

ALTERNATIVE SURGICAL THERAPY TO STANDARD TREATMENT FOR HYDROCEPHALUS: VENTRICULO-EPIPLOOIC SHUNT AND VENTRICULOPORTAL EXTRAPERITONEAL TRANSOMPHALIC SHUNT

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Accepted August 2, 2016

Introduction: Hydrocephalus is an extremely invalidating disease, with high impact on population, requiring frequent medical care.

Material and methods: Classical ventriculoperitoneal and ventriculocardiac shunts have high risk of complications. We propose two new surgical techniques, ventriculo-epiploic and ventriculoportal extraperitoneal transomphalic shunts for treatment of hydrocephalus. First, we performed ventriculo-epiploic and ventriculoportal extraperitoneal transomphalic shunt, on experimental animals (pigs), according with the laws in force. Latter, we performed ventriculo-epiploic shunt on humans, and we retrospectively reviewed medical records of patients.

Results: Experimental animals in both studies had favorable outcome, with no postoperative early or late morbidity. Between 2008 and 2012 we performed ventriculo-epiploic shunt in 12 patients. The outcome was favorable in all cases, without significant postoperative complications.

Conclusions: Ventriculo-epiploic and ventriculoportal extraperitoneal transomphalic shunts are two new, safe and efficient surgical techniques for treatment of hydrocephalus. They can be performed as a saving solution in patients with repetitive classical shunt failures. Complications specific to classical shunt systems can be successfully avoided. Ventriculo-epiploic and ventriculoportal shunts have multiple advantages compare to other classic shunt techniques, with no additional stipulated complications. In case of ventriculoportal shunt further research is needed and this surgical technique must be performed on human subjects with hydrocephalus.

Key words: epiploon, hydrocephalus, umbilical vein, ventriculo-epiploic shunt, ventriculoportal shunt.

INTRODUCTION

Hydrocephalus is impairment in production, flow, or absorption of cerebrospinal fluid (CSF).^{1,2} Hydrocephalus is an extremely invalidating disease with high impact on population, requiring frequent medical care. The prevalence of hydrocephalus is 1–1.5%.^{3,4} Hydrocephalus can be congenital, secondary or normal pressure hydrocephalus.³

Surgery is the treatment of choice in hydrocephalus. Over the times many surgical procedures were tried. They can be grouped into surgical techniques of internal drainage (third ventriculostomy), external drainage (external ventricular drainage) and extracranial drainages (ventriculoperitoneal shunt, ventriculoatrial shunt,

lumboperitoneal shunt, ventriculopleural shunt, Torkildsen shunt, ventriculosubgaleal shunt, ventriculosinusal shunts (ventriculosagittal or ventriculotransverse), ventriculocholecystic shunt, ventriculoureteral shunt, lumboureteral shunt, ventriculomastoid drainage, ventriculosternal shunt, drainage into the thoracic duct, salivary gland, spinal epidural space, bone stomach, ileum and fallopian tube, etc.).

Ventriculoperitoneal and ventriculocardiac shunts are the most common surgical techniques used for treatment of hydrocephalus, but they carry high risk of complications.⁵

Patients with ventriculoperitoneal shunts can develop specific complications such as: inguinal hernia, CSF pseudocysts, CSF ascites, visceral perforations and ileus.⁵ Complications specific to ventriculocardiac shunts, such as sepsis or shunt

nephritis, carries an extremely high mortality and morbidity.⁵⁻⁷

The aim of this study is finding new surgical techniques for hydrocephalus.

MATERIAL AND METHODS

We propose two new surgical techniques, ventriculo-epiploic and ventriculoportal extraperitoneal transomphalic shunts for treatment of hydrocephalus. First, we performed the two surgical techniques, ventriculo-epiploic and ventriculoportal extraperitoneal transomphalic shunt, on experimental animals with highly similar anatomy (pigs). Latter, we performed ventriculo-epiploic shunt on humans, and we retrospectively reviewed medical records of these patients.

