



Posterior vault distraction osteogenesis: indications and expectations

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Abstract

Cranial vault remodeling (CVR) in patients with craniosynostosis serves to correct abnormal skull morphology and increase intracranial volume to prevent or treat pathologic increases in intracranial pressure (Taylor and Bartlett, *Plast Reconstr Surg* 140: 82e-93e, 2017). Distraction osteogenesis is a well-established technique for bony repositioning and growth stimulation in the facial and long bones, in which the gradual separation of bony segments at an osteotomy site results in generation of new bone and subsequent bone lengthening (Greene, 2018). While initially described in the orthopedic literature, the relevance and applicability of distraction osteogenesis to craniofacial surgery has been well-studied and is now well-established (Steinbacher et al., *Plast Reconstr Surg* 127: 792-801, 2011). Posterior cranial vault distraction osteogenesis (PVDO) was introduced as a treatment option for cranial vault expansion in patients with craniosynostosis in 2009 by White et al., based upon the premise that posterior vault distraction could provide greater intracranial volume expansion than fronto-orbital advancement and remodeling (FOAR), but that acute posterior cranial vault expansions were limited by the soft tissue envelope of the infant scalp and prone to relapse related to the supine positioning typical of infants (White et al., *Childs Nerv Syst* 25: 231-236, 2009). Since this introduction, significant evidence has accrued regarding the safety of, and outcomes after, PVDO. PVDO is now known to provide larger increases in intracranial volume in comparison to anterior cranial vault remodeling procedures (Derderian et al., *Plast Reconstr Surg* 135:1665-1672, 2015) and to provide morphologic improvements in both the posterior and anterior cranial vaults (Goldstein et al., *Plast Reconstr Surg* 131:1367-1375, 2013). Perioperative major morbidity is comparable to conventional vault remodeling (Taylor et al., *Plast Reconstr Surg* 129:674e-680e, 2012) and the procedure has been safely applied to patients of various ages with syndromic and non-syndromic craniosynostosis (Zhang et al., *J Craniofac Surg* 29:566-571, 2018; Li et al., *J Craniofac Surg* 27:1165-1169, 2016). Many high-volume craniofacial centers now consider PVDO the preferred first operation in infants with syndromic craniosynostosis, and indications for this procedure continue to expand as evidence accrues regarding its utility and safety (Steinbacher et al., *Plast Reconstr Surg* 127: 792-801, 2011; Swanson et al., *Plast Reconstr Surg* 137:829e-841e, 2016).

Keywords Craniosynostosis · Distraction osteogenesis · Posterior cranial vault distraction osteogenesis · Cranial vault reconstruction

The rationale for posterior vault distraction osteogenesis

Posterior vault distraction osteogenesis is by design (and in contradistinction to conventional cranial vault remodeling) a non-devascularizing operation relative to the cranial bones.

The osteotomized transport segment remains pedicled on the underlying dura, thus preserving blood supply to the bone even after craniotomy [1]. Distraction histogenesis of the overlying scalp soft tissues accompanies the underlying bony movement as the distractors are activated, in theory decreasing the risk for trauma and ischemic injury to the skin in the context of large bony advancements. Due to new vascularized bone creation, the propensity for bony relapse is decreased and requirement for use of hardware to maintain advancement is eliminated [1]. Overall, larger bony movements can be both achieved and maintained with distraction osteogenesis due to the collective expansion of bone and soft tissue [2, 3].

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Indications for posterior vault distraction osteogenesis

Posterior vault distraction osteogenesis was originally described for volumetric expansion and morphologic improvement of patients with syndromic multi-suture synostosis and accompanying turribrachycephaly [4, 5]. The use of PVDO has been expanded to the population of non-syndromic craniosynostosis patients, with phenotypes treated using PVDO including non-syndromic bicoronal, multi-sutural (e.g., 2 or more major cranial sutures), and pan-sutural (e.g., 3 or more major cranial sutures [5]). Published uses for PVDO have grown and phenotypes of non-syndromic craniosynostosis patients are varied enough such that at this juncture, there is not a simply defined phenotype for which PVDO is definitively indicated or not indicated. Noting the variability in syndromic and non-syndromic craniosynostosis presentations, above all, the determination of PVDO as an appropriate treatment should consider (1) the degree of cranial volume expansion required and (2) the phenotypic result desired.

