

Controversies: Optic nerve sheath fenestration versus shunt placement for the treatment of idiopathic intracranial hypertension

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Background: Idiopathic intracranial hypertension (IIH) has been increasing in prevalence in the past decade, following the obesity epidemic. When medical treatment fails, surgical treatment options must be considered. However, controversy remains as to which surgical procedure is the preferred surgical option – optic nerve sheath fenestration (ONSF) or cerebrospinal fluid (CSF) shunting – for the long-term treatment of this syndrome. **Purpose:** To provide a clinical update of the pros and cons of ONSF versus shunt placement for the treatment of IIH. **Design:** This was a retrospective review of the current literature in the English language indexed in PubMed. **Methods:** The authors conducted a PubMed search using the following terms: Idiopathic IIH, pseudotumor cerebri, ONSF, CSF shunts, ventriculo-peritoneal shunting, and lumbo-peritoneal shunting. The authors included pertinent and significant original articles, review articles, and case reports, which revealed the new aspects and updates in these topics. **Results:** The treatment of IIH remains controversial and lacks randomized controlled clinical trial data. Treatment of IIH rests with the determination of the severity of IIH-related visual loss and headache. **Conclusion:** The decision for ONSF versus shunting is somewhat institution and surgeon dependent. ONSF is preferred for patients with visual symptoms whereas shunting is reserved for patients with headache. There are positive and negative aspects of both procedures, and a prospective, randomized, controlled trial is needed (currently underway). This article will hopefully be helpful in allowing the reader to make a more informed decision until that time.

Key words: Idiopathic intracranial hypertension, lumbo-peritoneal shunt, optic nerve sheath fenestration, pseudotumor cerebri, ventriculo-peritoneal shunt

Idiopathic intracranial hypertension (IIH), also known as pseudotumor cerebri, is a syndrome that has significantly increased in prevalence in the last decade.^[1] Since the percentage of obese (body mass index [BMI] >30) persons in the population have grown, there has been an increase in the number of cases of IIH.^[2] A direct dose-relationship has been demonstrated between increasing BMI or weight gain (>5-15%) and an increased risk for IIH.^[3] Specific medications (e.g. steroid withdrawal, lithium, tetracyclines, and vitamin A analogs) and systemic conditions (e.g. obstructive sleep apnea, renal failure, coagulopathies, and anemia) have also been associated with IIH.^[4-6] The most common IIH patients are non-obese females of childbearing age, although IIH can occur in nonobese patients, males, children, and older adults. The clinician must be careful in these atypical cases as IIH remains a diagnosis of exclusion.^[6]

The diagnosis of IIH rests upon the modified Dandy criteria, which include: (1) Signs and symptoms only consistent with increased intracranial pressure (ICP) (i.e. headaches, nausea, vomiting, transient visual obscurations, papilledema); (2) no

localizing focal neurological signs except unilateral or bilateral sixth nerve paresis or other signs associated with increased ICP; (3) cerebrospinal fluid (CSF) opening pressure ≥ 25 cm of water (20-25 cm of water are borderline measurements) and without CSF cytological or chemical abnormalities; and (4) normal neuroimaging adequate to exclude cerebral venous thrombosis (i.e. usually magnetic resonance imaging combined with magnetic resonance venogram).^[7]

The pathophysiology of IIH remains idiopathic but is most likely due to increased CSF production, reduced CSF absorption, increased cerebral venous pressure, venous sinus stenosis, increased brain water content, or a combination of these factors.^[8]

The treatment of IIH can include observation (with weight loss and diet management) for asymptomatic or mildly affected individuals; medical management (typically diuretic therapy) for symptomatic patients; and surgical management for patients failing maximum medical therapy. Treatment goals in IIH include alleviation of increased ICP symptoms (e.g. headache or diplopia) and signs (i.e. visual loss from papilledema).^[9]

Medical management can be initiated for symptomatic patients (i.e. carbonic anhydrase inhibitors, loop diuretics, topiramate). However, if conservative maximal medical management fails or the onset of disease is acute and severe, then surgical intervention should be considered. Serial lumbar punctures or a continuous lumbar drain can be employed in the acute setting, although these options are generally only appropriate for temporary use (e.g. prior to definitive surgery or during pregnancy), as the CSF reforms rapidly.^[9]