First study: We performed a new surgical technique, ventriculo-epiploic shunt, as an experimental study on three animals. We chose three pigs because they have highly similar anatomy with humans. The experiment was done into the Center of Experimental Medicine, University of Medicine and Pharmacy Iuliu Hatieganu, Cluj-Napoca, according to the laws in force. Surgery for the cranial step was standard. We inserted the proximal catheter into the right lateral ventricle, using a right posterior parietal burr hole and proximal catheter was connected with the distal one using a valve. Then, on each pig we performed one of the three surgical techniques of ventriculo-epiploic shunts: distal end of the abdominal catheter was placed between the two layers of the great omentum through open surgery, distal end of the abdominal catheter was placed into a large gastroepiploic vein through open surgery and distal end of the abdominal catheter was placed

between the two layers of the great omentum laparoscopically.

Second study: We performed another new surgical technique, ventriculoportal extraperitoneal transomphalic shunt on an experimental animal. From the same reasons, because it has similar anatomy with humans we chose a pig, for this experiment. The experiment was performed into the Center of Experimental Medicine, University of Medicine and Pharmacy Iuliu Hatieganu, Cluj-Napoca, according to the laws in force. Surgery for the cranial step was also standard, as described in the first experiment. The key to ventriculoportal extraperitoneal transomphalic shunt is inserting the distal end of the catheter into the reopened umbilical vein and draining CSF into the portal vein. The round ligament of the liver was found near the umbilicus and was dissected free extraperitoneally for 4 cm length. The ligament was opened and the umbilical vein was found inside it and circumdissected free for 3 cm. Following circumdissection, the vein was suspended using a thread. After lifting the vein, an oblique hemisection in the walls of the vein was done. The umbilical vein was progressively reopened with a stylet, aiming toward the right shoulder. The distal end of the catheter was inserted into the umbilical vein and CSF was drained into the portal system.

Third study: We performed a retrospective review of medical records of consecutive patients with hydrocephalus, in which ventriculo-epiploic shunt was done between February 2008 and July 2012.

RESULTS

First and second study: Experimental animals in both studies had favorable outcome, with no postoperative early or late morbidity.

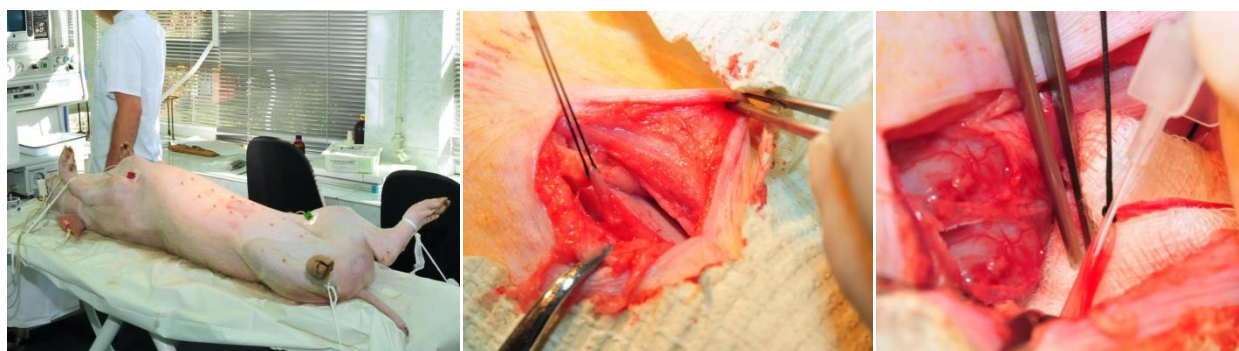


Fig. 1. Ventriculoportal extraperitoneal transomphalic shunt. a) Positioning of the animal, under general anesthesia, on the experimental table. b) Extraperitoneal circumdissection of the umbilical vein. c) Testing of umbilical vein patency after complete reopening.

Third study: Between February 2008 and July 2012 we performed ventriculo-epiploic shunt in 12 patients. There were 6 men and 6 women. Age varied from 6 to 70 years. Four patients had congenital hydrocephalus, 6 patients had secondary hydrocephalus and 2 patients had normal pressure hydrocephalus.