Retrospective analyses of patients undergoing PVDO have helped to delineate the operation's role in varying patient populations. A 2018 study by Zhang et al. compared populations of syndromic and non-syndromic patients undergoing PVDO with the findings that non-syndromic patients undergoing PVDO were older (mean age of 4.17 years for non-syndromic, mean age of 2.72 years for syndromic), less likely to carry a diagnosis of bicoronal synostosis, and more likely to present with clinical signs of increased ICP (all $p < 0.05$). These data suggest that PVDO was chosen for syndromic populations at a younger age (presumably, to both preemptively expand the cranial vault and improve anterior and posterior cranial vault morphology), and in older, non-syndromic populations to relieve increased intracranial pressure in settings in which a major change in cranial shape and/or morphology was likely less necessary [5].

Two particular patient groups in whom PVDO should be considered include (1) syndromic craniosynostosis patients with turribrachycephaly [4], with a role to both preemptively expand the cranial vault and improve cranial vault morphology, and (2) syndromic or non-syndromic patients in whom cranial expansion is required, with varying degrees of brachycephaly, but in whom a frontal reshaping operation is not necessarily indicated. At the Children's Hospital of Philadelphia, PVDO is a cornerstone of our algorithm to treat patients with syndromic craniosynostosis (see "Treatment algorithms utilizing PVDO" and Fig. 1), whereas its use in non-syndromic craniosynostosis patients and older patients remains a case-by-case decision based upon the aforementioned clinical findings.

Clinical outcomes after posterior vault distraction osteogenesis

Intracranial volume

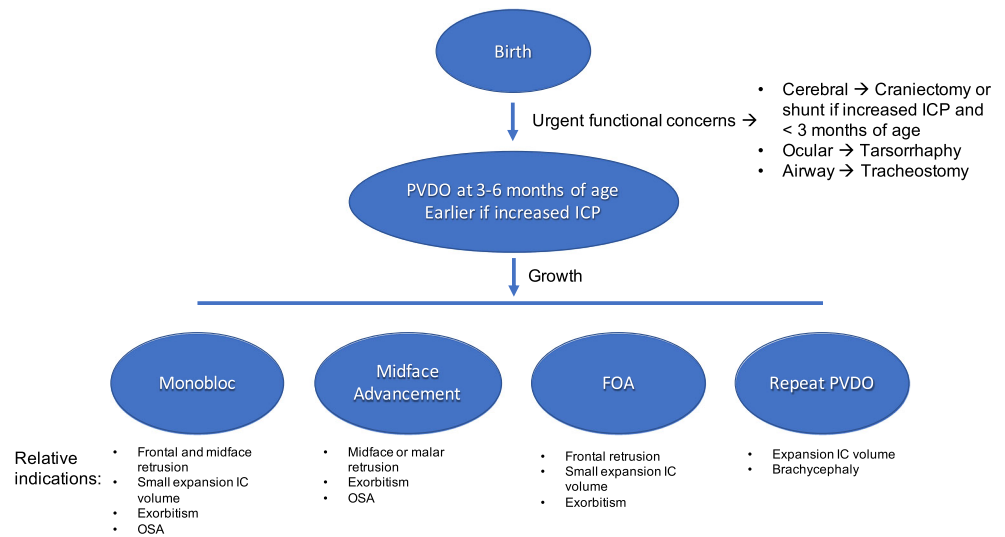
Numerous studies have been performed on PVDO's capacity for volumetric and craniometric expansion, and the findings of these studies are summarized in Table 1. PVDO has been consistently shown to increase intracranial volume by between 20 and 30% and to provide a greater increase in intracranial volume than anterior vault remodeling [3, 6–9, 12, 15]. The posterior fossa demonstrates the greatest increase in size relative to the remaining intracranial space [13].

Conflicting data exist regarding whether this difference in volume achieved after posterior versus anterior cranial vault surgery is attributable directly to a greater increase in intracranial volume per millimeter distracted or, more simply, to the fact that greater overall distances of distraction are possible in the posterior vault. A 2015 retrospective study by Derderian et al. compared craniometric measurements after FOAR and PVDO and identified that while the volumetric gains achieved per millimeter of bony advancement are similar in the two operations, total expansion with PVDO was almost twice that of FOAR [3]. Derderian et al.'s data differed slightly from a 2012 study by Choi and colleagues who identified significantly greater increases in intracranial volume after PVDO compared to FOAR when controlling for distance of advancement. Specifically, PVDO was found to increase intracranial volume by 23.9% after a 20 mm advancement, in comparison to a 17.7% increase in intracranial volume after a 20 mm fronto-orbital advancement, indicating a 35% greater increase in intracranial volume with PVDO compared to fronto-orbital advancement with the same distance of distraction [8]. Of note, this latter referenced study was a theoretical study performed with computer-simulated distraction as opposed to pre- and postoperative CT scans of patients undergoing FOAR and PVDO.