Access this article online

Website:

www.ijo.in

DOI:

10.4103/0301-4738.146012

Quick Response Code:



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Manuscript received: 28.06.14, Revision accepted: 27.08.14

The most common long-term surgical treatment options available include optic nerve sheath fenestrations (ONSFs) or CSF shunts (lumbo-peritoneal shunt [LPS], ventriculo-peritoneal shunt [VPS], or ventriculo-atrial shunt [VAS]). However, since no prospective, randomized trials have been performed to compare these procedures in a head to head manner, the preferred surgical treatment remains controversial.^[3]

Venous sinus stenting has also recently been discovered and utilized in select patients as a third surgical consideration to reduce both ICP and cerebral venous pressure. However, due to the higher and more severe rate of complications and mechanism of direct venous sinus drainage, venous sinus stenting is for select patients only.^[10] Thus, we will not cover the venous sinus stenting controversy in this manuscript. Instead, we will compare and contrast the pros and cons of two well-established surgical treatments for IIH - ONSF and CSF shunting (LPS, VPS, or VAS).

Optic Nerve Sheath Fenestration

Pros

Optic nerve sheath fenestration was first described surgically in 1872 when deWecker incised the retro-orbital optic nerve.^[11] However, the use of ONSF for the treatment of vision loss caused by IIH was not well-known until 1964 when Hayreh described the resolution of papilledema after incision of the optic nerve sheath.^[12] Thus, when headaches are not a predominant symptom, ONSF is often the preferred surgical option.^[13] Despite this convention, ONSF has recently been reported to improve headaches. In one review, a significant improvement in or resolution of headaches was reported in over half of IIH patients who were status post-ONSF. This has led some to believe that ONSF may be a superior surgical option to CSF shunting, even in IIH patients with predominantly headache symptoms.^[14]

In one of the largest retrospective, noncomparative, interventional case series studying the effectiveness of ONSF, Banta and Farris described a 94% improvement in visual acuity (148 of 158 eyes) and 88% rate of visual field stabilization or improvement (71 of 81 eyes) after ONSF. There was only one eye (<1%) that suffered any severe, vision-limiting surgical complications.^[15] Table 1 for a summary and comparison of all referenced ONSF literature reviewed.

Chandrasekaran *et al.* have reported visual outcomes (visual acuity and visual field mean deviations) in 51 eyes of 32 patients with IIH pre- and post-ONSF. Patients with only mild visual field defects were found to improve or stabilize postoperatively, while patients with more severe visual field defects were found to stabilize postoperatively. More favorable results were seen in younger patients. Complications were self-limited (e.g. diplopia, anisocoria, disc hemorrhage) and resolved without further surgical or medical intervention.^[16]

The benefit of ONSF despite multiple prior LPS placements has also been documented. In a study of 23 patients with multiple previous LPS, Sergott *et al.* demonstrated visual improvement in six patients after ONSF. This demonstrates that even with functional CSF shunts, ONSF may be beneficial for protective prophylaxis in the event of shunt failure, or in-patients with progressive visual deterioration.^[17]

In general, ONSF is an effective long-term procedure for visual loss associated with IIH. However, as would be expected, patients with acute symptoms had better responses after ONSF than patients with chronic, atrophic disc edema. Spoor and McHenry retrospectively studied postoperative ONSF outcomes in 75 eyes and found that 68% with good preoperative visual acuity had improvement (36%) or stabilization (32%) of visual function.^[18]

Another retrospective review of 62 patients described the additional benefit of unilateral ONSF to reduce the grade of papilledema in the contralateral, unoperated eye. Although the reduction in papilledema was the greatest on the operated eye, a reduction in papilledema grade was seen in the contralateral eye at 3, 6, and 12 months postoperatively. The reduction in papilledema was also correlated with a corresponding increase in visual function in both operated and contralateral eyes. The mechanism of contralateral eye improvement after unilateral ONSF is not entirely clear, but is hypothesized to be related to decreased CSF content in both sheaths after unilateral fenestration, spontaneous improvement, or improved patient compliance post-surgically.^[19]

Cons

Although many benefits of ONSF have been demonstrated, complications can also occur. Many studies have reported that the majority of complications are usually transient and resolve without sequelae.^[14-16,20]

