A systematic review and meta-analysis of endoscopic versus open treatment of craniosynostosis. Part 1: the sagittal suture

Han Yan, MD,¹ Taylor J. Abel, MD,² Naif M. Alotaibi, MD, MSc,¹ Melanie Anderson, MLIS,³ Toba N. Niazi, MD,⁴ Alexander G. Weil, MD,⁵ Aria Fallah, MD, MSc,⁶ John H. Phillips, MD, MA,⁷ Christopher R. Forrest, MD, MSc,⁷ Abhaya V. Kulkarni, MD, PhD,^{1,2} James M. Drake, MBBCh, MSc,^{1,2} and George M. Ibrahim, MD, PhD^{1,2}

¹Division of Neurosurgery, Department of Surgery, University of Toronto; ²Division of Neurosurgery, The Hospital for Sick Children, Toronto; ³Library and Information Services, University Health Network, University of Toronto, Ontario, Canada; ⁴Division of Neurosurgery, Nicklaus Children's Hospital, University of Miami Miller School of Medicine, Miami, Florida; ⁵Division of Neurosurgery, CCHU-Ste-Justine Children's, Montreal, Quebec, Canada; ⁶Department of Neurosurgery, Mattel Children's Hospital, David Geffen School of Medicine at University of California Los Angeles, California; and ⁷Division of Plastic and Reconstructive Surgery, Department of Surgery, University of Toronto, Ontario, Canada

OBJECTIVE In this systematic review and meta-analysis the authors aimed to directly compare open surgical and endoscope-assisted techniques for the treatment of sagittal craniosynostosis, focusing on the outcomes of blood loss, transfusion rate, length of stay, operating time, complication rate, cost, and cosmetic outcome.

METHODS A literature search was performed in compliance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines. Relevant articles were identified from 3 electronic databases (MEDLINE, EMBASE, and CENTRAL [Cochrane Central Register of Controlled Trials]) from their inception to August 2017. The quality of methodology and bias risk were assessed using the Effective Public Health Practice Project Quality Assessment Tool for Quantitative Studies. Effect estimates between groups were calculated as standardized mean differences with 95% CIs. Random and fixed effects models were used to estimate the overall effect.

RESULTS Of 316 screened records, 10 met the inclusion criteria, of which 3 were included in the meta-analysis. These studies reported on 303 patients treated endoscopically and 385 patients treated with open surgery. Endoscopic surgery was associated with lower estimated blood loss ($p < 0.001$), shorter length of stay ($p < 0.001$), and shorter operating time ($p < 0.001$). From the literature review of the 10 studies, transfusion rates for endoscopic procedures were consistently lower, with significant differences in 4 of 6 studies; the cost was lower, with differences ranging from \$11,603 to \$31,744 in 3 of 3 studies; and the cosmetic outcomes were equivocal ($p > 0.05$) in 3 of 3 studies. Finally, endoscopic techniques demonstrated complication rates similar to or lower than those of open surgery in 8 of 8 studies.

CONCLUSIONS Endoscopic procedures are associated with lower estimated blood loss, operating time, and days in hospital. Future long-term prospective registries may establish advantages with respect to complications and cost, with equivalent cosmetic outcomes. Larger studies evaluating patient- or parent-reported satisfaction and optimal timing of intervention as well as heterogeneity in outcomes are indicated.

<https://thejns.org/doi/abs/10.3171/2018.4.PEDS17729>

KEYWORDS craniosynostosis; scaphocephaly; endoscopic; sagittal suture; craniofacial

THE incidence of craniosynostosis is estimated to be 3.0–6.4 per 10,000 live births; the majority of the cases are sporadic and a minority occur as part of a syndrome.^{6,24,29} There is a predominance of isolated sagittal synostosis or scaphocephaly, which accounts for more

than half of all reported craniosynostosis cases and is estimated to occur at a rate of 1.0–2.5 in 5000 live births.^{10,24} Indications for surgical management of sagittal synostosis focus heavily on correcting dysmorphism (scaphocephaly) and the remote possibility of resolving elevated intracra-

ABBREVIATIONS EPHPP = Effective Public Health Practice Project; LOS = length of hospital stay; PRISMA = Preferred Reporting Items for Systematic Reviews and Meta-Analyses; SMD = standardized mean difference.

SUBMITTED January 29, 2018. **ACCEPTED** April 3, 2018.

INCLUDE WHEN CITING Published online July 6, 2018; DOI: 10.3171/2018.4.PEDS17729.

FIG. 1. PRISMA flowchart for systematic review.

nial pressure. It has been hypothesized that untreated sagittal craniosynostosis may lead to early speech and language problems and subsequent literacy issues, and to problems in related functions such as working memory, attention, and planning.²⁰ Such adverse outcomes are the result of the elongation of the skull affecting the dorsolateral prefrontal cortex.