Craniometrics and cranial morphology

Craniometric alterations after PVDO have been demonstrated in both the anterior and posterior segments of the cranial vault and are summarized in Table 2. Posterior cranial height increases by 12.2% on average [9], and the posterior cranial base length and size of foramen magnum both increase [15]. No changes in anterior or middle cranial vault *height* have been identified to date, but improvements in overall anterior cranial vault *morphology* have been consistently established. Goldstein et al. demonstrated a decrease in the baso-frontal angle (angle generated by lines from sella-nasion and nasion-anterior most aspect of frontal bone, Fig. 2) by 3.9%, indicating improvement in

Fig. 1 The Children’s Hospital of Philadelphia algorithm for the early use of posterior vault distraction osteogenesis in the operative management of patients with syndromic craniosynostosis. ICP, intracranial pressure; PVDO, posterior vault distraction osteogenesis; FOA, fronto-orbital advancement; OSA, obstructive sleep apnea. Reprinted with permission from: Swanson JW, Samra F, Bauder A, Mitchell BT, Taylor JA, Bartlett SP (2016) An algorithm for managing syndromic craniosynostosis using posterior vault distraction osteogenesis. *Plast Reconstr Surg* 137: 829e-841e



frontal bossing after PVDO. These findings have been corroborated by additional direct craniometric measurements by Ter Maaten et al., showing a decrease in the baso-frontal angle between approximately 2 and 4 degrees, and a significant decrease in directly measured supraorbital retrusion after PVDO [12].

Morphologic and craniometric outcome variation by patient age

The aforementioned morphologic and craniometric alterations vary by age of the patient, with studies generally identifying increased magnitude of change and/or improvement in

Table 1 Volumetric changes after posterior vault distraction osteogenesis

Study	Patients	Changes in intracranial volume
Serlo et al. [6]	N = 10	Increase by 20.2% (10.2–28.5%)
Nowinski et al. [7]	N = 2	Increase by 22–29% Overall greater volumetric increase than vault expansion with floating posterior cranial bone flap (13–24%) or lambdoid springs (18–25%)
Choi et al. [8]	N = 13	Increase by 23.9% 35% greater volumetric increase than vault expansion with FOAR (17.7%)
Goldstein et al. [9]	N = 11	Increase by 21% (7.5–50%) Increase by 28% (10.8–66%) in age < 1 year
Derderian et al. [3]	N = 15	Increase by 274 cm ³ Greater volumetric increase than expansion with FOAR (144 cm ³) Gain per millimeter of advancement similar
Shimizu et al. [10]	N = 7	Increase by 21% (13–34%)
Salokorpi et al. [11]	N = 31	Increase by 20.8% (19.3–21.9%)
Ter Maaten et al. [12]	N = 12	Increase by 249 cm ³ or 23% Increase by 45.5% in age < 1 year Increase by 10.5% in age > 1 year
Bauder et al. [13]	N = 10	Increase by 299 cm ³
Di Rocco et al. [14]	N = 21	Increase by 186 cm ³ (SD 86 cm ³) or 13.9% (SD 11.9%)

FOAR, fronto-orbital advancement and remodeling

Table 2 Craniometric changes after posterior vault distraction osteogenesis

Study	Patients	Anterior craniometrics	Posterior craniometrics
Goldstein et al. [9]	$N = 11$	Baso-frontal angle decreases by 3.9%	Posterior cranial height increases by 12.5% Posterior cranial height increases by 19.5% in patients < 1 year
Samra et al. [16]	$N = 7$	Frontal bossing angle decreases by 7.6%	
Ter Maaten et al. [12]	$N = 12$	Supraorbital retrusion decreases from 5.44 to 4.45mm Baso-frontal angle decreases by 2.92 degrees Baso-frontal angle decreases by 3.33 degrees in age < 1 year Baso-frontal angle decreases by 2.58 degrees in age > 1 year	

younger patients. Specifically, infants < 12 months of age undergoing PVDO have been found to differ from a cohort including all ages of patients in that increase in intracranial volume is larger (Goldstein et al.: 28.4% versus 21% [9]; Ter Maaten et al.: 45.5% versus 10.5% [12]) and that increase in posterior cranial height is greater (Goldstein et al.: 19.5% versus 12.2% [9]). The baso-frontal angle has been found to decrease by 3.33 degrees in patients < 1 year of age and by 2.58 degrees in patients > 1 year of age [9].

