

Lumboperitoneal Shunt with No Valve or Antisiphon Device in Management of Idiopathic Intracranial Hypertension at The National Center of Neurological Sciences Among Sudanese Patients

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Abstract

Background: Surgical treatment of idiopathic intracranial hypertension includes cerebrospinal fluid diversion procedures most commonly lumboperitoneal shunt. Lumboperitoneal shunt addresses the cause of both headache and papilledema more directly by effecting a global reduction of intracranial pressure.

Objective: To study surgical outcome of lumboperitoneal shunt in management of idiopathic intracranial hypertension.

Methods: This is retrospective study conducted at The NCNS (January 2017– June 2019). All patients diagnosed with IIH and operated by LP shunt with no valve or antisiphon device were included in this study. The data was inserted in a designed Excel sheet and the data analysis was conducted using Statistical Packages for Social Sciences version 23.

Results: The number of patients was 22 patients. (90.9%) less than 40 years. Male to female ratio was 1:10. Presenting symptoms and signs were papilledema (100.0%), headache (95.5%), blurring of vision (72.7%), diplopia (4.5%), vomiting (18.2%), convulsion (4.5%), neck pain (4.5%), hypoesthesia on the face (4.5%), these features subsided in (66.7%), (85.7%), (57.1%) and (100.0%) for the remainder symptoms, respectively. Blindness (27.3%) and optic atrophy (54.5%) did not subside in affected cases, one case lost follow up. Operative site pain with radiculopathy in one case, subsided without intervention. Radiological images of the brain were unremarkable except in one case diagnosed with sinus thrombosis and managed with warfarin. No cognitive or motor deficit was reported. One case of convulsion was controlled with medication. Reduction in weight associated with visual improvement was reported in only one patient who lost 14kg after operation. Surgical revision was done for 27.3%. The causes for revision involved shunt malfunction, CSF leakage, slippage of catheter either from lumbar or abdominal part which may be associated with migration.

Conclusion: IIH is a serious disease with variable symptoms and signs related to high ICP and can progress to irreversible visual loss. Lumboperitoneal shunt (LP) operation is a very effective surgical procedure in management of IIH. Meticulous technique during insertion of the LP catheter is very crucial so as to optimize the efficacy of the operation and decrease the failure rate.

Keywords: lumboperitoneal shunt, surgical outcome, idiopathic intracranial hypertension

Introduction

Idiopathic intracranial hypertension is not a single disease entity but rather a complex syndrome of variable presentations. According to its clinical presentation, it may be acute, subacute, or chronic, and depending on the opening lumbar CSF pressure, it could be mild, moderate, severe, or fulminant^[1]. Surgery is often indicated for patients with failed medical treatment or medications intolerance, manometric predictors for surgical decision may include severe and fulminant opening CSF pressures as well as poor manometric response to repeated lumbar taps^[2].

Lumboperitoneal shunt is very effective procedure for management of intractable headaches and visual impairment associated with idiopathic intracranial hypertension. Its benefit can be increased by appropriate patient selection and meticulous technical placement of the shunt, which is guided by strict protocols for shunt procedures [3].

Idiopathic intracranial hypertension (IIH) is a syndrome of raised intracranial pressure in the absence of intracranial mass lesion or ventricular dilatation, associated with normal cerebrospinal fluid (CSF) composition, normal level of consciousness, and usually normal findings on neurological examination except for papilledema and occasional VI nerve palsy [4].

Diagnosis of IIH is a process of exclusion depending on clinical symptoms and neurological and ophthalmological data, as well as imaging and CSF studies. The disease reflects a nonfatal course; however, it can disturb normal life and cause significant visual impairment [5].

In the absence of a clear understanding of the pathophysiology of IIH, the appropriate and most effective treatment strategy for this complex disease remains unknown [6].

Treatment options include medical and surgical modalities. Currently, patients are often treated medically and surgery is reserved for patients with severe and acute visual loss at initial presentation or in whom medical management has failed, i.e., those with intractable headache or a permanent visual deficit [7].