Infants with sagittal synostosis can be managed surgically with either traditional open surgery or the more contemporary technique of minimally invasive endoscope-assisted craniectomy followed by helmet therapy. As detailed in Bir et al.,³ Lannelongue²⁶ and Lane²⁵ described the earliest treatment of sagittal craniosynostosis relying on suturectomies, and since then the treatment options for sagittal craniosynostosis greatly overshadow those for any other type of craniosynostosis.¹⁰ Open surgery historically includes a large range of techniques—from strip craniectomy, to the pi procedure, to occipital reduction–biparietal widening, which are each associated with longer operating time, longer hospital stay, and greater blood loss.³⁴ Recognizing these suboptimal outcomes, Jimenez

and colleagues^{18,19} proposed the novel technique of using endoscopy to perform sagittal suturectomy, followed by 4–6 months of helmet therapy to prevent anteroposterior growth. This technique is offered during the first 6 months of life, optimizing cosmetic and functional results.

Despite numerous adopters of the endoscopic technique, the clinical efficacy of the endoscopic method as it compares to traditional open techniques remains debated. Thus, in this systematic meta-analysis we aimed to directly compare open surgical and endoscope-assisted techniques for the treatment of sagittal craniosynostosis, focusing on the outcomes of blood loss, transfusion rate, length of hospital stay (LOS), operating time, complications, cost, and cosmetic outcome.

Methods

Search Strategy

This systematic review and meta-analysis was conducted according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines

and recommendations.^{30,40} A literature search was performed on MEDLINE, EMBASE, and CENTRAL (Cochrane Central Register of Controlled Trials) on August 17, 2017, by a librarian (M.A.). The database searches used keywords (individually and/or in combination) such as “craniosynostosis,” “endoscopic,” and “neuroendoscope” with the appropriate subject headings. The reference lists of retrieved articles were reviewed to identify additional relevant articles. An additional search was conducted prior to publication for inclusion of newly published studies.

Study Selection

Retrieved studies were systematically assessed using inclusion and exclusion criteria by 2 reviewers (H.Y. and T.J.A.). Inclusion criteria were 1) diagnosis of sagittal craniosynostosis by a plastic surgeon or neurosurgeon; 2) at least 50% of the patient population had isolated sagittal craniosynostosis; 3) cohorts were divided into open surgery and endoscopically assisted surgery; and 4) follow-up outcomes were measured at 12 months or more. Exclusion criteria for the meta-analysis included 1) lack of quantitative comparison between open surgery and endoscopically assisted surgery; 2) nonhuman subjects; 3) inclusion of patients with syndromic craniosynostosis; and 4) editorials, abstracts, review articles, case reports, and dissertations. When duplicate studies were found, only the most complete reports were included for quantitative assessment. Studies from the same institution and possibly the same cohort of patients were only included if different outcome variables were reported.

Data Extraction and Critical Appraisal

All data were extracted from article texts, tables, and figures. Each retrieved article was reviewed by 2 investigators independently (H.Y. and T.J.A.). Authors were contacted when there were missing or incomplete data. The quality of the methodology and the risk of bias across studies were assessed by 1 reviewer (H.Y.) using the Effective Public Health Practice Project (EPHPP) Quality Assessment Tool for Quantitative Studies.⁴³ Measured variables included selection bias, study design, confounders, blinding, data collection methods, and withdrawals and dropouts, as well as a global rating, and the tool rates each as “strong,” “moderate,” or “weak.”

Statistical Analysis

Pooled effects between groups were calculated as standardized mean difference (SMD) with 95% CIs. Inter-study heterogeneity was not applicable because there were only 2 studies for each outcome. We used both random and fixed effects models to estimate the overall effect. A p value < 0.05 was set for statistical significance. Analysis was performed on “Comprehensive Meta-analysis” software (version 3.3, BIOSTAT).

Results

Literature Search

The search strategy identified a total of 503 studies (Fig. 1). After removal of 187 duplicate studies, inclusion and exclusion criteria were applied to the titles of the remain-

TABLE 1. Exclusion of eligible studies

Authors & Year	Excluded	Explanations & Comments
Abbott et al., 2012	No	NA
Arts et al., 2018	No	NA
Bonfield et al., 2016	No	NA
Chan et al., 2013	Yes	Includes pts w/ syndromic craniosynostosis; pts w/ sagittal craniosynostosis represent 33% of cohort
Dvoracek et al., 2015	Yes	Overlapping pt population suspected
Esparza & Hinojosa, 2008	No	NA
Farber et al., 2017	Yes	Pts w/ metopic craniosynostosis only
Garber et al., 2017	No	NA
Ghenbot et al., 2015	No	NA
Ghosh et al., 2014	Yes	Overlapping pt population suspected
Han et al., 2016	No	Overlapping pt population confirmed; included for largest cohort size
Hashim et al., 2014	Yes	Analysis of neuropsychological outcomes only
Keshavarzi et al., 2009	Yes	Pts w/ metopic craniosynostosis only
Keshavarzi et al., 2010	Yes	Pts w/ sagittal craniosynostosis represent 45% of cohort
Kohan et al., 2008	Yes	Case studies of twin pts
Le et al., 2014	Yes	Overlapping pt population suspected
MacKinnon et al., 2009	Yes	Pts w/ unilat coronal craniosynostosis only; overlapping pt population & data suspected
MacKinnon et al., 2013	Yes	Pts w/ unilat coronal craniosynostosis only; overlapping pt population & data suspected
Nguyen et al., 2015	Yes	Pts w/ metopic craniosynostosis only
Nowaková et al., 2015	Yes	Translated from Czech; nonquantitative data
Rogers et al., 2015	Yes	Measured outcome is hand preference
Shah et al., 2011	No	NA
Siu et al., 2014	Yes	Case studies of 2 pts w/ unilat coronal craniosynostosis & Down syndrome
Tan et al., 2013	Yes	Pts w/ unilat coronal craniosynostosis only
Thompson et al., 2018	No	NA
Vogel et al., 2014	No	Overlapping pt population confirmed w/ authors; included for unique data
Yarbrough et al., 2014	Yes	Case series of 5 pts w/ mixed suture types
Zubovic et al., 2015	Yes	Pts w/ unilat lambdoid craniosynostosis only

NA = not applicable; pts = patients.