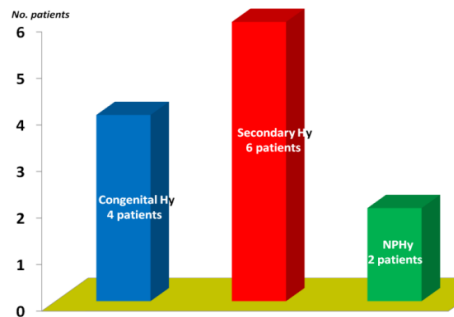


Fig. 2. Types of hydrocephalus in patients with ventriculo-epiploic shunt.

The technique was performed in 10 patients with multiple classical shunt revisions and in two was done per primam. Except for the 2 patients in which ventriculo-epiploic shunt was done as first choice, all have prior history of repeated distal complications.

The outcome was favorable in all cases, with no significant postoperative complications. Following ventriculo-epiploic shunt we have only 2 cases with shunt failure and in both cases it was proximal, caused by ventricular catheter or valve occlusion with debris. No distal shunt failure was encountered in our series of cases. The outcome was favorable in all cases, with no significant postoperative complications. No additional neurological deficits were seen. The follow-up period varied from 36 to 75 months.

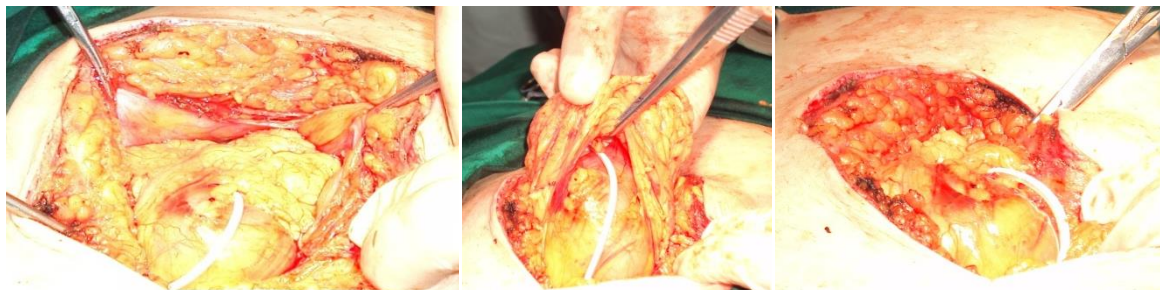
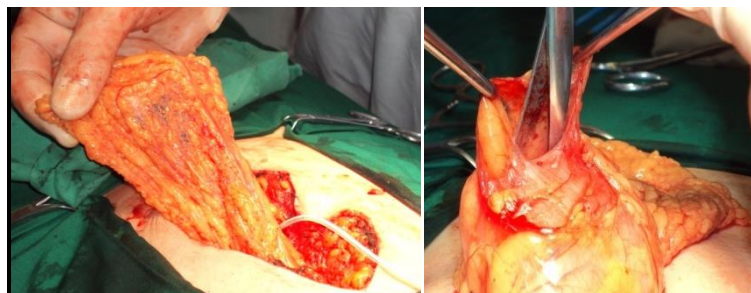


Fig. 3. Ventriculo-epiploic shunt, intraoperative aspects. a) Insertion of the proximal catheter into the right lateral ventricle. b) Great omentum. c. Dissection of the two omental layers. d, e, f) Introducing the distal tip of the peritoneal catheter into the two layers of the great omentum.