Surgical treatment of IIH involve CSF diversion procedures most commonly lumboperitoneal (LP) shunt, and optic nerve sheath fenestration (ONSF) [8], and sagittal sinus stenting for certain cases. LP shunt manage the cause of headache and papilledema more directly by effecting a general reduction of intracranial pressure. However, despite its effectiveness in the early stages of IIH, LP shunt failure often occurs and it may require multiple revisions in some patients. Reasons for LP shunt revision include shunt obstruction, low intracranial pressure headache, lumbar radiculopathy, abdominal pain, and infection [9].

Rationale and justification

This study was designed to assess the outcome of lumboperitoneal shunt in management of idiopathic intracranial hypertension which is a serious disease complicated by irreversible blindness.

Research Problem

Failure rate of lumboperitoneal shunt

Research Question

Is lumboperitoneal shunt effective in management of Idiopathic intracranial hypertension

Objectives

General objectives

To study outcome of lumboperitoneal shunt with no valve or antisiphon device in management of idiopathic intracranial hypertension at The National Center of Neurological Sciences (January 2017 – June 2019).

Specific objectives

1. To determine the outcome of lumboperitoneal shunt with no valve or antisiphon device in management of idiopathic intracranial hypertension.
2. To determine the potential factors affecting the lumboperitoneal shunt with no valve or antisiphon device in management of idiopathic intracranial hypertension.

Methodology

1. Study design

This is a hospital based retrospective study.

2. Study area

The study was conducted at The National Center of Neurological Sciences at Khartoum State. The center is belonged to the federal ministry of health, and receiving neurological and neurosurgical cases. It consists of neurological and neurosurgical department. The neurosurgical department had 5 consultants, 2 specialists, and rotating registrars of neurosurgery.

3. Study duration

From January 2017 to June 2019.

4. Study population

Patients with idiopathic intracranial hypertension at The National Center of Neurological Sciences who underwent Lumboperitoneal shunt operation during the study duration.

Inclusion criteria

- Idiopathic intracranial hypertension.
- Lumboperitoneal shunt management.

Exclusion criteria

- Refused to participate.

5. Sample size

Total coverage was used to select the study sample, by which all the Patients with idiopathic intracranial hypertension and managed by lumboperitoneal shunt operation at The National Center of Neurological Sciences during the study duration and fulfill the inclusion criteria were selected. The study sample was 22 patients.

6. Data Collection tools and methods

Data was collected using a questionnaire that fulfills the objective of the study.

7. Study variables

- 1- Demographic data.
- 2- Presentations of idiopathic intracranial hypertension.
- 3- lumboperitoneal shunt management.

8. Data Analysis

Data was analyzed by Statistical Packages for Social Sciences (SPSS) 23.0. Test of hypothesis was done with Chi-square test, as appropriate $P < 0.05$ is considering statistically significant (Confidence Interval: CI 95%, Margin error 5%).

9. Ethical considerations

- Ethical clearance was obtained from SMSB Ethical Committee.
- Consent was obtained from The National Center of Neurological Sciences.

Privacy of data collected was considered (No names, data was coded; data was interpreted in form of statement tables & figures).

Results

The number of patients was 22 patients. The majority of the patients 20(90.9%) aged less than 40 years and 2(9.1%) aged between 40 – 60 years (Figure 1). The mean age was 32 ± 4.3 years.

Females were 20(90.9%) and males were 2(9.1%). Male to female ratio was 1: 10 (Figure 2).

Normal BMI reported in eight (36.4%), overweight eight (36.4%) and obese six (27.2%) (Figure 3).