TABLE 2. Results of quality assessment of methodology

Authors & Year	Selection Bias	Study Design	Confounders	Blinding	Data Collection Methods	Analyses	Global Rating
Abbott et al., 2012	Moderate	Moderate	Weak	Moderate	Moderate	Weak	Weak
Arts et al., 2018	Strong	Moderate	Moderate	Moderate	Moderate	Moderate	Strong
Bonfield et al., 2016	Strong	Moderate	Weak	Moderate	Moderate	Weak	Weak
Esparza & Hinojosa, 2008	Strong	Weak	Weak	Moderate	Moderate	Weak	Weak
Garber et al., 2017	Strong	Moderate	Moderate	Moderate	Moderate	Strong	Strong
Ghenbot et al., 2015	Moderate	Moderate	Weak	Moderate	Moderate	Weak	Weak
Han et al., 2016	Strong	Moderate	Weak	Moderate	Moderate	Moderate	Moderate
Shah et al., 2011	Strong	Moderate	Weak	Moderate	Moderate	Moderate	Moderate
Thompson et al., 2018	Strong	Moderate	Strong	Moderate	Moderate	Strong	Strong
Vogel et al., 2014	Strong	Moderate	Weak	Moderate	Moderate	Moderate	Moderate

ing 316 articles. This yielded 28 studies that underwent full-text analysis, of which 15 did not meet the inclusion criteria of 1) having more than 50% of patients present with sagittal synostosis; and 2) reporting on quantitative results. Three of the studies had overlapping cohorts for which unique outcomes were not reported (Table 1). Of the 10 studies included in the qualitative analysis, only 3 reported appropriate data from nonoverlapping cohorts that could be used for meta-analysis.

Seven of the 28 full-text analysis studies originated from Washington University School of Medicine, with confirmed or potential overlapping cohorts. Therefore, for each key question only the study with the largest cohort was included in the literature review. This process resulted in 10 studies with unique cohorts for specific outcomes. There were 4 weak, 3 moderate, and 3 strong papers (Table 2).

Cohort Description

Patient demographic characteristics are outlined in Table 3. The cohort size of the meta-analysis was 688 patients, with 303 (44%) children undergoing endoscope-assisted craniostomosis surgery and 385 (56%) children undergoing open repair for craniostomosis. The age of the endoscopic cohort was younger than that of the open cohort (median 2.9, mean 3.4 months, and median 5.6, mean 15.5 months, respectively).

In the literature review, the reported age of the patients in the endoscopic and open cohorts ranged from 2.9¹¹ to 4.1⁸ months and 4.8⁴ to 15.5¹⁶ months, respectively. The proportion of male patients in the studies treated by endoscopic and open procedures ranged from 68% to 75% and 42% to 76%, respectively. The follow-up ranged from 4.2 months to 95.4 months.

Perioperative Outcomes

Estimated Blood Loss

Endoscope-assisted craniostomosis surgery has a significantly lower estimated blood loss compared to open craniotomies, based on 3 studies (SMD -1.57 ml/kg, 95% CI -1.75 to -1.39; $p < 0.001$) (Fig. 2A). From the literature review, Abbott et al.¹ similarly reported medians of 4.1 ml/kg and 59.4 ml/kg when comparing endoscopic to open procedures.

Length of Stay

Endoscope-assisted craniostomosis surgery has a significantly shorter LOS compared to open craniotomies, based on 2 studies (SMD -1.66 days, 95% CI -1.86 to -1.46; $p < 0.001$) (Fig. 2B). Thompson et al.⁴⁴ conducted a multicenter retrospective study analyzing 933 propensity score-matched children and reported a median LOS of 2 and 4 days for endoscopic and open surgery, respectively. Abbott et al.¹ also described a shorter LOS for endoscopic surgery compared to open surgery, with medians of 1 day and 3 days, respectively. A Spanish study⁸ measured mean time of hospitalization as a consequence of postoperative complications and found that endoscopic osteotomies (11.2 days) resulted in shorter stays compared to open sagittal suturectomy, open sagittal suturectomy with frontal dismantling, and total cranial vault remodeling (12.4, 11.5, and 16.2 days, respectively).

Operating Times

Endoscope-assisted craniostomosis surgery had a significantly lower operating time compared to open craniotomies, based on 2 studies (SMD -1.68 days, 95% CI -1.87 to -1.48; $p < 0.001$) (Fig. 2C). Likewise, Thompson et al.⁴⁴ and Abbott et al.¹ reported medians of 1.16 and 2 hours for endoscopic procedures and 2.16 and 6 hours for open procedures.

