma, GOS: Glasgow Outcome Scale, H: Hydrocephalus, HE: Hematoma evacuation, ICH: Intracerebral hemorrhage, LP: Low pressure, MP: medium pressure, NA: Not available, NPH: Normal pressure hydrocephalus, PV: Programmable valve, SL: Shunt ligation.

Intracranial bleeding developed spontaneously in 7 of these cases including our one. In 12 of the cases, the bleeding occurred into the subdural space and in 5 into the other compartments: cerebellar hematoma in 2 cases, subarachnoid hemorrhage (SAH) accompanying intracerebral hematoma (ICH) in 2 cases, and ICH in one case. Most of the cases with ASDH had a history of mild trauma. There were only two cases with ASDH developing spontaneously including our patient. However, there was no history of trauma in any of the 5 cases with bleeding into the other compartments (p=0.0033).

The time span from LP shunting to bleeding was not mentioned for two cases in one article (13), while it was 16.2±25.9 months (average±SD, ranging from a few hours to 7 years) in the others.

In most cases (8 cases), a nonprogrammable medium pressure (MP) valve was used. In the cases with spontaneously developed bleeding, the LP shunt system was valveless in 2 cases, with distal slit valve in 4 cases, and with programmable valve in one case.

Treatment Modalities

After development of bleeding, only hematoma evacuation was performed in 5 cases, all of whom had ASDH. In one of them, LP shunt ligation was required after a few days because of rebleeding (13). One of these patients was died, and the GOS scores of the others were 3 in two cases and 5 in other two.

Lumboperitoneal shunt ligation or removal was performed in 5 cases. One of them died, and GOS scores of the others were 3 in two cases, 4 in one and 5 in the last one. In 2 of them, including our patient, a chronic subdural hematoma developed at the site of ASDH, and it was evacuated a few days after shunt ligation/removal.

Hematoma evacuation and shunt ligation/removal were performed in the same session in 4 cases. Two of them died. The GOS score was 2 in one case and 5 in the other.

In 2 cases, conservative treatment was applied without any intervention (10,14). One of them died and the GOS score of the other was 5.

Factors Affecting Outcome

Out of 17 cases, 5 cases died (31.2%). The GOS score was 2 in one, 3 in 4, 4 in one and 5 in 5 cases. The outcome of one patient (12) was not mentioned in the article. In total, 10 of the other 16 patients (62.5%) had worse outcome (GOS 1-3).

Age and sex were not statistically significant when comparing patients with worse (GOS 1-3) or good outcome (GOS 4-5) (p=0.87 and p=1, respectively), nor between the dying and surviving patients (p=0.44 and p=1, respectively). Presence or absence of trauma history and bleeding compartment also were not statistically significant comparing patients with worse and good outcome nor between the dying and surviving patients (p=0.6 and p=0.29, respectively, for presence/absence of trauma history, and p=0.6 and p=0.54, respectively, for bleeding compartment). Type of the primary disease treated with LP shunting was also not significantly different in the patients with worse and good outcome and between the dying and surviving patients (p=0.6 and p=0.54, respectively). Seven patients were comatose after bleeding and 3 of them died; however, 2 of 9 patients who were not comatose also died (p=0.59).

On the other hand, the time span from LP shunting to intracranial bleeding was statistically shorter in the cases with worse outcomes than in the cases with good outcomes (3.7±4.7 months and 38.6±28.1 months, respectively, p=0.0283). A similar result was also found between the time spans of the dying and surviving patients (1.7±2.9 months versus 28.2±27.3 months, respectively, p=0.0198).

Discussion

Although LP shunting is a simple operation and provides a lower complication rate than VP shunting, it may cause serious and life-devastating complications. Intracranial hypotension due to overdrainage of CSF is very frequently seen in cases with LP shunting. Friedman and Jacobson (3) reported a rate of up to 25% in a review evaluating cases treated with LP shunting for idiopathic intracranial hypertension, and Wang et al. (2) reported 14.8% in a series consisting of 74 cases treated for various reasons. In most cases with intracranial hypotension, symptoms and signs are mild, such as headache with changing position, nausea and vomiting, dizziness, or tinnitus (16). However, in some cases more serious conditions may be seen due to intracranial hypotension, the most feared ones being development of intracranial bleeding and acquired tonsillar descent (7).

Development of intracranial hypotension and chronic subdural effusions in patients treated with LP shunting is actually quite frequent, as it is in cases treated with VP shunt. Duthel et al. (17) reported development of chronic subdural effusion in 8 out of 195 cases with LP shunt. However, intracranial acute bleeding into the various compartments, spontaneously or after mild head injury, is rarely reported in cases with LP shunt. Selman et al. (5) reported only one case in a series consisting of 130 patients treated with LP shunting. We found 16 such cases with adequate data in the literature, which made a total of 17 cases with our patient. Various types of shunt systems had been used in these patients, such as valveless T tube, valves with constant medium pressure, distal slit valves, or programmable valves. Most of the cases had ASDH developing after mild head injury.

Unfortunately, the outcome of these patients was not very good, with 62.5% of them having GOS scores from 1-3. We could not find any factor significantly affecting the outcome of the patient except the time span from LP shunting to intracranial bleeding. Interestingly, the time span was significantly shorter in patients with worse outcome than in patients with good outcome (3.7±4.7 months and 38.6±28.1 months, respectively). This finding was thought to indicate that intracranial hypotension secondary to LP shunting in these patients developed quickly and severely and resulted in more massive bleeding. However, this finding gave us an an important clue for following patients with LP shunting. We recommended that these patients must be very closely followed for symptoms and signs of intracranial hypotension, especially during the first few months after shunting. Warning signs and symptoms of intracranial hypotension such as postural headache, tinnitus, nausea, and vomiting, etc. must be inquired about during outpatient control examinations.

Other factors including age, gender, type of primary disease treated with LP shunting, presence or absence of head injury, neurological condition after bleeding, and bleeding compartment did not significantly affect the outcome of the patients in this small series. It was expected that a valveless shunt system or one with smaller pressure might more frequently cause intracranial hypotension and bleeding, but unfortunately we could not show such an effect of LP shunt type, probably because of the small subject number.

In conclusion, although LP shunting is an easy and smart technique to treat patients with communicating hydrocephalus

or benign intracranial hypertension, it must be kept in mind that this procedure may cause intracranial bleeding, occurring spontaneously or after mild head injury, due to overdrainage of the CSF. Because mortality and morbidity are quite high in this condition, patients with LP shunting should be very closely followed for warning signs and symptoms of intracranial hypotension, especially during the first a few months.

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Contribution Categories	Name of Author
Development of study idea	F.K.G., O.A., A.T., B.E., M.S.V., I.G., E.D., A.T2., E.O.
Methodological design of the study	F.K.G., O.A., A.T., B.E., M.S.V., I.G., E.D., A.T2., E.O.
Data acquisition and processing	F.K.G., O.A., A.T., B.E., M.S.V., I.G., E.D., A.T2., E.O.
Data analysis and interpretation	F.K.G., O.A., A.T., B.E., M.S.V., I.G., E.D., A.T2., E.O., M.K.
Literature review	F.K.G., O.A., A.T., B.E., M.S.V., I.G., E.D., A.T2., E.O., M.K.
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Manuscript review and revision	F.K.G., O.A., A.T., B.E., M.S.V., I.G., E.D., A.T2., E.O.

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