Presenting symptoms were headache 21(95.5%), blurring of vision 16(72.7%), diplopia with 6CN palsy only one (4.5%), blindness, either both or one eye were six (27.3%), blindness in both eyes three (13.6%), blindness in right eye two (9.0%), blindness in left eye only one (4.5%), vomiting four (18.2%), generalized convulsion 1(4.5%) and neck pain only one patient (4.5%) (Table 1). In the majority of the patients no neurological cognitive or motor deficit was reported 21 (95.5%) and only one case reported of decrease sensation on the right region of the face (hypoesthesia) (4.5%) (Figure 4).

The duration of complains in all patients ranged between 1 – 12 months.

Severe papilledema (grade 4) reported in four (18.2%), Moderate papilledema (grade 2,3) three (13.6%), Mild papilledema (grade 1) eight (36.4%), Optic atrophy 12 (54.4%) and congested vessel three (13.6%) (Table 2). In all patients high opening pressure was reported. Diagnostic radiological images of the brain were unremarkable except in one case which was a suspicious of superior sagittal sinus thrombosis and received medication with warfarin.

The majority of the patients 16(72.7%) underwent one shunt operation, two operations in four (18.5%) patients, three in only one (4.5%) patient and four in only one (4.5%) patient (Table 3). The causes of repeated operations were slippage of the abdominal part of the LP shunt (three cases) two of them remain into the sub-cutaneous fat of the abdomen and back, while the third one migrated upward into the sub-cutaneous fat of the lower part of the right thoracic region. CSF leakage from the wound at the lumbar site was reported in one case. Slippage of lumbar part of the catheter in another case due to release of the T tube part of the catheter from the plastic fixator leading to slippage of the lumbar part outside the thecal sac, which spontaneously followed by malfunctioning of the catheter. Finally, malfunctioning of the abdominal peritoneal part was reported in the last case.

Twenty-one of the patients accessed at follow up, one patient lost follow up. The follow up of all patients has been conducted through clinical, ophthalmological exam, and diagnostic radiological images of the brain which involve CT for all patients, MRI for most, and MRV for just a few numbers. In the 21 patient's headache resolved in 18(85.7%), incomplete recovery in two (9.5%) and remain static in one (4.8%). Vomiting recovered in of all of the four patients. Blurring of vision recovered in 12(57.1%), incomplete recovery in four (19.0%). Diplopia recovered completely in the only one patient (4.8%), and 6CN palsy also recovered, while the remainder one did not recover (4.8%). Blindness remain static in all five (23.8%) out of six patients (one lost follow up).The only one case(4.8%) of decrease sensation on the right region of the face completely recovered .Only one patient(4.8%) developed intermittent abdominal pain at the site of operation associated with electrical like sensation radiated down to the right leg but that symptom resolved completely without any intervention. The only one case of generalized convulsion (4.8%) was controlled with medication, and the only one case of neck pain (4.8%) completely recovered (Table 4). No change in weight was reported except in only one patient, who showed 14kg weight reduction during post- operative follow up.

Regarding fundoscopic examination of the eye, all patients presented with papilledema which varies from mild, moderate to severe degree, 14 (66.7%) patients recovered completely, the remaining seven (33.3%) showed some improvement but still have papilledema. All Twelve (57.1%) patients came with optic atrophy either unilateral or bilateral showed no any improvement in a matter of optic nerve atrophy despite significant improvement of vision in non-blind cases.

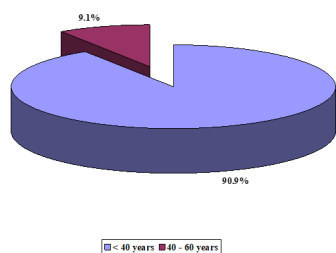


Figure 1. Distribution of the patients with idiopathic intracranial hypertension managed by lumboperitoneal shunt according to age group.

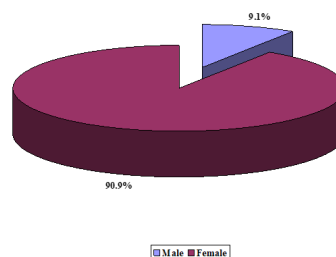


Figure 2. Distribution of the patients with idiopathic intracranial hypertension managed by lumboperitoneal shunt according to gender.