Transfusions

Six studies^{1,2,4,11,16,44} reported a lower need for intraoperative and postoperative transfusions for patients undergoing endoscopic procedures compared to open cranial surgery. Endoscopic procedures showed a transfusion rate ranging from 0%¹ to 26%^{2,44} of patients and open procedures ranged from 16%⁴⁴ to 100%¹ (Table 4).

Complications

Eight studies^{1,2,4,8,11,16,39,44} reported on complications of open and endoscopic treatment of sagittal synostosis (Table 5). These studies all used different metrics to analyze rates of complications at various time points, including during the operation, during the hospital stay, and at various lengths of follow-up. Although statistical analysis was not reported, 6 studies reported equal or lower rates of

complications for endoscopically treated patients. Han et al. observed complication rates at 4 time points and found no statistical difference between endoscopic and open surgery (range, $p = 0.140$ to $p = 0.921$). Most notably, 3 studies^{2,8,11} reported a lower reoperation rate for endoscopic techniques compared to open surgery.

Cosmetic Outcomes

Two studies from different patient populations compared postoperative cranial index, both of which showed no statistical difference. Abbott et al.¹ reported a postoperative cranial index of 0.77 and 0.75 for endoscopic and open procedures, respectively, with no significant difference. Likewise, Shah et al.³⁹ also demonstrated a cranial index of 0.76 and 0.77 ($p = 0.346$) for endoscopic and open procedures. Finally, Ghenbot et al.¹² looked at cranial vault volume, showing no significant difference for the endoscopic and open cohorts ($p = 0.31$), although the patients in this cohort were probably also all analyzed by Shah et al.³⁹

Cost of Treatment

Three studies looked at the cost of craniostylosis treatment. Abbott et al.¹ and Vogel et al.⁴⁵ analyzed the total cost of treating craniostylosis, including the medical cost of hospital and physician fees, orthotic cost of helmet fittings, and indirect patient costs of travel and lost work days. The median (range) of the endoscopic and open surgery cost was \$23,377 (\$20,987–\$24,977) and \$55,121 (\$44,690–\$86,313), respectively,¹ and the reported mean cost of endoscopic and open surgery was \$37,356 and \$56,990, respectively.⁴⁵ Looking solely at medical costs,¹¹ endoscopic procedures again were found to be cheaper (\$21,203) than open procedures (\$32,806).

Discussion

In this meta-analysis and literature review, we found that surgical treatment of sagittal synostosis by endoscope-assisted surgery was associated with shorter operating room times, shorter LOS, reduced rates of transfusion, and similar cosmetic results at last follow-up (as measured by cranial index), compared with open surgery. Important differences highlighted include the age at time of surgery, which was consistently younger for children undergoing endoscopic repair. The study by Esparza and Hinojosa⁸ shows the differences between North American and European treatment; despite the difficulty in performing a direct comparison, the inclusion of this study highlights how different healthcare systems will have an impact on treatment options. Although a myriad of techniques for both open and endoscopic repair have been reported, rendering comparisons difficult, our meta-analysis provides the most comprehensive synthesis of the available literature pertaining to the surgical treatment of sagittal synostosis with open and endoscopic methods.

Perioperative Outcomes

Endoscopic treatment of craniostylosis has significantly less estimated blood loss than open surgery.^{1,2,11,16} Consequently, the rate of transfusion has been found to be

TABLE 3. Demographic characteristics of patients in literature review studies

Authors & Year	City & State or Country	EPHPP Rating	Study Design	No. of Pts	% Pts w/ Sagittal Craniostylosis	No. of Pts Treated		Mean or [Median] Age (mos)*		% Male		FU (mos)	
						Endo	Open	Endo	Open	Endo	Open	Endo	Open
Abbott et al., 2012	Boston, MA	Weak	R OS	20	100	10	10	[3.0]	[13.1]	70	60	4.2	22.2
Arts et al., 2018†	Nijmegen, Netherlands	Strong	R OS	93	100	63	30	4.0 ± 1.0	10 ± 8.9	NR	NR	12	12
Bonfield et al., 2016	Vancouver, BC, Canada	Weak	R OS	99	100	28	71	[3.8]	[4.8]	NR	NR	NR	NR
Esparza & Hinojosa, 2008	Madrid, Spain	Weak	R OS	155	100	39	116	4.1	6.9	NR	NR	NR	NR
Garber et al., 2017†	Salt Lake City, UT	Strong	R OS	300	100	100	200	[2.9]	[5.6]	75	76	51.6	95.4
Ghenbot et al., 2015	St. Louis, MO	Weak	R OS	27	100	12	15	NR	NR	NR	NR	NR	NR
Han et al., 2016†	St. Louis, MO	Moderate	R OS	295	57.6	140	155	3.4 ± 1.2	15.5 ± 16.5	NR	NR	25.2	37.4
Shah et al., 2011	St. Louis, MO	Moderate	P OS	89	100	47	42	3.6	6.8	68	42	13	25
Thompson et al., 2018	Multicenter, USA	Strong	R OS	933	61.8	311	622	[3]	[5]	70	65	NR	NR
Vogel et al., 2014	St. Louis, MO	Moderate	R OS	42	100	21	21	3.1 ± 0.2	6.8 ± 0.4	NR	NR	NR	NR

Endo = endoscope-assisted surgery; FU = follow-up; NR = not recorded; OS = observational study; P = prospective; R = retrospective.

