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Over-drainage and persistent shunt-dependency in patients with idiopathic intracranial hypertension treated with shunts and bariatric surgery

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Abstract

Background: Idiopathic intracranial hypertension (IIH) may lead to visual impairment. Shunt surgery is indicated for refractory IIH-related symptoms that persist despite medical treatment, or those presenting with significant visual decline. Obesity is a risk factor for IIH; a reduction in weight has been shown to improve papilledema. Bariatric surgery (BS) has been suggested for treating IIH associated with morbid obesity. In this study, we describe a high rate of over-drainage (OD) seen in patients following shunts and BS.

Methods: The study cohort includes 13 patients with IIH that underwent shunt surgery for treatment of the IIH-related symptoms. Six patients underwent BS in addition to the shunt surgery (but not concomitantly). Seven patients had only shunt surgeries with no BS. Data were collected retrospectively.

Results: BS effectively led to weight reduction (body mass index decreasing from 43 ± 4 to 28 ± 5). Patients undergoing BS had 1–6 (2.5 ± 1.9) shunt revisions for OD following BS, as opposed to 0–3 (1.4 ± 1.1) revisions prior to BS over similar time spans (statistically insignificant difference), and 0–6 (1.6 ± 2.5) revisions among the non-BS patients over a longer time span (statistically insignificant difference). Two patients in the BS group underwent shunt externalization and closure; however, they proved to be shunt-dependent.

Conclusions: Patients with IIH that undergo shunt surgery and BS (not concomitantly) may suffer from OD symptoms, necessitating multiple shunt revisions, and valve upgrades. Despite BS being a valid primary treatment for some patients with IIH, among shunted patients, BS may not lead to resolution of IIH-related symptoms and patients may remain shunt-dependent.

Key Words: Bariatric surgery, idiopathic intracranial hypertension, shunt

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INTRODUCTION

Idiopathic intracranial hypertension (IIH), also known as pseudotumor cerebri, is a disease of unknown origin, affecting primarily young women, often with morbid obesity. Major clinical presentations are headaches and transient visual obscurations. Treatment typically consists of medications (such as acetazolamide or Topamax) that decrease cerebrospinal fluid (CSF) production.

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Patients with refractory headaches and/or severe visual decline are often treated by various surgical options, including optic nerve sheath fenestration (ONSF) and CSF diversion procedures such as lumboperitoneal shunt (LPS) or ventriculoperitoneal shunt (VPS). Despite the proven positive effect of CSF diversion procedures on visual outcome, many patients continue to suffer from chronic headaches, and often undergo multiple shunt revisions (treating symptoms of over and under-drainage) or multiple episodes of intracranial pressure monitoring (ICPm) to try and correlate the symptoms to ICP.[25]

Recently, bariatric surgery (BS) has gained popularity, and has proven to be effective in weight reduction, as well as improvement of various obesity-related morbidities such as diabetes, ischemic heart disease, and general wellbeing.[1,6,23,26]

Previous reports among patients with IIH and obesity have shown that BS may lead to major symptom relief and visual improvement.[3,5,11,13,18,20,27-29] Despite these encouraging publications, there is a scarcity of information on the effect of BS on obese patients that had prior CSF diversion surgeries.[15]

This study focuses on patients diagnosed with IIH and treated with a shunt, who underwent BS at a later stage, or those treated primarily with BS, followed at a later stage with a shunt.

METHODS

Following an Institutional Review Board approval, data were collected retrospectively. Patient approval was waived. Collected data included: Demographics, surgical history (relating to ONSF, shunt surgeries, ICPm, and BS), and clinical course. Clinical data included degree of headache (none [minimal impact], existing [with no major impact on daily life], severe [with significant impact on daily life]), and data relating to patients' weight.

Ophthalmological variables included visual acuity (VA), optic disc appearance (ODA), and visual fields (VFs).

Between 2002 and June 2014, 14 patients, all females, underwent CSF diversion procedures (LPS or VPS) for the treatment of IIH. Of these, 7 have undergone both BS and shunt procedures. However, 1 of the patients underwent BS several years before diagnosing IIH, undergoing a gastric banding procedure 9 years prior to an LPS while had no symptoms of IIH. The banding sleeve in this patient was removed 1 year later due to abdominal complications. One patient from the non-BS group was lost to follow-up after the shunt surgery, and thus was not included in this study. Thus, for practical reasons, we divided the patients into the following categories: Those that underwent both BS and a shunt (6 patients), and those that did not undergo BS (7 patients, including the patient that underwent a band removal 9 years prior to the shunt surgery).