Table 1

Patients with ventriculo-epiploic shunt for hydrocephalus

No	Sex	Age*	Diagnosis	BEFORE VENTRICULO-EPIPLOIC SHUNT						AFTER VENTRICULO-EPIPLOIC SHUNT				
				Initial operations	Clinical and neurological findings	No of revisions‡	Shunt failure		Cause of distal shunt failure	Age†	No of revisions§	Cause of failure	Outcome	Follow-up
							Proximal + valve	Distal						
1	m	7 mo	Congenital internal hydrocephalus, right T porencephalic cyst, left hemiparesis	Cysto-ventriculoperitoneal shunt in Y	Increased head circumference, altered mental state, vomiting, left hemiparesis	23	9	14	CSF pseudocysts Extensive peritoneal adhesions syndrome	6 y	0	-	good	6 y 3 mo
2	f	42 y	Secondary hydrocephalus after operated colloid cyst. Hysterectomy.	Third ventriculostomy	Headache, nausea, vomiting	0	-	-	-	45 y	0	-	good	6 y 1 mo
3	m	5 mo	Congenital hydrocephalus, left porencephalic cyst, right hemiparesis	Cysto-ventriculoperitoneal shunt in Y	Increased head circumference, altered mental state, vomiting, right hemiparesis	28	14	14	CSF pseudocysts Extensive peritoneal adhesions syndrome	7 y	0	-	good	5 y 10 mo
4	f	8 y	Secondary hydrocephalus after operated right cerebellar pilocytic astrocytoma	Ventriculoperitoneal shunt, total resection of the right cerebellar pilocytic astrocytoma	Headache, vomiting, right-side balance and coordination disturbances	7	1	6	CSF pseudocysts Extensive peritoneal adherence syndrome	11 y	0	-	good	5 y 6 mo
5	f	50 y	Secondary hydrocephalus after nonaneurysmal subarachnoid hemorrhage. Cholecystectomy. Appendectomy	Third ventriculostomy	Headache, nausea, vomiting	0	-	-	-	51 y	0	-	good	5 y 1 mo
6	f	44 y	Secondary hydrocephalus after ruptured basilar tip aneurysm	Ventriculoperitoneal shunt, coils embolization of the basilar tip aneurysm	Headache, nausea, vomiting, meningismus	1	-	1	Distal shunt occlusion with debris	46 y	0	-	good	4 y 9 mo
7	m	6 mo	Congenital hydrocephalus, pineal cyst, Dandy-Walker malformation, agenesis of corpus callosum	Ventriculoperitoneal shunt	Increased head circumference, altered mental state, vomiting,	21	12	9	CSF pseudocysts Extensive peritoneal adhesions syndrome	6 y	1	Ventricular catheter and valve occlusion with debris	good	3 y 11 mo

Table 1
(continued)

8	m	70 y	Normal pressure hydrocephalus	Ventriculoperitoneal shunt	Gait disturbances, memory loss, gatism	1	-	1	Distal shunt occlusion with debrides	70 y	0	-	good	3 y 10 mo
9	m	56 y	Secondary hydrocephalus after operated left vestibular schwannoma, left hipoacusia	Ventriculoperitoneal shunt, subtotal resection of the left vestibular schwannoma	Headache, vomiting, left hipoacusia	2	0	2	Extensive peritoneal adherence syndrome	58 y	0	-	good	3 y 8 mo
10	f	32 y	Secondary hydrocephalus after meningitis	Ventriculoperitoneal shunt	Headache, altered mental state, meningismus	4	2	2	CSF pseudocysts	34 y	0	-	good	3 y 5 mo
11	m	67 y	Normal pressure hydrocephalus	Ventriculoperitoneal shunt	Gait disturbances, memory loss, gatism	2	1	1	Distal shunt occlusion with debrides	68 y	0	-	good	3 y 4 mo
12	f	8 mo	Congenital internal hydrocephalus, posterior fossa arachnoid cyst, pineal region cystic tumor, agenesis of the corpus callosum, ventriculitis with Acinetobacter, obesity, depressive syndrome	Ventriculoperitoneal shunt, posterior fossa cystotomy, left frontal Ommaya reservoir for pineal tumor	Altered mental state, somnolence, vomiting, Parinaud syndrome, bilateral III nerve palsy	38	23	16	CSF pseudocysts Extensive peritoneal adherences syndrome	21 y	1	Ventricular catheter occlusion with debrides		3 y

Age* age at diagnosis of hydrocephalus

Age† age at ventriculo-epiploic shunt

No of revisions‡ numbers of shunt revision before ventriculo-epiploic shunt

No of revisions§ number of revisions after ventriculo-epiploic shunt

DISCUSSIONS

In ventriculo-epiploic shunt the distal tip of the peritoneal catheter is introduced between the two omental layers or intravascular into an epiploic vein.⁸ Epiploon has a very good absorption capacity, leaving only a small amount of CSF to flow into the peritoneal cavity.