* The means are reported ± SD when the original study included the SD.

† Study included in meta-analysis.

FIG. 2. Meta-analysis of perioperative measures. Std diff in means = SMDs. Figure is available in color online only.

TABLE 4. Transfusion rates between endoscopic and open treatments of sagittal craniosynostosis

Authors & Year	Treatment	Transfusion (%)	
		Endo	Open
Abbott et al., 2012	pRBC	0*	100*
	Platelets	0	20
	FFP	0	20
	Cryoprecipitate	0	10
	Albumin	30*	90*
Arts et al., 2018	pRBC	26	81
Bonfield et al., 2016		7.1	17.0
Garber et al., 2017	Endo vs CVR	13*	83*
	Endo vs SSC	13	23
Han et al., 2016	Intraop pRBC	5.0*	96.1*
	Postop pRBC	5.0*	39.4*
Thompson et al., 2018	RBC blood products	26*	81*
	Coagulation products	3*	16*

CVR = total cranial vault reconstruction; FFP = fresh frozen plasma; pRBC = packed red blood cells; SSC = open sagittal strip craniectomy.

* $p < 0.05$.

less for endoscopically treated patients.^{1,2,4,11,16,44} Bonfield et al.⁴ summarized 37 published studies that have investigated blood transfusion rates in craniofacial procedures, although they did not report any studies that directly compared endoscopic to open procedures. Although blood transfusion safety has improved over time, infants have a greater incidence of adverse outcomes than adults (37 vs 13 cases per 100,000 red blood cells transfused, respectively).^{15,27} This is most often due to complications such as transfusion-related acute lung injury (TRALI) and transfusion-related circulatory overload (TACO). Therefore, it is important to minimize the need for transfusion when performing surgery on infants. In their multicenter study of 31 institutions, Thompson et al.⁴⁴ regretted the documented inaccuracies of blood loss estimates and sought alternatives to study this important metric.

The length of operating time and LOS for endoscopically treated patients was significantly shorter with endoscope-assisted correction of sagittal synostosis, as seen in 5 studies.^{1,8,11,16,44} Although the American studies show LOS means ranging from 1 to 4 days, Esparza and Hinojosa⁸ admit that their mean time of 11.9 days is unacceptable; their longer hospital stays seen in sagittal expansive osteotomies (12.4 days) and sagittal holocranial dismantling (16.2 days) are related to postoperative complications. This further suggests that the less invasive endoscopic techniques may reduce operating time and hospital stay.

TABLE 5. Complications in patients with sagittal craniosynostosis undergoing endoscopic and open surgery

Authors & Year	No. of Pts		Complication	Endoscopic		Open	
	Endo	Open		%	No. of Pts	%	No. of Pts
Abbott et al., 2012	10	10	Postop emergency visits	0.0	0	30.0	3
Arts et al., 2018	63	30	Intraop complications	1.6	1	3.3	1
			Postop complications	4.7	3	3.3	1
			Reoperation	1.6	1	3.3	1
Bonfield et al., 2016	28	71	Cardiovascular or wound healing	0.0	0	0.0	0
Esparza & Hinojosa, 2008	39	116	Reoperation	2.6	1	9.5	11
			Wound infection	2.6	1	0.0	0
			Subgaleal hematoma	0.0	0	5.2	6
			Infected hematoma	0.0	0	3.4	4
			Dural tear	0.0	0	0.8	1
			Craniolacunae	0.0	0	2.6	3
			Plate scarring	0.0	0	1.7	2
			Postop hyperthermia	10.3	4	15.5	18
			Infection*	1.7	4	2.3	16
Garber et al., 2017	100	200	Revision surgery	1.0	1	7.0	14
Han et al., 2016†	140	155	Surgical complications	2.1	3	1.3	2
			Intraop durotomies	3.6	5	7.8	12
			Postop complications	3.6	5	4.5	7
			Readmit <30 days	1.4	2	1.3	2
Shah et al., 2011†	47	42	Wound revision	2.1	1	2.4	1
Thompson et al., 2018	311	622	Hypotension requiring pressors	3	8	4	23
			Venous air embolism	1	4	1	5
			Hypothermia, temp <35°C	22	70	26	160
			Postop intubation	2	6	10	60
			Cardiac arrest	0	0	0.2	1

* Bacterial and/or viral infection sites include the urinary tract, respiratory system, ophthalmic system, and/or central and mechanical intravenous lines.

† Possible overlapping cohorts.

Complications

Eight studies^{1,2,4,8,11,16,39,44} seem to support an equivalent or decreased complication rate for endoscopically assisted surgeries compared to open surgery. Future research should document the most common or most severe complications at each stage of treatment to provide the opportunity for analysis across studies. None of the included studies reported a higher incidence of injury to the sagittal sinus due to endoscopic surgery compared to open surgery. Because it is a novel technique, endoscopic surgery encompasses a large variability in outcomes and complications between different surgeons and procedures.