However, complication rates seem to vary greatly between different studies. Using a medial orbitotomy approach for optic nerve sheath decompression, Plotnik and Kosmorsky reported a 40% overall complication rate, including temporary motility disorders (29%); pupillary dysfunction (11%); and vascular complications (11%) comprised of two central retinal artery occlusions, one episode of transient outer retinal ischemia, and one superotemporal branch retinal artery occlusion.^[21] In contrast, Chandrasekaran *et al.* found a 15.6% overall complication rate (5 out of 32 patients) after ONSF. All complications were self-limiting (three patients had diplopia, two had anisocoria, and one patient had a disc hemorrhage). In this study, all surgeries were also performed by the same surgeon using a medial subconjunctival approach.^[16] Although there is a wide range of complication rates associated with ONSF, most studies are in agreement with a range between 5% and 45%.^[20]

To better characterize the etiology and management of severe complications associated with ONSF, Mauriello, *et al.* retrospectively studied post-ONSF patients, looking specifically for patients with post-operative visual decline within 1 month of the procedure. The authors identified 5 patients with the following complications: One post-operative retrobulbar bleed, one infectious post-operative optic neuropathy, and three gradually declined in vision without a particular surgical complication identified (mechanism was suspected to be persistently elevated ICP). Four out of the five patients experienced an improvement of vision after a LPS was placed. The patient with the suspected post-operative infection also experienced visual recovery after treatment on intravenous (IV) antibiotics, even though no infectious organism was identified.^[22]

Table 1: ONSF Article Summary

Authors and year	# of patients	# of eyes	Pre-ONSF visual acuity (range)	Acuity improved or stable	Visual acuity improved	Visual field improved	Visual acuity worsen	Visual field worsen	Visual acuity stable	Visual field stable	Complications
Sergott <i>et al.</i> , 1988 ^[17]	23	29	20/20-20/400	23	21	29	1	0	2	0	Conjunctival filtering bleb (one patient), adduction deficit (one patient)
Spoor <i>et al.</i> , 1993 ^[18] - acute cases	35	69	20/20-20/400	69	44	68	0		25		Excessive scarring, arachnoidal adhesions, swollen optic disc, atrophic optic disc
Spoor <i>et al.</i> , 1993 ^[18] - chronic cases	18	32	20/20-20/200	32	14	7	0		18		
Banta and Farris, 2000 ^[15]	86	158		148		71	10	10			Diplopia, dellen, anisocoria, orbital apex syndrome, presumed traumatic optic neuropathy
Chandrasekaran <i>et al.</i> , 2006 ^[16]	32	51	6/6-6/18	32	13	13	10	1	19	17	Diplopia, anisocoria, disc hemorrhage
Mauriello <i>et al.</i> , 1995 ^[22]	5	6	20/20-20/80				4	1			Nerve sheath vessel bleed, infectious optic neuropathy
Wilkes and Siatkowski, 2009 ^[23]	1	2	20/25 OD, 20/40 OS				2	2			Progressive optic neuropathy
Rizzo III and Lessell, 1994 ^[27]	2	2					2	2			Choroidal infarction
Flynn <i>et al.</i> , 1994 ^[26]	1	1	20/400 OD, 20/20 OS				1				Transient blindness
Corbett <i>et al.</i> , 1988 ^[28]	28	40	20/20-20/200	34	12	21	6	7	22	10	Permanent tonic pupils, accommodative paresis, loss of vision, retrobulbar hemorrhage

KEY: Table summarizing similar retrospective reviews of optic nerve sheath fenestration. All numbers presented are numbers of patients. If certain categories were not included in the article, that section was left blank in the Table, ONSF: Optic nerve sheath fenestrations, OD: Oculus dexter, OS: Oculus sinister