Functional outcomes

A critical corollary to the volumetric and morphologic gains of PVDO is the presence of functional improvement in intracranial pressure elevation, cerebrospinal fluid hydrodynamics, and presence or severity of a Chiari malformation after the operation. In its original description, PVDO was confirmed to improve

intracranial pressure in all subjects when measured by fundoscopy and/or radiologic evaluation [4]. Subsequently, a retrospective study by Zhang et al. identified resolution of increased ICP in syndromic and non-syndromic patients after PVDO (determined by reduction in irritability, headaches, nausea, and ophthalmologic data (papilledema) when available) [5].

Given the craniometrically confirmed anatomical expansion of the posterior fossa achieved with PVDO, it furthermore makes empiric sense that Chiari malformations may be improved and/or treated by PVDO and that cerebrospinal fluid hydrodynamics may be altered or improved. A retrospective study by Lin et al. identified that syndromic patients treated with PVDO were less likely to develop a new-onset Chiari malformation after PVDO than after conventional cranial vault remodeling [17]. In this same series, two patients experienced improvement in Chiari malformation postoperatively after PVDO, while no patients experienced the same after conventional open posterior vault remodeling [17]. No difference was identified in the rate of new-onset hydrocephalus requiring shunt or in improvement in shunted hydrocephalus [17]. A retrospective study by Di Rocco et al. identified a decrease in symptoms attributable to tonsillar herniation after PVDO (sleep apnea, papilledema, headaches); however, a significant change in the measurement of tonsillar herniation on imaging was not able to be established [14].

Spruijt et al. identified a decreased incidence of tonsillar herniation and papilledema in patients after occipital expansion when compared to patients after fronto-orbital advancement [18]. This study was performed in patients undergoing acute occipital expansion (not distraction osteogenesis); however, their findings are worth noting in the context of a discussion of functional improvements after PVDO given that one would expect even greater volumetric gains after PVDO than after acute occipital expansion [18, 19]. While definitive indications for PVDO for the treatment of intracranial pathology other than increased intracranial pressure have not been established, it stands to reason that it may be beneficial in treating some forms of idiopathic intracranial hypertension and slit ventricle syndrome.

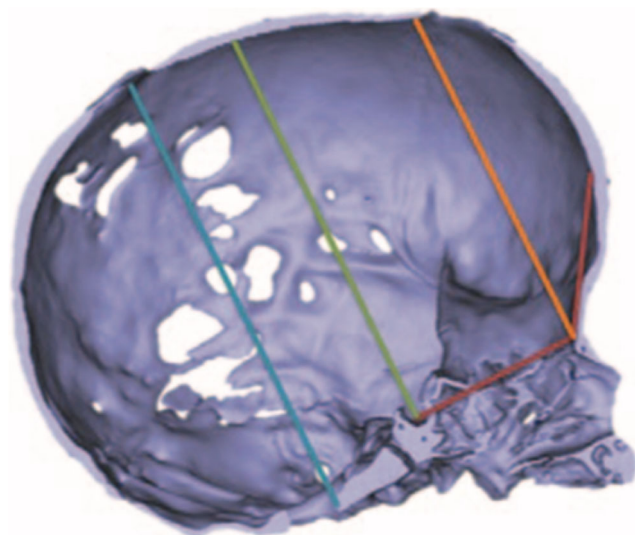


Fig. 2 Craniometric measurements in posterior vault distraction osteogenesis. Anterior cranial height, orange; middle cranial height, green; posterior cranial height, blue; baso-frontal angle, red. Reprinted with permission from: Goldstein JA, Paliga JT, Wink JD, Low DW, Bartlett SP, Taylor JA (2013) A craniometric analysis of posterior cranial vault distraction osteogenesis. *Plast Reconstr Surg* 131: 1367-1375