Cosmetic Outcomes

It has not yet been clearly defined how to best measure the postoperative aesthetic improvements after craniosynostosis surgery. The average cranial index (i.e., maximal cranial width/length) of children younger than 3 years of age was measured at 0.815.³¹ In children with sagittal synostosis, the cranial index is as low as 0.65.^{14,35} Our study shows that both endoscopic and open corrections, with cranial index ranging from 0.75 to 0.77, are able to achieve comparable postoperative results.^{1,39} Ghenbot et

al.¹² analyzed cranial vault volume in addition to cranial index and found no difference in cranial vault volume, assuaging the concern that molding helmet therapy would lead to volume restriction. Future studies evaluating these techniques must take into account variability in outcomes among children as well as other measures of cosmesis, including parent or patient impression and satisfaction.

Cost of Treatment

In 3 different populations,^{1,11,45} the cost of endoscopic treatment with helmet therapy follow-up was found to be lower than the cost of open surgery, with differences ranging from \$11,603 to \$31,744. The difference is mostly attributable to the more invasive and extensive nature of open cranial surgery, often requiring ICU observation, transfusions, or a higher rate of revision. The costs of helmets were reported in all included studies; however, there are several indirect costs and also additional time needed for postoperative helmet servicing that have not been measured in the published literature. Furthermore, there may be unaccounted obstacles and unknown patient or parent impressions of helmet usage that have not been studied. The long-term efficacy and costs require ongoing research.

Strengths and Limitations

Strengths of this study include an extensive search of the current literature, strict adherence to PRISMA guidelines, and quality of evidence analysis by EPHPP protocol. Currently, there exists no randomized controlled trial comparing the surgical approaches of endoscopic and open sagittal suture repair. Given the data suggesting absence of equipoise between the two techniques, it is unlikely that such a trial would be conducted. Ultimately, the findings of this meta-analysis require confirmation by larger, multicenter studies with extended follow-up.

Only 3 studies met PRISMA criteria for full meta-analysis; this low number indicates the need for future studies. The studies included in this review consisted mostly of moderate and weak observational studies. One must also consider the selection bias due to preference of surgeons or institutions and shifts of practice over time. There have yet to be any studies that examine patient or parent satisfaction, or the required duration and comfort of helmet usage. It is also evident that there is a tradeoff between the potential benefits of endoscopic repair and the disadvantage of the length of time spent in a helmet. These compromises are mediated by parental preferences, which are difficult to capture in current literature. Future studies aimed at assessing “value-based medicine” by using methodologies such as decision-tree analyses may elucidate the benefit of endoscopic repair.

Furthermore, there is still a lack of understanding regarding the variability of endoscopic outcomes and, therefore, longer follow-up and further analyses are indicated. The current meta-analysis was additionally limited by the heterogeneity of suture types in some studies.¹⁶ Given the clinical and surgical heterogeneity that we have identified, it is of value to conduct future observational studies that separate different entities and account for patient and surgical variability. Given the dearth of data available for meta-analysis, interstudy heterogeneity could not be calculated.

Conclusions

Endoscope-assisted correction of sagittal synostosis has some benefits over open surgical repair. Endoscopic procedures are associated with lower estimated blood loss, operating time, days in hospital, and transfusion rates. Current evidence in the literature comparing endoscopic and open sagittal craniosynostosis repair is mostly of weak to moderate quality—only 3 papers were found to be strong. Although these early studies suggest several benefits of endoscopic procedures, consideration is needed for each individual family to understand the usage of helmets and unknown long-term outcomes. Future large prospective registries or randomized controlled trials are required to validate the findings of this study.