Although various studies state the majority of ONSF complications are usually not severe or vision-threatening, there have been cases reported with devastating outcomes after ONSF. One case was a 16-year-old woman who presented with decreased vision secondary to IIH (right eye 20/25 and left eye 20/40). She underwent ONSF and initially improved after the first week. Fourteen days post-operatively; however, she experienced worsening of her headache, further decline in vision, and was found to have an opening pressure of 65 cm H₂O on lumbar puncture. She then underwent VPS, but her vision continued to decline, with optic atrophy observed at 10 months post-operatively (20/150 in the right eye and 20/125 in the left eye). Possible etiologies for the continued visual decline of this patient could include previous irreversible damage that occurred before the ONSF, or persistent and prolonged elevated ICP.^[23] Gellrich *et al.* observed a significant reduction in the number and size of retinal ganglion and amacrine cells post-ONSF in an experimental Wistar rat model, despite surgical and histological exclusion of iatrogenic optic nerve axon damage and verification of ocular perfusion. This suggests that other factors may be contributing to cellular damage during ONSF and further studies to elucidate this mechanism are needed. This also suggests that ONSF is not necessarily protective against further optic nerve damage

and patients must be monitored very closely after ONSF. It also questions the isolated use of ONSF without shunting in preventing eventual visual loss.^[24]

In another IIH case with ONSF via the medial approach, the patient presented 6 h after the procedure with no light perception (NLP) vision in the operated eye. After 36 h of treatment with IV methylprednisolone and oral acetazolamide, the patient began to recover vision. Three months later, vision had recovered to 20/30. The proposed mechanism was hypothesized to be axonal demyelination possibly due to a stretch injury with a subsequent re-myelination.^[25]

A similar case of "NLP" vision was described by Flynn *et al.* 5 h after ONSF through the medial approach. After 12 h of IV steroids, the vision recovered to 20/800, only slightly worse than the pre-operative vision of 20/400. The mechanism was speculated to be likely multi-factorial, possibly due to axonal stretch with an interruption in axoplasmic flow, or perhaps secondary to the development of steroid-responsive extraneural edema.^[26] Two other cases were reported with significant temporal visual field loss due to choroidal infarctions after ONSF (also using a medial approach technique). The mechanism of choroidal infarction was postulated to be excessive manipulation of the vortex veins or retraction of the globe.^[27]

Corbett *et al.* performed ONSFs on 40 eyes using the lateral orbitotomy approach instead of the more commonly described medial approach. Visual acuity improved in only 30% (12 eyes), remained stable in 55% (22 eyes), and worsened in 15% (6 eyes). In addition to the eyes with visual worsening, other significant complications included: 16 eyes with permanent tonic pupils, one retrobulbar hemorrhage, and one sixth nerve palsy.^[28] These cases suggest that the rate of complications can be dependent upon not only the amount of stretch placed on the optic nerve, but ONSF surgical techniques.

Although some improvement in headaches has been reported after ONSF, CSF shunting is generally regarded as a more effective procedure to improve headache symptoms.^[29] Some studies report up to 50% improvement in headaches post-ONSF, although this still leaves at least 50% of patients requiring further medication or having to undergo another procedure to treat intractable headaches.^[13-14,28] Banta and Farris reported even less favorable outcomes for headache symptoms; in 61 patients who actually presented with headaches as their primary symptom, only 8 of these patients (31%) reported improvement in their headaches post-ONSF.^[15]

In summary, ONSF has been shown to stabilize or improve visual loss due to papilledema in many but not all patients with IIH. The major complications are direct orbital and intraocular related effects and a worsening of visual function. ONSF is generally safe however, and to our knowledge there have been no reported mortalities from the procedure, but systemic anesthesia-related complications can obviously occur. Although bilateral surgery may eventually be necessary, many patients have sufficient visual improvement from unilateral surgery. Although the mechanism remains unclear, headaches can improve after ONSF but to a lesser degree than that reported for CSF shunting.

Shunts (Ventriculo-Peritoneal, Ventriculo-Atrial, and Lumbo-Peritoneal)

Pros

While ONSFs are generally favored in patients with severe visual loss and minimal to no headache, VPS, VAS, or LPS is the preferred treatment for IIH in patients with mild to moderate refractory headache with or without visual loss due to papilledema, or in patients symptomatic headache failing maximal medical therapy alone.^[29] In addition, some institutions may not have access to ONSF, and thus CSF shunting may be the only available surgical option for refractory IIH.

Although no prospective head to head comparative studies have been performed to evaluate the difference in effectiveness of VPS versus LPS, retrospective reviews have not revealed any significant differences in outcomes.^[29] Table 2 for a summary and comparison of all referenced CSF shunting literature reviewed.