The distal tip is isolated from abdominal viscera and complications specific to ventriculoperitoneal shunt can be prevented. CSF is irritating and causes local inflammatory response, peritoneal congestion and inflammatory adhesions between catheter and abdominal viscera, leading, in some cases to a extensive peritoneal adherence syndrome.⁷ Placing the distal catheter between the great omental layers avoids direct contact of catheter and bowel and adhesions occurrence, preventing ileus, bowel volvulation and visceral perforations.

The incidence of shunt infections is also diminished because the great omentum has lymph nodes which limit infection spreading. Liver is the second station in stopping infection spreading.

The etiology of CSF pseudocysts is not clear, but is supposed to be the consequence of shunt infections, extensive abdominal adhesions, history of prior abdominal surgery, hyperproteinorrachia, or impaired absorption of the peritoneum.^{9,10} After isolating the distal end between the two layers of the great omentum, the irritating CSF is not drained any more into the peritoneal cavity, preventing adherence and pseudocyst formation. By limiting the amount of CSF which reaches the abdominal cavity, processus vaginalis can close and the rate of inguinal hernia, in infants, toddlers or young children^{5,11} is lowered. Through the same mechanism, limiting the quantity of CSF in the peritoneal cavity, the incidence of CSF ascites¹² is diminished.

In refractory CSF ascites intravascular ventriculo-epiploic shunt is recommended. By placing the distal catheter into an epiploic vein, the entire quantity of CSF flows into the portal system, being a good therapeutic option in refractory ascites.

Ventriculo-epiploic shunt can represent a saving option in patients with ventriculoperitoneal shunt with multiple distal shunt failures due to repetitive abdominal complications, or it can be done in patients with high risk of developing abdominal complications. Ventriculo-epiploic shunt allows keeping the abdominal cavity for distal shunting, in patients in which classic techniques carry high morbidity.

The umbilical vein was catheterized by Carbalhaes¹³, Bayly and Carbalhaes¹⁴ and Roberti¹⁵ for exploration of the portal system and treatment. In Romania Burlui et al. described the procedure of umbilical vein recanalization in 1966.¹⁶⁻¹⁸

During the embryonic and fetal life the umbilical vein plays an important role in carrying oxygenated blood from placenta to the fetus. Following birth the umbilical vein undergoes degeneration and closes.¹⁹ The umbilical vein can become patent spontaneously in patients with portal hypertension²⁰ or it can be reopened surgically. Reopening of the umbilical vein can be done intra or extraperitoneally.¹⁷ The round ligament, containing the umbilical vein has a distal juxtaumbilical segment, which is entirely extraperitoneal.¹⁷ The extraperitoneal approach is preferred, because the peritoneal cavity is not opened, and the risk of accidental intraoperative bowel perforation is completely eliminated.

In normal conditions CSF is absorbed into dural venous sinuses, so shunting the CSF into the venous flow is physiologic. Pressure in the portal system is 5–10 mm Hg²¹, lower than in hydrocephalus, so CSF flow is not hindered. We do not recommend ventriculoperitoneal shunt in patients with portal hypertension, even if the pressure gradient still exists, because extra load of fluid may aggravate the liver disease. Patients with cirrhosis or liver failure are also contraindicated from the same reasons. A total amount of 2000 ml fluid per day can be administrated into the portal flow. The daily quantity of CSF is only 500 ml, so this amount of fluid is well tolerated.

But most important, liver has an immunological function. Liver plays an important role in controlling innate and acquired immunity and ensures clearance of antigen-antibody immune complexes.^{22,23} Due to immunological function of the liver antigens and immune complexes passing through the liver are stopped and do not reach systemic circulation. Complications specific to ventriculoatrial shunts, such as sepsis, shunt nephritis or tumor cells spreading are eliminated. None of the complications related to placement of the distal catheter in the right atrium, such as extrasystoles, arrhythmias, atrial wall perforations, cardiac tamponade, tricuspid valve insufficiency, tricuspid stenosis, endocarditis, pulmonary thromboembolism, pulmonary hypertension, cor pulmonale and right heart failure, can occur.^{5,7,24-27}

Also because of the immunological function of the liver, long-term antibiotic treatment, mandatory

in ventriculoatrial shunts, can be discontinued earlier without increasing morbidity.