References

- Abbott MM, Rogers GF, Proctor MR, Busa K, Meara JG: Cost of treating sagittal synostosis in the first year of life. **J Craniofac Surg** 23:88–93, 2012
- Arts S, Delye H, van Lindert EJ: Intraoperative and postoperative complications in the surgical treatment of craniosynostosis: minimally invasive versus open surgical procedures. **J Neurosurg Pediatr** 21:112–118, 2018
- Bir SC, Ambekar S, Notarianni C, Nanda A: Odilon Marc Lannelongue (1840–1911) and strip craniectomy for craniosynostosis. **Neurosurg Focus** 36(4):E16, 2014
- Bonfield CM, Sharma J, Cochrane DD, Singhal A, Steinbok P: Minimizing blood transfusions in the surgical correction of craniosynostosis: a 10-year single-center experience. **Childs Nerv Syst** 32:143–151, 2016
- Chan JW, Stewart CL, Stalder MW, St Hilaire H, McBride L, Moses MH: Endoscope-assisted versus open repair of craniosynostosis: a comparison of perioperative cost and risk. **J Craniofac Surg** 24:170–174, 2013
- Di Rocco F, Arnaud E, Renier D: Evolution in the frequency of nonsyndromic craniosynostosis. **J Neurosurg Pediatr** 4:21–25, 2009
- Dvoracek LA, Skolnick GB, Nguyen DC, Naidoo SD, Smyth MD, Woo AS, et al: Comparison of traditional versus normative cephalic index in patients with sagittal synostosis: measure of scaphocephaly and postoperative outcome. **Plast Reconstr Surg** 136:541–548, 2015
- Esparza J, Hinojosa J: Complications in the surgical treatment of craniosynostosis and craniofacial syndromes: apropos of 306 transcranial procedures. **Childs Nerv Syst** 24:1421–1430, 2008
- Farber SJ, Nguyen DC, Skolnick GB, Naidoo SD, Smyth MD, Patel KB: Anthropometric outcome measures in patients with metopic craniosynostosis. **J Craniofac Surg** 28:713–716, 2017
- Fearon JA, McLaughlin EB, Kolar JC: Sagittal craniosynostosis: surgical outcomes and long-term growth. **Plast Reconstr Surg** 117:532–541, 2006
- Garber ST, Karsy M, Kestle JRW, Siddiqi F, Spanos SP, Rivacambrian J: Comparing outcomes and cost of 3 surgical treatments for sagittal synostosis: a retrospective study including procedure-related cost analysis. **Neurosurgery** 81:680–687, 2017
- Ghenbot RG, Patel KB, Skolnick GB, Naidoo SD, Smyth MD, Woo AS: Effects of open and endoscopic surgery on skull growth and calvarial vault volumes in sagittal synostosis. **J Craniofac Surg** 26:161–164, 2015
- Ghosh TD, Skolnick G, Nguyen DC, Sun H, Patel K, Smyth MD, et al: Calvarial thickness and diploic space development in children with sagittal synostosis as assessed by computed tomography. **J Craniofac Surg** 25:1050–1055, 2014
- Gociman B, Marengo J, Ying J, Kestle JR, Siddiqi F: Minimally invasive strip craniectomy for sagittal synostosis. **J Craniofac Surg** 23:825–828, 2012
- Goodnough LT: Risks of blood transfusion. **Anesthesiol Clin North America** 23:241–252, v, 2005
- Han RH, Nguyen DC, Bruck BS, Skolnick GB, Yarbrough CK, Naidoo SD, et al: Characterization of complications associated with open and endoscopic craniosynostosis surgery at a single institution. **J Neurosurg Pediatr** 17:361–370, 2016
- Hashim PW, Patel A, Yang JF, Travieso R, Terner J, Losee JE, et al: The effects of whole-vault cranioplasty versus strip craniectomy on long-term neuropsychological outcomes in sagittal craniosynostosis. **Plast Reconstr Surg** 134:491–501, 2014
- Jimenez DF, Barone CM: Endoscopic craniectomy for early surgical correction of sagittal craniosynostosis. **J Neurosurg** 88:77–81, 1998
- Jimenez DF, Barone CM, McGee ME, Cartwright CC, Baker CL: Endoscopy-assisted wide-vertex craniectomy, barrel stave osteotomies, and postoperative helmet molding therapy in the management of sagittal suture craniosynostosis. **J Neurosurg** 100 (5 Suppl Pediatrics):407–417, 2004
- Kapp-Simon KA, Speltz ML, Cunningham ML, Patel PK, Tomita T: Neurodevelopment of children with single suture craniosynostosis: a review. **Childs Nerv Syst** 23:269–281, 2007