Shunt revisions were categorized as primary shunt insertions, revisions due to shunt infections, revisions treating under-drainage symptoms (originating from a restrictive valve), revisions treating over-drainage (OD) symptoms (originating from a permissive valve), and technical revisions (for treating disconnections, catheter migrations, and valve rotations).

Data were summarized in an excel file, and basic statistical analysis (mean ± standard deviation) was performed for the numerical data. Comparison of number of surgeries done in the various groups was done using an unpaired Student's *t*-test.

RESULTS

Bariatric surgery group

Of the 6 patients in the BS group, 5 had an LPS 3–70 months (43 ± 26) prior to the BS, and 1 had a VPS 2 years following BS. Ages at shunt surgeries were 21–31 years (27 ± 4). Two patients underwent ONSF prior to the shunt. Prior to BS, patients underwent an average of 3–11 shunt revisions (6.2 ± 3.3). The indication for BS was for weight reduction (and not control of IIH). The indication for shunt (in both the BS and non-BS groups) was the treatment of refractory IIH despite maximal medication treatment, accompanied with a visual decline that necessitated immediate pressure reduction.

Four patients underwent sleeve gastrectomy, and 2 underwent gastric banding (see an overview on BS techniques and efficacy by DeMaria, 2007).[10] At the time of the shunt surgeries, patients weighed 95–118 kg (109 ± 9), and had a body mass index (BMI) of 37–47 (43 ± 4). Following BS, patients lost 5–54 kg (37 ± 17) over a period of 10–84 months (39 ± 25), and the BMI reduced in all patients (from 43 ± 4 to 28 ± 5). Following BS, patients underwent 1–14 (5.7 ± 5) shunt related surgeries.

Nonbariatric surgery group

Of the 7 patients in the non-BS group, 6 underwent LPS and 1 VPS. Ages at primary surgery were 12–43 years (26 ± 10). Two patients underwent ONSF prior to the shunt surgery. At the time of shunt surgeries, patients weighed 60–120 kg (92 ± 23), and had a BMI of 26–43 (35 ± 7). At last follow-up, patients weighed 56–126 kg (88 ± 21), and had a BMI of 22–45 (33 ± 8).

Clinical course and shunt revisions

Follow-up duration after the first shunt procedure was 23–181 (86 ± 47) months for the entire group (39–116 (75 ± 25) in the BS group, and 23–181 (95 ± 61) in

the non-BS group). Follow-up period following the BS procedure was 10–84 (39 ± 25) months.

All the BS patients underwent multiple shunt revisions due to various indications (as stated in the methods section). The exact surgical course was available for 11 patients (the 6 BS patients, and 5 of 7 non-BS patients). Only 2 patients underwent 1 revision each for under-drainage. We focused on patients undergoing revisions for OD and technical reasons [Table 1]. Regarding OD indications, the BS group had 1–3 (1.4 ± 1.1) revisions prior to the BS, but 1–6 (2.5 ± 1.9) revisions for after BS. The non-BS group underwent 0–6 (1.6 ± 2.5) revisions for OD indications. The difference in number of revisions due to OD did not reach statistical significance between the various patient groups. Following OD, valves were upgraded, changed to dual switch valve (DSV), or added an assist device. Current shunt valves are summarized in [Table 2].

Regarding technical indications, prior to BS, patients underwent 0–5 (2.4 ± 1.8) revisions, as opposed to 0–5 (1.5 ± 1.9) following BS, and 0–4 (1.4 ± 1.9) revisions in the non-BS group.

Table 1: Number of shunt revisions

Patient	BS group				Non-BS group	
	Pre-BS		Post-BS			
	OD	Technical	OD	Technical	OD	Technical
1	1	2	6	2		
2	1	5	2	5		
3	2	0	1	1		
4	0	3	2	0		
5	3	2	1	0		
6			3	1		
7					6	3
8					1	0
9					0	4
10					0	0
11					1	0

BS: Bariatric surgery, OD: Over-drainage

Table 2: Current valve status

	BS group	Non-BS group
DSV	1	3
DSV + SA	2	1
Medos	2	1
MP + SA	1	-
Medos + SA	1	-
Unknown	-	2

DSV: Dual switch valve (Miethke), SA: Shunt assist (Miethke), MP: Medium pressure (Medtronic), HP: High pressure (Medtronic), Medos: Codman Hakim Programmable Shunt (Johnson and Johnson), BS: Bariatric surgery

In 2 of the 5 patients that underwent BS following LPS, the LPS was externalized to evaluate the need for any CSF diversion. These patients lost 47 and 42 kg and had clear OD symptoms. Both patients had the shunt externalized and elevated gradually, and developed severe under-drainage symptoms over a course of hours, which improved once the height of the bag was lowered. Both patients improved with shunt revisions.