In one retrospective review of 34 patients with 63 shunt placements by Tarnaris *et al.*, no significant difference in visual outcomes or headaches was observed between patients receiving VPS when compared to LPS. However, more first time complications and revisions were observed in the

LPS group when compared to the VPS group, even though LPS have the theoretic advantage of avoiding acute intracranial complications. These authors did not find any identifiable factors to help predict patient outcomes or whether either shunt would need revision. Overall, as would be expected, headaches improved more than visual disturbances for both groups.^[30]

Another retrospective review by Abubaker *et al.* studied outcomes of 25 different IIH patients post-CSF shunting (72% LPS and 28% VPS). Although Tarnaris *et al.* had suggested that VPS lasted longer in duration than LPS,^[30] Abubaker found a lower failure rate for LPS (11%) versus VPS (14%), but a higher revision rate for LPS (60%) versus VPS (30%). Both types of shunts improved patient symptoms (headaches, visual changes) significantly (89% improvement post-LPS, 80% improvement post-VPS).^[31] In the 10 year retrospective review of 53 patients with IIH, Sinclair *et al.* reported at 12 months post-CSF shunt placement, significant improvements were demonstrated in visual acuity (logMAR scores of 0.31 ± 0.36 at baseline to 0.10 ± 0.30), visual obscurations (53% improvement), deteriorating visual deficits (43% improvement), papilledema (44% improvement), and headaches (19% improvement). LPS made up 92.5% of shunting procedures, while VPS made up 7.5% of shunting procedures.^[32]

In another retrospective review of 22 IIH patients with LPS by El-Saadany *et al.*, patients with both headaches (86.4% or 19/22) and papilledema (72.7% or 16/22) showed significant improvements post-LPS. Consistent with previous studies, the authors found LPS to be a generally effective procedure. The authors also suggested that targeting patients more likely to benefit from LPS could enhance the effectiveness of the procedure. Predictors for increased success in LPS were patients with severe or fulminant CSF pressures or poor manometric response to repeated lumbar taps.^[33]

Cons

Despite decreasing ICP, CSF shunts do not always improve symptoms; this could partly be due to the non-ICP related nature of many IIH associated headaches. In their 10 year review, even though Sinclair *et al.* reported an overall improvement in visual symptoms post-shunting, headaches remained in a majority of patients (79%). In addition, shunt revisions were commonplace with 51% of the patients requiring a shunt revision and 30% requiring multiple revisions. Although CSF shunting was found helpful in halting visual deterioration, the authors suggested that due to the high rate of shunt complications, shunt revisions, and persistent post-shunt headaches, headaches should not be the sole indication for shunting.^[32] Similar to the study reported by Sinclair *et al.*, Karabatsou *et al.* retrospectively reviewed the outcomes of 21 patients post-LPS placement and also reported high shunt revision rates. There was an average of 3 revisions per patient, with a total of 63 revisions in 21 patients over an average follow-up period of 24 months. Only 3 patients did not undergo revision. Other complications included 17 shunt migrations, one case of temporary radiculopathy, 7 shunt-related infections (1% infection rate per procedure, or 33% per patient), 7 patients with tonsillar herniation (although only two were symptomatic). Of the two symptomatic tonsillar herniation patients, one had a VPS placed, and the other ultimately required a sub-occipital decompression.^[34]

Table 2: CSF Shunting Article Summary

Authors and year	Type of shunt	# of patients	# of patients with shunt revisions	Headache improved	Headache no change/worse	Visual outcome improved	Visual outcome no change/worse	Papilledema treated	Papilledema Improved	Papilledema unchanged	Complications
Tarnaris <i>et al.</i> , 2011 ^[30]	VPS	5	2	3	2	2	3				Shunt infection, shunt obstruction, intraabdominal pain, low-pressure type headaches, malposition of catheter tip in the lumbar spinal canal, CSF leakage through the wound
	LPS	24	10	17	7	10	14				
Abubaker <i>et al.</i> , 2011 ^[31]	VPS	10	2	8	1	8			1		Chronic subdural hematoma, distal end dislodgement
	LPS	18	10	11	2	18	2	11		1	Shunt infection, new pressure headaches, radicular pain, abdominal pain, shunt blockage, distal end and proximal end mal-position
Sinclair <i>et al.</i> , 2011 ^[32]	Both		27								Shunt block, shunt migration, shunt disconnection
	VPS	4									
	LPS	49									
El-Saadany <i>et al.</i> , 2012 ^[33]	LPS	12	6	3	19			16	2	2	Shunt infection, shunt obstruction due to migration of distal catheter, shunt overdrainage
Karabatsou <i>et al.</i> , 2004 ^[34]	LPS	21	18								Migration of shunt, fracture of shunt, radiculopathy, shunt infections, tonsillar herniation