Long-term expectations after PVDO

Anterior cranial vault and midfacial surgery

The greatest benefits of PVDO are perhaps identified in the context of additional cranial surgery needs in patients with syndromic craniosynostosis. Early performance of PVDO may delay the need for fronto-orbital advancement or decrease the number of procedures required to achieve desired frontal morphology in the syndromic craniosynostosis population [20]. A Kaplan-Meier analysis of patients undergoing PVDO showed that early PVDO both increases the time to requirement of frontal remodeling surgery and decreases the number of frontal remodeling procedures required in the first 5 years of life [20] (Fig. 3). Delaying FOA to a time frame in which bony relapse is less significant and less likely may decrease the need for additional/secondary interventions [9]. A significant delay in the need for frontal surgery may allow for the choice of monobloc distraction to address brow position, corneal position, and midface hypoplasia in one operation, rather than in a combination of frontal surgery and midfacial surgery timed over several years [9]. Of note, FOAR after PVDO appears to be increasingly technically challenging, with a retrospective study by Zhang et al. identifying longer anesthetic and operative durations and an increase in technical difficulty with closure in patients with a history of PVDO undergoing FOAR, compared to patients undergoing primary FOAR [21].

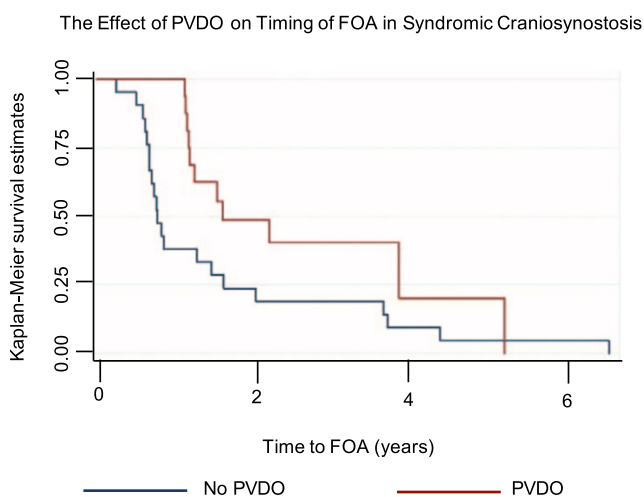


Fig. 3 Kaplan-Meier analysis identifies increased time to first fronto-orbital operation in patients with syndromic craniosynostosis who underwent posterior vault distraction osteogenesis. PVDO, posterior vault distraction osteogenesis; FOA, fronto-orbital advancement. Reprinted with permission from: Swanson JW, Samra F, Bauder A, Mitchell BT, Taylor JA, Bartlett SP (2016) An algorithm for managing syndromic craniosynostosis using posterior vault distraction osteogenesis. *Plast Reconstr Surg* 137: 829e-841e

Posterior fossa physiology

The long-term effects of improvement of posterior fossa morphology are less well defined than the effects on frontal anatomy and the subsequent need for additional cranial operations. Apart from the decreased likelihood of a new-onset Chiari malformation, the described benefits on intracranial circulation and venous outflow and the physiologic improvements to the cranial base have not been definitively shown to improve intracranial physiology over a prolonged period of time [4].

New-onset craniosynostosis

An interesting radiological finding after PVDO is new-onset suture closure in previous unfused sutures [22]. The postoperative fusion of open cranial vault sutures was studied retrospectively by Tahiri et al. in 2015 via evaluation of postoperative CT scans in 30 patients with turribrachycephaly and/or increased ICP treated with PVDO. Of patients with patent lambdoid sutures prior to PVDO, 89.5% developed postoperative lambdoid suture closure; of patients with patent sagittal sutures prior to PVDO, 41.2% developed postoperative sagittal suture closure. By physical exam, no patients developed phenotypic changes associated with lambdoid (occipital flattening, occipitomastoid bulge) or sagittal (scaphocephaly) synostosis. No variables (age, sex, underlying diagnosis, distance of distraction) were found to correlate with new-onset craniosynostosis. Ultimately, based upon the absence of significant morphologic changes, it was determined that new-onset cranial suture fusion likely does not bear significant volumetric consequences in these patients [22]. Nonetheless, this radiologic finding and its potential consequences on cranial morphology or volume require additional study in the future.