At last follow-up, 2 patients in the BS group, and 4 in the non-BS group still complain of headache.

Visual exams

Preshunting visual evaluation was available for 12 of 13 patients (24 eyes). VA was intact or only mildly compromised in 22 eyes preshunting and in 19 eyes postshunting. Overall, VA deteriorated in 3 eyes. VFs were intact or with minor defects in 17 eyes preshunting, and in 20 postshunting. Overall, VF deteriorated in 4 eyes and improved in 7. ODA was normal in only 1 eye before surgery but was normal in 15 following shunt surgery. Overall, the ODA improved in 12 eyes and deteriorated in 7.

When comparing the BS and the non-BS groups, both had similar improvement and deterioration numbers in both VA and VF. However, ODA normalized in 6 eyes in the BS as opposed to 4 in the non-BS group. Of the 6 patients with remaining headaches, 1 had no available preoperative visual evaluation. The remaining 5 had stable intact VA (8 eyes), deteriorated VA (2 eyes), stable VF (5 eyes), deteriorated VF (2 eyes), and improved VF (3 eyes). ODA was stable (1 eye), deteriorated (4 eyes), and improved (5 eyes) at last follow-up.

DISCUSSION

This is the largest report on the effect of BS on IIH patients that have a functioning shunt. The study points out two main issues:

- OD is common following shunt surgeries for IIH. However, OD is more common in patients with a shunt and BS
- While BS and the subsequent weight loss can potentially cure IIH in nonshunted patients; patients with a shunt already in place may remain shunt-dependent even after BS. Two shunted patients that underwent BS, developed OD, and were subsequently challenged for shunt dependency. Both proved to be shunt-dependent.

IIH is a poorly understood disease, which typically affects young, obese women. The association of IIH to overweight has been repeatedly described, although the mechanism is poorly understood. Between 64% and 70% of IIH patients are obese[3] High BMI is a risk factor for

IIH,[9] and a poor prognostic factor once IIH has been diagnosed.[3] Several mechanisms have been suggested to correlate between overweight and IIH, including mechanical reasons such as increased intra-abdominal and intrathoracic pressures leading to reduced intracranial venous drainage, and hormonal reasons such as elevated leptin and estrogen.[2,5,9] Other contributing factors may include hypoventilation and sleep apnea associated with obesity, leading to hypercarbia and elevated ICP,[3,5] or occult cerebral sinus thrombosis affecting cerebral venous drainage.[3]

Treatment of IIH includes diet and medications such as acetazolamide and Topamax. However, in refractory IIH causing incapacitating headaches or continuous visual decline, surgical alternatives are applied, such as ONSF and CSF diversion surgeries.[17] Currently, there are no clear indications for surgical treatment or technique; decisions are based on each center's experience.[4] In general, it is accepted that following both ONSF and CSF diversions, headache remains a common complaint over the years in about 40–60% of patients.[14,17,19,21,25,30] In additional, both LPS and VPS have a high rate of malfunction, necessitating shunt revisions, mostly due to OD or blockage.[21,25,30,31,33] Many valves have been utilized to treat and prevent OD; our personal experience has been to place horizontal-vertical valves (such as the DSV by Miethke), sometimes coupled with a gravitational component (shunt assist [SA], by Miethke).[32] DSV includes two valves that toggle between them depending on the patient's position. At the upright position, the higher pressure valve functions while at the horizontal position, the lower pressure valve functions. This mechanism reduces OD occurrence compared with single valve systems.[32]

Weight loss has shown positive effects on IIH-related symptoms, especially when meaningful weight reduction is achieved.[3,16] BS in its various techniques has been repeatedly shown to cause not only subjective improvement in symptoms such as headaches and visual decline but also objective improvement in visual tests[5,13] and reduction in lumbar puncture pressure.[24,29] These improvements in IIH-related symptoms, together with other health related advantages of weight reduction, have made BS a valid and even a preferred alternative for IIH treatments,[3,5,11,13,18,20,27-29] and is increasingly utilized for IIH.[7] Despite these encouraging results, shunt dependency has been reported following BS, although rare.[8]

The main finding of the current series highlights the association of BS with OD symptoms in a small group of patients that had undergone both BS and shunt surgery (not concurrently). This concept of BS-induced shunt OD has been described in a previously published case report of a patient with an LPS that underwent BS and developed secondary Chiari.[15] Symptoms and radiological findings responded to a valve upgrade.