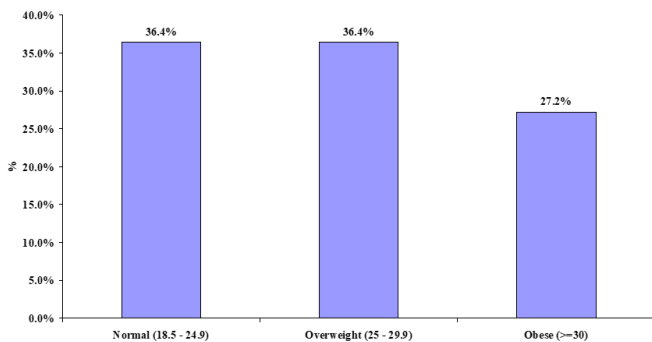


Figure 3. Distribution of the patients with idiopathic intracranial hypertension managed by lumboperitoneal shunt according to BMI.

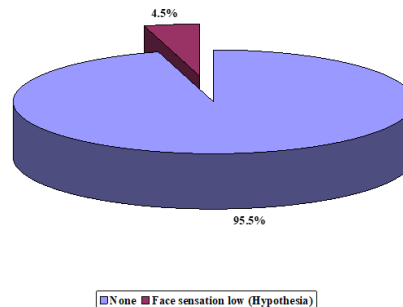


Figure 4. Distribution of the patients with idiopathic intracranial hypertension managed by lumboperitoneal shunt according to neurological deficit (Cranial Nerves were not included)

Table 1: Distribution of the patients with idiopathic intracranial hypertension managed by lumboperitoneal shunt according to presenting symptoms.

Symptoms	N	%
Headache	21/22	95.5
Convulsion	1/22	4.5
Neck pain	1/22	4.5
Vomiting	4/22	18.2
Blurring of vision	16/22	72.7
Blindness	6/22	27.3
Diplopia with unilateral 6CN palsy	1/22	4.5

Table 2: Distribution of the patients with idiopathic intracranial hypertension on initial presentation according to Visual Fundoscopic examination.

Papilledema	N	%
Mild (Grade 1)	15/22	68.2
Moderate (Grade 2,3)	3/22	13.6
Severe (Grade 4)	4/22	18.2
Optic atrophy	12/22	54.4
Unilateral optic atrophy	3/22	13.6
Bilateral optic atrophy	9/22	40.1
Congested vessel	3/22	13.6

Table 3: Distribution of the patients with idiopathic intracranial hypertension managed by lumboperitoneal shunt according to number of follow up LP shunt operations.

Number of follow up shunt op	N	%
One	16	72.7
Two	4	18.2
Three	1	4.5
Four	1	4.5
<i>Total</i>	22	100.0

Table 4: Distribution of the patients with idiopathic intracranial hypertension managed by lumboperitoneal shunt according to clinical outcome.

Outcome	N	%
Headache Outcome		
Not recovered	1	4.8
Incomplete recovery	2	9.5
Completely recovered	18	85.7
Total	21	100.0
Vomiting Outcome		
Completely recovered	4	
Total	4	100.0
Blurring of vision Outcome		
Incomplete recovery	4	25.0
Completely recovered	12	75.0
Total	16	100.0

Table continued.....

Pain at operative site with radiculopathy Outcome		
Recovered	1	
Total	1	100.0
Convulsions Outcome		
Completely recovered with medication	1	
Total	1	100.0
Blindness Outcome		
Not recovered	5	
Total	5	100.0
Diplopia with unilateral 6CN Palsy Outcome		
Completely recovered	1	
Total	1	100.0
Neck pain Outcome		
Recovered	1	
Total	1	100.0
Right site facial hypoesthesia Outcome		
Recovered	1	
Total	1	100.0

Table 5: Distribution of the patients with idiopathic intracranial hypertension managed by lumboperitoneal shunt according to ophthalmology outcome.