The distal end of the catheter inserted into the umbilical vein is isolated from abdominal viscera and complications specific to ventriculoperitoneal shunts, such as volvulus around the catheter or visceral perforations, are avoided. Other specific complications, such as peritoneal adhesions or CSF pseudocysts, are also excluded because the irritative CSF does not reach the peritoneal cavity.

The ventriculoportal shunt combines the advantages and avoids complications of ventriculoperitoneal and ventriculocardiac shunts.

Ventriculoportal shunt is indicated as the first choice treatment or it can be done in patients with multiple complications following classical shunt procedures. Patients, prior contraindicated for ventriculoperitoneal shunt, such as those with history of repeated extensive lower abdominal surgery, extensive peritoneal adhesions, necrotizing enterocolitis and peritonitis and refractory CSF ascites, may benefit following ventriculoportal shunt, so indications for surgery are extended. As in any vascular shunt, 3 consecutive sterile CSF cultures are needed.

Besides general contraindication for surgery in patients with hydrocephalus, ventriculoportal shunt must not be performed in CSF infections, portal hypertension, cirrhosis and liver failure. History of extensive upper abdominal surgery is not a contraindication per se, but in these patients the round ligament may have been cut during operation and catheterization of the umbilical vein may not be possible.

CONCLUSIONS

Ventriculo-epiploic and ventriculoportal extraperitoneal transomphalic shunts are two new, safe and efficient surgical techniques for treatment of hydrocephalus. They can be performed as a saving solution in patients with repetitive classical shunt failures. Complications specific to classical shunt systems can be successfully avoided. Ventriculo-epiploic and ventriculoportal shunts have multiple advantages compare to other classic shunt techniques, with no additional stipulated complications. In case of ventriculoportal shunt further research is needed and this surgical technique must be performed on human subjects with hydrocephalus.

Authors' note: Both authors had equal contribution to the article, and they both are principal authors. VTG participated in conception and design of the study, performed surgery, acquired data, analyzed and interpreted data, wrote and reviewed the manuscript and gave final approval of the version to be published; AMS participated in conception and design of the study, participated in surgery, acquired data, analyzed and interpreted data, wrote, translated and reviewed the manuscript and gave final approval of the version to be published

Funding: The authors received no financial support for the research, authorship, and/or publication of this article.

Conflict of interests: The authors declare that there is no conflict of interests with respect to the research, authorship, and/or publication of this article.

Funding: None

ACKNOWLEDGEMENT

This manuscript was presented at "41st World Congress of ISMH", 18-21 May 2016, Bucharest, Romania, as invited paper.

REFERENCES

1. Rekate H.L., *A contemporary definition and classification of hydrocephalus*, *Semin Pediatr Neurol*, **2009**, 16, 9-15.
2. Rekate H.L., *A consensus on the classification of hydrocephalus: its utility in the assessment of abnormalities of cerebrospinal fluid dynamics*, *Childs Nerv Syst*, **2011**, 27, 1534-1541.
3. Greenberg M.S., "Hydrocephalus". In *Handbook of neurosurgery*. Thieme Medical Publisher, New York, 2006, 180-207.
4. Hord E.D., *Hydrocephalus*, *eMedicine*, **2004**, eMedicine Specialties/Neurology/Behavioral Neurology and Dementia.
5. Tamburrini G.; Caldarelli M.; Di Rocco C., *Diagnosis and management of shunt complications in the treatment of childhood hydrocephalus*, *Reviews in Neurosurgery*, **2002**, 1, 3.
6. Fried A.H.; Epstein M.H., *Childhood hydrocephalus: clinical features, treatment, and the slit-ventricle syndrome*, *Clinical Trials and Noteworthy treatments for brain tumors treatment of hydrocephalus: shunts*, **2005**.
7. Popescu M.; Grigorean V.T., "Hidrocefalia". Editura Universitară Carol Davila, București, 2006.
8. Grigorean V.T.; Popescu M.; Sandu A.M.; Toader S., *Ventriculo-epiploic shunt, a new surgical technique for treatment of hydrocephalus*, *J Exp Med Surg Res*, **2010**, 17, 55-63.
9. Gaskill S.J.; Marlin A.E., *Pseudocysts of the abdomen associated with ventriculoperitoneal shunts: a report of twelve cases and a review of the literature*, *Pediatr Neurol*, **1989**, 15, 23-27.
10. Adegbite A.B.; Khan M., *Role of protein content in CSF ascites following ventriculoperitoneal shunting*, *J Neurosurg*, **1982**, 57, 423-425.
11. Clarnette T.D.; Lam S.K.; Hutson J.M., *Ventriculoperitoneal shunts in children reveal the natural history of closure of the processus vaginalis*, *J Pediatr Surg*, **1988**, 33, 413-416.