Perioperative outcomes after posterior vault distraction

A systematic review by Grieves et al. summated complications after PVDO and identified an overall perioperative complication rate of 30% [23]. Complications and their rate of occurrence are summarized in Table 3 and include CSF leak/dural injury (9.8%), pin infection/wound dehiscence/device exposure (6.9%), device failure (5.8%), intraoperative sinus bleed (2.3%), lambdoid suture separation during distraction (2.3%), and external trauma to distractor device (2.3%) [23]. No deaths or major long-term morbidities were identified in this review [23]. A notable consideration not discussed by Grieves et al. is the presence of complications in patients with ventriculoperitoneal (VP) shunts undergoing PVDO, who may be at particularly increased risk for shunt malfunction given the possibility of shunt exposure during the initial

Table 3 Complications of posterior vault distraction osteogenesis

Complication	Incidence
Dural injury/CSF leak [23]	9.8%
Device exposure [23]	6.9%
Device failure [23]	5.8%
Dural sinus injury/bleeding [23]	2.3%
External device trauma [23]	2.3%
Lambdoid suture separation [23]	2.3%
VP shunt complications [24]	35.7%

distraction operation or distractor removal, and the possible location of the shunt device on the transport segment, potentially predisposing to catheter malposition. Azzolini et al. retrospectively compared patients with VP shunts undergoing PVDO versus conventional posterior vault remodeling and identified significantly greater frequency of shunt complications in the PVDO group including prolonged CSF leak, shunt infection or malfunction, wound infection, and readmission [24]. Overall, 35.7% of patients with a VP shunt undergoing PVDO developed a shunt-related complication [24].

Comparison to conventional posterior vault remodeling

The perioperative morbidity of PVDO has been directly compared to the morbidity of conventional posterior vault expansion [25]. In this context, intraoperative and postoperative major complications, blood loss and transfusion, total operative time, ICU stay, and hospital stay were found to be similar between the two operations [25].

Varying age groups of syndromic and non-syndromic patients

Perioperative characteristics and morbidity of PVDO (including operative time, anesthesia time, blood loss, transfusions volume, ICU stay duration, and all major/minor complications) has been found to be similar across syndromic and non-syndromic craniosynostosis patients, suggesting similar safety profiles across multiple varying populations subject to PVDO [5]. Similarly, when postoperative complications were compared between older and younger patients undergoing PVDO, major perioperative morbidity rates were found to be similar [26].

Treatment algorithms utilizing PVDO

Given the ability of PVDO to safely provide robust intracranial volume expansion, improve frontal and occipital cranial morphology, and delay and/or decrease the need for frontal craniofacial surgery until an age at which relapse is less severe

and/or less likely, the procedure has become first-line treatment for syndromic craniosynostosis at the Children's Hospital of Philadelphia for approximately the past 10 years [1, 20] (Fig. 1). In this setting, at initial evaluation, patients are studied for the presence of increased intracranial pressure, ocular exposure, and airway obstruction. If no findings are present that necessitate early intervention such as regional craniectomy, tarsorrhaphy, or tracheostomy, PVDO is typically performed as the first operation between 3 and 9 months of age [20]. Following PVDO, patients are monitored for midfacial and frontal growth and clinical findings that suggest readiness or necessity for frontal surgery. The specific frontofacial operation(s) performed and the timing of such an operation are determined on an individual basis, and an algorithmic approach to this decision-making process has been published elsewhere [1]. As mentioned previously, since this algorithm has been implemented, the time to first fronto-orbital operation has increased and the number of total fronto-orbital operations in the first 5 years of life has decreased [20].

Conclusions

Over the past decade, PVDO has transformed algorithms for care of patients with many forms of craniosynostosis, and most significantly syndromic craniosynostosis. PVDO provides relatively large increases in intracranial volume with simultaneous favorable alterations in anterior and posterior cranial morphology and is safe and effective in a wide age range of patients with syndromic or non-syndromic craniosynostosis. Ongoing study of the functional benefits of posterior vault expansion will help us tailor its utilization, including the possibility of treating diagnoses other than craniosynostosis. Enthusiasm for PVDO must be weighed against relatively high complication rates, and patients may benefit from efforts to mitigate risks from transcutaneous hardware.

Author contribution Both authors (ARC, JAT) contributed equally to the drafting of the manuscript, critical revision of the manuscript, final approval of the version to be published, and agreement to be accountable for all aspects of the work.

Declarations

Conflict of interest The authors have no financial support, funding, conflicts of interest, or off-label use of pharmaceuticals to disclose.

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