Outcome	N	%
Papilledema Outcome		
RECOVERED	14	66.7
Incomplete recovery	7	33.3
Total	21	100.0
Optic atrophy Outcome		
Not recovered	12	100.0
Recovered	0	0.0
Total	12	100.0

Discussion

In the present study 22 patients underwent LP shunt for management of IIH, the majority of the patients 20(90.9%) aged less than 40 years, with the mean age 32±4.3 years. Females were predominated with male to female ratio was 1:10. This is similar to a study which was done by Bruce et al and showed that more than 90% of patients are women of childbearing age^[16]. Moreover, Idiopathic Intracranial Hypertension (IIH) can occur in the pediatric population; and these younger patients are often not obese^[15].

In the present study, the hall mark presenting symptom and sign were headache (95.5%) and papilledema (100.0%) respectively. Similar to these findings, another study which was reported by González-Hernández et al.^[13], who reported headaches in almost all IIH patients, and similar to Yadav et al^[23] who also reported headache and papilledema in all patients. In the present study headache subsided immediately after the lumboperitoneal shunt operation in 18 patients (85.7%), and papilledema completely subsided gradually in 14 patients (66.7%). This outcome is similar to EL-Saadany et al ^[22] who reported that 19 patients (86.4%) showed resolution of their headache, and 16 patients (72.7%) showed complete resolution of papilledema. Headache is being described as a typically non-specific and vary in type, anatomical location, and frequency (Mark S Gans et al) ^[25]. The headache is generally described as being diffuse, worsening in the morning and being exacerbated by coughing and sneezing. The pathophysiology and mechanisms of headache in IIH is still not understood and there was no statistical relationship found between headache severity and intracranial pressure (ICP) (Mollan et al.) ^[26].

Other presenting symptoms in the present study was blurring of vision (72.7%), this is similar to Yadav et al [23] who reported (79.2%) presented with blurring and decrease vision. Blurring and distortion (ie, metamorphopsia) of central vision is mainly due to macular edema or optic neuropathy due to the effect of papilledma (Feldon SE. et al) [10]. Blurring and decrease of vision have subsided gradually postoperatively in 75% of patients in the present study, this delay in improvement can be explained by the associated gradual resolution of papilledma which is the main cause of blurring of vision (Feldon SE. et al [10]), this result was not very similar to Yadav et al [23] study which showed vision improvement in 55.6% of patients.

In the present study diplopia associated with unilateral sixth CN palsy reported in only one patient (4.5%), and completely subsided gradually. In contrast to study published by Yadav et al [23], which reported diplopia in 45.8% of patients.

Patients who present with double vision most frequently complain of horizontal displacement of the images. Horizontal diplopia is a symptom of non-localizing CN VI palsy due to high ICP. Abnormal CSF pressure can affect the eye muscles and movement producing diplopia (Bradley K. Farris et al) [27]. Vertical diplopia is rare, but it has been reported (Friedman et al) [11]. Same study by Yadav et al [23] also reported CSF leak, and temporary over-drainage in form of headache were showed in one, and 15 cases respectively. In the present study we had CSF leak in one case which was similar to the above study, but in contrast to the above study and most of studies, no any patient in the present study showed symptoms or signs of over-drainage. Over-drainage and intracranial hypotension have been overcome in most of international centers by using a valve system device with mild resistant or anti-syphon device [17]. In the present study 27.3% of patients presented with blindness either in one or both eyes, those patients did not show any signs of visual improvement after lumboperitoneal shunt operation. This result is justified as a consequence of severe and prolong effect of papilledema, which eventually leads to optic atrophy and irreversible visual loss which sometimes occurs suddenly (Feldon SE et al) [10]. In the present study 54.5% of patients presented with optic atrophy with different intensity, those who had a residual vision (27.3%) have showed significant gradual improvement in terms of vision in post-operative out-patient follow-up, despite the existence of optic atrophy in all of them. The remainder six patients who were blind, five of them (one lost follow up) remained static without showing any improvement as mentioned previously. This is similar to Yadav et al [23] study which mentioned that, only one patient, out of nine who had only perception of light and optic atrophy, has improved in vision after LP shunt operation, while all patients with vision of finger counting or better without optic atrophy improved after shunt.