The mechanisms by which BS may induce OD are unclear, but may include:

- Cure or improvement of IIH by weight reduction, thus making the CSF diversion redundant. It has been suggested by several authors that in this context, when significant long-term weight loss is anticipated based on a planned BS, LPS may be a valid interim solution for IIH despite the "short life expectancy of these shunts"[20,25]
- Reduced abdominal pressure secondary to weight loss may increase the pressure gradient across the shunt valve and lead to OD. We speculate that this is a major factor, and thus have continued to upgrade the valve system (eventually achieving good clinical outcome)
- Reduced abdominal pressure leads to a reduction of pressure in the inferior vena cava. This may increase venous drainage from the epidural venous (Batson) plexuses. In a similar fashion, it has been suggested that increased drainage from these plexuses may be the basis for spontaneous intracranial hypotension[12]
- BS leading to significant weight loss improves sleep study parameters and reduces obstructive sleep apnea (OSA).[22] OSA is thought to contribute to elevated ICP levels.

When relating to shunt revisions due to technical reasons (tubing disconnections, catheter migrations, and valve rotations), the rate prior to BS was higher than after. This may pertain to obesity-related factors that lead to shunt migration and valve rotations. However, the numbers are too small to draw any concrete conclusions.

Another important observation in this small patient group was the incidence of shunt dependency even after BS with effective weight reduction. In 2 patients, following several valve upgrades, we suspected that the BS had cured the IIH and actually made CSF diversion unneeded. Thus, both patients underwent externalization of the shunt. Both patients proved to be shunt-dependent with a low compliance needing effective CSF drainage within hours. We suspect that a potential explanation may be that patients with shunts have a more severe form of IIH as compared to the nonshunted patients. The shunted patients have more severe presenting symptoms of headaches or visual decline and are more prone to be refractory to medical treatments. Thus, weight loss may not be sufficient to cure their IIH.

Our current policy in the treatment of IIH is medical treatment combined with weight loss programs (including BS). When there is deterioration in visual function despite maximum treatment, we advocate CSF diversion (preferably LPS). We advocate using a restricted valve

(such as the DSV) and avoid using programmable valves (as it is difficult to palpate the exact location of the valve and assure a correct pressure programming). Despite the disappointing results of BS on IIH-related headaches in this group of previously shunted patients, BS has an important role on the general wellbeing of IIH patients suffering from morbid obesity, and we encourage patients to pursue it. We acknowledge and instruct our patients that OD symptoms may arise, and that a need for valve upgrades may arise once weight loss is achieved. If these symptoms occur, we upgrade the DSV to higher pressures and add an SA, or convert the LPS to VPS. It is accepted that VPS tend to have less malfunctions (especially obstructions) and less chiari than LPS; however, OD rates seem to be generally similar.[19,30] The reason for less shunt obstructions in VPS compared to LPS is unknown, but may be associated with the larger tube diameter and lumen compared to LPS. VPS may be more adequate for programmable shunts, as they are adjacent to the skull and safer to program.

This study has several limitations. First, the small retrospective group limits the ability to generalize our results and even perform meaningful statistics. Second, data concerning indication for shunt revision and the exact valve details were missing for 2 of 7 non-BS patients (they were previously treated elsewhere). Third, the two groups (BS and non-BS) may not represent the same IIH severity, for instance because of significantly different BMI values. Fourth, it is not absolute that BS was the trigger to the OD symptoms, as these symptoms occurred prior to BS (although at a lower rate), and also in the non-BS group. However, the rate of OD following BS (100%), makes BS a probable contributing factor in the OD occurrence.

CONCLUSIONS

BS has an important role treating morbidly obese patients with IIH. In some patients, this may result in improvement of IIH-related symptoms. However, in patients that underwent a previous shunt treatment, or in patients with prior BS that undergo a later shunt placement, BS may provoke OD symptoms, leading to future shunt related surgeries. In additional, these shunted patients may still be shunt-dependent despite undergoing BS and significantly reducing their BMI.

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Conflicts of interest
There are no conflicts of interest.

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