21. Keshavarzi S, Hayden MG, Ben-Haim S, Meltzer HS, Cohen SR, Levy ML: Variations of endoscopic and open repair of metopic craniosynostosis. **J Craniofac Surg** **20**:1439–1444, 2009
22. Keshavarzi S, Meltzer H, Cohen SR, Breithaupt A, Ben-Haim S, Newman CB, et al: The risk of growing skull fractures in craniofacial patients. **Pediatr Neurosurg** **46**:193–198, 2010
23. Kohan E, Wexler A, Cahan L, Kawamoto HK, Katchikian H, Bradley JP: Sagittal synostotic twins: reverse pi procedure for scaphocephaly correction gives superior result compared to endoscopic repair followed by helmet therapy. **J Craniofac Surg** **19**:1453–1458, 2008
24. Kolar JC: An epidemiological study of nonsyndromal craniosynostoses. **J Craniofac Surg** **22**:47–49, 2011
25. Lane LC: Pioneer craniectomy for relief of mental imbecility due to premature sutural closure and microcephalus. **JAMA** **267**:230, 1992
26. Lannelongue OM: De la craniectomie chez les microcéphales, chez les enfants arriérés et chez les jeunes sujets présentant, avec ou sans crises épileptiformes, de troubles moteurs ou psychiques. **Congr Franc de Chir Paris** **5**:73–81, 1891
27. Lavioie J: Blood transfusion risks and alternative strategies in pediatric patients. **Paediatr Anaesth** **21**:14–24, 2011
28. Le MB, Patel K, Skolnick G, Naidoo S, Smyth M, Kane A, et al: Assessing long-term outcomes of open and endoscopic sagittal synostosis reconstruction using three-dimensional photography. **J Craniofac Surg** **25**:573–576, 2014
29. Lee HQ, Hutson JM, Wray AC, Lo PA, Chong DK, Holmes AD, et al: Changing epidemiology of nonsyndromic craniosynostosis and revisiting the risk factors. **J Craniofac Surg** **23**:1245–1251, 2012
30. Liberati A, Altman DG, Tetzlaff J, Mulrow C, Gøtzsche PC, Ioannidis JP, et al: The PRISMA statement for reporting systematic reviews and meta-analyses of studies that evaluate health care interventions: explanation and elaboration. **PLoS Med** **6**:e1000100, 2009
31. Likus W, Bajor G, Gruszczyńska K, Baron J, Markowski J, Machnikowska-Sokołowska M, et al: Cephalic index in the first three years of life: study of children with normal brain development based on computed tomography. **Sci World J** **2014**:502836, 2014
32. MacKinnon S, Proctor MR, Rogers GF, Meara JG, Whitecross S, Dagi LR: Improving ophthalmic outcomes in children with unilateral coronal synostosis by treatment with endoscopic strip craniectomy and helmet therapy rather than fronto-orbital advancement. **J AAPOS** **17**:259–265, 2013
33. MacKinnon S, Rogers GF, Gregas M, Proctor MR, Mulliken JB, Dagi LR: Treatment of unilateral coronal synostosis by endoscopic strip craniectomy or fronto-orbital advancement: ophthalmologic findings. **J AAPOS** **13**:155–160, 2009
34. Mehta VA, Bettegowda C, Jallo GI, Ahn ES: The evolution of surgical management for craniosynostosis. **Neurosurg Focus** **29**(6):E5, 2010
35. Murray DJ, Kelleher MO, McGillivray A, Allcutt D, Earley MJ: Sagittal synostosis: a review of 53 cases of sagittal suturectomy in one unit. **J Plast Reconstr Aesthet Surg** **60**:991–997, 2007
36. Nguyen DC, Patel KB, Skolnick GB, Naidoo SD, Huang AH, Smyth MD, et al: Are endoscopic and open treatments of metopic synostosis equivalent in treating trigonocephaly and hypotelorism? **J Craniofac Surg** **26**:129–134, 2015
37. Nowaková M, Kordoš P, Hladík M, Medfická H, Rosický J, Kaleta E, et al: Endoskopické operační řešení kraniosynostózy z pohledu dětského intensivisty. **Přehledové články** **16**:308–311, 2015
38. Rogers GF, Wood BC, Amdur RL, Jeelani Y, Reddy S, Proctor MR, et al: Treatment type is associated with population hand preferences in patients with unilateral coronal synostosis: implications for functional cerebral lateralization. **Plast Reconstr Surg** **136**:782e–788e, 2015
39. Shah MN, Kane AA, Petersen JD, Woo AS, Naidoo SD, Smyth MD: Endoscopically assisted versus open repair of sagittal craniosynostosis: the St. Louis Children's Hospital experience. **J Neurosurg Pediatr** **8**:165–170, 2011
40. Shamseer L, Moher D, Clarke M, Ghera D, Liberati A, Petticrew M, et al: Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) 2015: elaboration and explanation. **BMJ** **350**:g7647, 2015
41. Siu A, Rogers GF, Myseros JS, Khalsa SS, Keating RF, Magge SN: Unilateral coronal craniosynostosis and Down syndrome. **J Neurosurg Pediatr** **13**:568–571, 2014
42. Tan SP, Proctor MR, Mulliken JB, Rogers GF: Early fronto-facial symmetry after correction of unilateral coronal synostosis: frontoorbital advancement vs endoscopic strip craniectomy and helmet therapy. **J Craniofac Surg** **24**:1190–1194, 2013
43. Thomas BH, Ciliska D, Dobbins M, Micucci S: A process for systematically reviewing the literature: providing the research evidence for public health nursing interventions. **Worldviews Evid Based Nurs** **1**:176–184, 2004
44. Thompson DR, Zurakowski D, Haberkern CM, Stricker PA, Meier PM, Bannister C, et al: Endoscopic versus open repair for craniosynostosis in infants using propensity score matching to compare outcomes: a multicenter study from the Pediatric Craniofacial Collaborative Group. **Anesth Analg** **126**:968–975, 2018
45. Vogel TW, Woo AS, Kane AA, Patel KB, Naidoo SD, Smyth MD: A comparison of costs associated with endoscope-assisted craniectomy versus open cranial vault repair for infants with sagittal synostosis. **J Neurosurg Pediatr** **13**:324–331, 2014
46. Yarbrough CK, Smyth MD, Holekamp TF, Ranalli NJ, Huang AH, Patel KB, et al: Delayed synostoses of uninvolved sutures after surgical treatment of nonsyndromic craniosynostosis. **J Craniofac Surg** **25**:119–123, 2014
47. Zubovic E, Woo AS, Skolnick GB, Naidoo SD, Smyth MD, Patel KB: Cranial base and posterior cranial vault asymmetry after open and endoscopic repair of isolated lambdoid craniosynostosis. **J Craniofac Surg** **26**:1568–1573, 2015

Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Yan, Ibrahim. Acquisition of data: Yan, Abel, Anderson. Analysis and interpretation of data: Yan, Abel. Drafting the article: Yan, Abel, Ibrahim. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Yan. Statistical analysis: Alotaibi. Administrative/technical/material support: Ibrahim. Study supervision: Yan, Ibrahim.

Supplemental Information

Companion Papers

Yan H, Abel TJ, Alotaibi NM, Anderson M, Niazi TN, Weil AG, et al: A systematic review of endoscopic versus open treatment of craniosynostosis. Part 2: the nonsagittal single sutures. DOI: 10.3171/2018.4.PEDS17730.

Correspondence

Han Yan: The Hospital for Sick Children, Toronto, ON, Canada. hhan.yan@mail.utoronto.ca.