This result may reveal that early identification of patients with optic nerve atrophy may have a great positive impact on post-operative visual outcome, and to avoid blindness which is a serious complication.

In the present study vomiting was reported in 18.2% of patients, generalized convulsion (4.5%), neck pain (4.5%), and hypoesthesia at the right side of the face which reported in only one patient (4.5%). Those minor presenting symptoms have been regarded as associated nonspecific symptoms (Wall M) [12], which resolved immediately after operation and high ICP returned to normal. The only one case (4.5%) presented with convulsions, has been completely controlled with medication. In the present study we had only one patient with superior sagittal sinus thrombosis diagnosed by MRV and has been managed with warfarin. In the present study no one diagnosed with cerebral venous sinus stenosis. This is markedly different from the study reported by Farb et al [14], which showed 29 patients with IIH with marked narrowing of the transverse dural venous sinus on MRV. Despite of this variation, we did not regard this as a reliable result as most of the patients in the present study did not undergo MRV, which is more sensitive (95%) than MRI (84%) in detecting sinus thrombosis, and is regarded the gold standard investigation of choice to detect sinus thrombosis (Dinkin et al) [29].

Moreover Digital subtraction angiography (DSA) was not done, which is more specific than MRV and MRI in detecting cerebral venous sinus stenosis (Hamdy M. et al) [28]. The financial and cost issues were the main obstacles to do these important investigations beside they are not part of the routine investigations in IIH patients unless there is a suspicion of sinus narrowing or thrombosis in MRI which is rare (Dinkin et al) [29].

In the present study we had only one case of pain at operative site associated with electrical shock like (radiculopathy) sensation radiated down to right lower limb, which resolved without intervention.

Radiculopathy leads to significant patient limitations in quality of life and work performance. This is due to direct spinal root irritation by the lumbar catheter. The treatment for this complication is surgical revision with repositioning of the lumbar catheter (Toma AK et al [18]).

Radicular pain (usually in the arms) is an uncommon presenting symptom (González-Hernández A et al) [13]. No patient in the present study present with this symptom.

In the present study only one case was known to be overweight preoperatively. This patient had a significant reduction in her weight, about 14 kg in post-operative follow up period. This patient showed significant improvement in terms of vision and papilledema. This is similar to a study published by Kupersmith et al [24], which reviewed 58 overweight women with IIH, divided in two groups. Thirty-eight patients underwent weight reduction and 20 patients with no weight reduction. Papilledema resolved in 28/38 in the first group, and 8/20 in the second group. These results confirmed the strong positive correlation between weight reduction and improvement of vision.