KEY: Table summarizing similar retrospective reviews of cerebrospinal fluid shunting. All numbers presented are numbers of patients. If certain categories were not included in the article, that section was left blank in the Table, CSF: Cerebrospinal fluid, VPS: Ventriculo-peritoneal shunt, LPS: Lumbo-peritoneal shunt

El-Saadany *et al.*, although reporting an improvement in headaches in the majority of patients, also reported a 9% shunt infection rate, 27% rate of shunt obstruction, and 13% rate of shunt over-drainage. Delayed timing of procedure scheduling or a prolonged duration of surgery may have been a factor in the cause of infection in those patients. Migration of the peritoneal catheter was the most common cause for obstruction, which was attributed to the technical and hardware-related factors for anchoring the catheter.^[33] Tarnaris *et al.* reported that overall, 20.5% of patients that underwent shunting procedures had complications, and 35% ultimately required a shunt revision. Complications included shunt infection, shunt obstruction, intra-abdominal pain, and CSF leak. LPS were advantageous in avoiding intracranial complications but involved more problems with infections,

subdural hematomas, cerebellar tonsil descent, and distal catheter migration and obstruction, increasing the need for greater revisions. VPS are suggested to have lower revision rates, but the number of true revisions could be distorted by the fact that far fewer VPS are performed than LPS. VPS also carry the much greater feared intracranial complications, as well as increased difficulty with shunt placement within the small ventricles of IIH patients.^[30] Nevertheless, it has been our practice at our institution to recommend stereotactically placed, programmable valve VPS over LPS for patients with symptomatic IIH who have failed conservative and maximal medical therapy for headache and visual loss due to papilledema.

The incidence of CSF shunting for IIH has increased by up to 350% nationwide (between 1988 and 2002). Out of

2779 admissions for CSF shunting in IIH patients, overall in-patient mortality was 0.5% (0.9% mortality rate for VPS and 0.3% LPS). Median length of hospital stay was 3 days for each type of shunt, and median hospital charges were \$12,050 for VPS, and \$10,400 for LPS. Limitations, however, included: CSF shunting for IIH was substituted for the true incidence of IIH, so true incidence may be much higher; the national hospital discharge database was used as a data source for IIH, which could produce biased data since many IIH cases are diagnosed and treated in the outpatient setting; and since specific patients cannot be identified in this database (only total number of shunt procedures performed), first time shunting versus shunt revision rates cannot accurately be determined.^[35]

Conclusions

The treatment of IIH remains controversial and lacks randomized controlled clinical trial data (although one such trial is underway in the United States).^[36] The decision for treatment of IIH rests with the determination of the severity of IIH related visual loss and headache. Patients who fail conservative (weight loss) and maximal medical therapy for papilledema and headache might require surgical intervention.^[37] We prefer ONSF for patients with predominantly visual symptoms and reserve VPS or LPS for patients with headache (with or without visual loss) who fail maximal medical therapy for ICP and headache. We prefer stereotactic placed programmable valve VPS, but the literature suggests that the procedures have similar morbidity and efficacy. The VPS however carries a higher in-patient mortality risk (0.9%) compared with LPS (0.3%) and to our knowledge despite the possible intraocular and intraorbital morbidity no patient has ever died from an ONSF. Ultimately, the decision for which surgical procedure is best in IIH remains surgeon and institution dependent based upon the local surgical expertise and availability.

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Cite this article as: Spitze A, Lam P, Al-Zubidi N, Yalamanchili S, Lee AG. Controversies: Optic nerve sheath fenestration versus shunt placement for the treatment of idiopathic intracranial hypertension. *Indian J Ophthalmol* 2014;62:1015-21.

Source of Support: This work was supported in part by an unrestricted grant from Research to Prevent Blindness (RPB) to the University of Texas Medical Branch, Galveston, Texas, USA. **Conflict of Interest:** None declared.