12. Chidambaram B.; Balasubramaniam V., *CSF ascites: a rare complication of ventriculoperitoneal shunt surgery*, *Neurology India*, **2000**, 48, 378-380.
13. Carbalhaes G.O., *Portography: a preliminary report of a new technique via the umbilical vein*, *Clin Proc Child Hosp Dist Columbia*, **1959**, 15, 120-122.
14. Bayly J.H.; Carbalhaes G.O., *The umbilical vein in the adult: diagnosis, treatment and research*, *Am Surg*, **1964**, 30, 56-60.
15. Roberti G.; D'Agnolo B.; Servello M.; Bottero M.; Matteucci M., *Omphaloportography: a new means of investigation of the portal system*, *Minerva Med*, **1956**, 56, 334-336.
16. Burlui D.; Teju G., *Repermeabilization of the umbilical vein, exploration route and per- and postoperative treatment*, *Presse Med*, **1966**, 74, 179-180.
17. Burlui D.; Rațiu O., „Vena ombilicală în chirurgia porto-hepato-biliară”, Editura Medicală, București, 1970.
18. Burlui D.; Rațiu O.; Manesco G.; Teju G., *Portal arterialization using the repermeabilized umbilical vein*, *Presse Med*, **1986**, 76, 581-582.
19. Butler H., *Post-natal changes in the intra-abdominal umbilical vein*, *Arch Dis Child*, **1954**, 92, 427-435.
20. Young T.H.; Lee H.S., *Recanalized umbilical vein*, *N Engl J Med*, **2007**, e17.
21. Carale J.; Mukherjee S., *Portal hypertension*, *Medscape Medscape Reference*, **2010**.
22. Mehal W.Z.; Azzaroli F.; Crispe I.N., *Immunology of the healthy liver: old questions and new insights*, *Gastroenterology*, **2001**, 120, 250-260.
23. Gao B.; Jeong W.I.; Tian Z., *Liver: an organ with predominant innate immunity*, *Hepatology*, **2008**, 47, 729-736.
24. Vargas-Barron J.; Buenfil-Medina C.; Sanchez-Ugarte T.; Keirns C.; Rocha-Maguey J.; Romero-Cardenas A.; Lupi-Herrera E., *Ventriculoatrial shunts for hydrocephalus and cardiac valvulopathy: an echocardiographic evaluation*, *Am Heart J*, **1991**, 121, 1498-1501.
25. Piatt J.H. Jr.; Hoffman H.J., *Cor pulmonale: a lethal complication of ventriculoatrial CSF diversion*, *Childs Nerv Syst*, **1989**, 5, 29-31.
26. Berhouma M.; Vallee B., *Pulmonary hypertension and migrated ventriculoatrial shunt*, *Rev Prat*, **2011**, 61, 602.
27. Kluge S.; Baumann H.J.; Regelsberger J.; Kehler U.; Gliemroth J.; Koziej B.; Klose H.; Meyer A., *Pulmonary hypertension after ventriculoatrial shunt implantation*, *J Neurosurg*, **2010**, 113, 1279-1283.