Taking the immediate surgical outcome and complications in the 22 operating patients, six (27.3%) of them required revision. One of them has been revised three times, while another one has been revised twice, and the remainder four underwent single revision. The causes or factors of repeated operations were, slippage of the abdominal part of the LP shunt in (three cases). In the first case the distal catheter (abdominal) part remained in subcutaneous fat of the abdomen, the second case has been revised twice but in the second revision the distal abdominal catheter was found collected in subcutaneous area of the back. In the third one the distal catheter migrated upward into the sub-cutaneous fat of the lower part of the right thoracic region. Those patients developed pain and uncomfortable foreign body sensation at abdominal wall and back region, and pain in the right lower thoracic region (sub-diaphragmatic), but without recurrence of headache and visual symptoms. Shunt radiological view was done for them and detected shunt migration. Shunt repositioning was done for them, and the patients were discharged in a good condition. They were symptoms-free during regular follow up. Only one case of CSF leakage from the lumbar site of the wound was reported. Again, this patient did not develop recurrence of his initial presenting symptoms. This patient underwent operative site exploration, shunt removal and replaced with a new one. The dura was found open, so it was closed in a water tight fashion and the patient was discharged in a good condition, and did not develop this complaint again. CSF leakage is a known rare complication (Sood S et al^[19]), that can be avoided by meticulous techniques, which might be influenced by the experience of the operator. Another case has showed slippage of the lumbar part, because the T tube part (proximal) catheter of the catheter has been found released from the plastic fixator and kinked which has led to malfunction of the catheter. This patient presented with headache without visual complaint. Unfortunately, her headache did not improve after operation, and the headache was controlled – to some extent – with analgesia and acetazolamide. Malfunction of abdominal peritoneal part of the catheter was recorded in the last case, she presented with headache and deterioration in vision. She underwent revision three times in different periods. She got blind, unfortunately, and was lost to follow up. Comparable to Jusue et al^[20] study which assessed the outcome, complications and surgical revision rates associated with LP shunt insertion in IHH patients, 26 patients with IHH who underwent lumboperitoneal shunt surgery for the first time by the senior author at an academy tertiary-care institution were retrospectively reviewed. Primary shunts were inserted into the 26 patients and 58% of them required revision surgery. Median time to surgical revision was 4 (3-22) months. This result was significantly higher than the present result. In the present result only 27.3% of patients underwent shunt revision as mentioned previously. Regarding the factors or causes of LP shunt revision, a study which has been done by Elshitany and Ali who followed seven patients with lumboperitoneal shunt malfunction at 1, 3, 6, and 12 months^[21]. They reported that in four of them the shunt malfunction was due to slippage of the peritoneal end either into subcutaneous fat of the abdomen (in three patients), or into subcutaneous fat of the back, with occurrence of shunt infection (in one patient), this was quite similar to the present result with the same causes of revision but none of our patients has developed post-operative infection. In another only one patient, the failure was due to the slippage of both tubes into the subcutaneous abdominal layer, while in another two patients, shunt failure was due to obstruction with adequate shunt position, this results also slightly similar to present study which has two cases underwent revision due to slippage of the lumbar part and the other one due to obstruction and shunt malfunction as mentioned previously. The present study is found to be quite different from EL-Saadany et al^[22] study(mentioned earlier) which has followed 22 patients who underwent LP shunt operation, those patients have been followed post operatively for 12 months. It has reported shunt infection in two (9%) of them who have required shunt extraction and antibiotic therapy, whereas non in the present study has developed this complication. In the same study six (27%) of patients have developed shunt obstruction and have required shunt revision, while only one patient in the present study has developed this complication.

Limitations

MRV (magnetic resonance venography) to confirm sinus thrombosis was done just for a very few cases.

DSA (digital subtraction angiography) to confirm cerebral venous sinus stenosis was not done at all.

Conclusion

Lumboperitoneal (LP) shunt is a very effective procedure in management of IHH, but needs meticulous technique to minimize the complications & rate of surgical revision.

Recommendations

- Lumboperitoneal shunt is an invasive procedure but very effective for treating intractable headaches and visual impairment associated with IHH.
- Its usefulness can be optimized by meticulous technical placement of the shunt guided by rigorous protocols for shunt procedures.
- Results in terms of improvement in vision were better in good pre- operatively vision group.
- Early detection of the disease is strongly recommended so as to achieve a better outcome especially in patients presented with optic atrophy.

- Increase the awareness of increase the aggressiveness of the disease in majority of patients by directly affects the vision leading to progressive visual loss.
- Early surgical intervention in patients who do not show improvement with medication is recommended to achieve a better prognosis especially in terms of vision.
- Na free diet, weight reduction, beside diuretics are mandatory to decrease the intensity of visual symptoms and signs during the postoperative period especially in overweight patient.

Conflict of Interest

The authors declare no conflict of interest.

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