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Short- and Long-Term Outcomes By Procedure Type for Non-Sagittal Single-Suture Craniosynostosis

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Abstract

Background: Minimally invasive approaches for the treatment of single-suture craniosynostosis are sometimes touted as equivalent to cranial vault reconstruction. While techniques for sagittal synostosis have been reviewed previously, evidence regarding open and less invasive surgical techniques for metopic, coronal, and lambdoid synostosis has yet to be reviewed.

Methods: Systematic searches were performed using [Embase.com](https://www.embase.com) and PubMed. Included studies reported short- or long-term outcomes, compared at least two standard techniques, discussed single-suture coronal, metopic, or lambdoid craniosynostosis, and enrolled at least 20 study participants. Two authors screened titles and abstracts, and also performed full text review and data extraction. Given heterogeneous outcomes, qualitative synthesis was performed after data extraction.

Results: The search strategy yielded 2,348 articles. Of these, 313 were removed as duplicates, and 1,935 were excluded during title/abstract review. After full text review of 100 articles, 19 were selected for data extraction. The heterogeneity of outcomes precluded meta-analysis and required qualitative synthesis. While short-term outcomes indicated decreased morbidity of minimally invasive techniques, only two articles presented long-term reoperation rates. One study reported higher reoperation rates in the less invasive technique, and the second reported no reoperations in the median follow-up period of 33 months.

Conclusions: Studies comparing long-term outcomes between different surgical techniques for single-suture craniosynostosis remain deficient. The development of standardized outcome measures is essential, and prospective, multicenter studies are necessary to assess the long-term efficacy of these procedures.

Keywords

craniosynostosis[MeSH Terms]; “reconstructive surgical procedures” [MeSH Terms]; cranial vault reconstruction; strip craniectomy; spring-assisted cranioplasty

INTRODUCTION

Craniosynostosis is defined by the premature fusion of any of the cranial sutures, affecting as many as 1 in 1700 children every year [1]. Patients with craniosynostosis develop abnormal head morphologies secondary to Virchow's law [2], and are also at increased risk for intracranial hypertension and developmental delays [3–7]. Operative intervention is the only treatment for patients with significant deformity, and open techniques alone fully address both hypoplastic and compensatory abnormalities simultaneously [8].

In recent years, less invasive procedures have been revived or newly developed for the management of craniosynostosis. Strip craniectomy was originally described in the 1940s [9], but was later abandoned due to poorer aesthetic results compared with open techniques [10, 11]. The operation was more recently described in conjunction with extended postoperative helmet therapy by Jimenez et al. in 1999 [12]. Other surgeons have promoted techniques like spring-mediated cranioplasty and distraction osteogenesis [13–17]. These operations are less invasive and rely on expansion of the calvarial vault over time [18–20]. Surgeons who promote these techniques assert equivalent aesthetic outcomes with significantly less morbidity at the time of surgery [21–23]. A recent systematic review and meta-analysis by Gerety et al. showed that strip craniectomy and spring-mediated cranioplasty may be equivalent to cranial vault reconstruction (CVR) in sagittal synostosis, although long-term studies are still needed [24]. However, studies comparing long-term outcomes between minimally invasive and open techniques in other types of craniosynostosis are noticeably absent. As such, we hypothesized that the craniofacial literature would provide short-term outcomes data from single centers utilizing one technique, but that relatively few studies would compare techniques or provide long-term results in this patient population.

To test our hypothesis and assess the current state of the literature on this topic, we performed a systematic review to identify studies comparing minimally invasive and open techniques in patients with metopic, unicoronal, and lambdoid synostosis. When possible, outcomes data were pooled in an effort to develop more robust recommendations for appropriate surgical interventions in single-suture craniosynostosis. Based upon our review, we also offer suggestions for potential avenues of future research.

MATERIALS AND METHODS

Search Strategy

We performed comprehensive searches on PubMed and [Embase.com](https://www.embase.com) on October 16, 2017. Each search consisted of a combination of controlled terms (MeSH in PubMed, and Emtree in Embase), as well as title and abstract keywords for *craniosynostosis*, *cranial vault reconstruction*, *strip craniectomy*, *spring-assisted cranioplasty*, and *endoscopic craniectomy* concepts. Duplicates were removed in Endnote X6. The reproducible searches for all databases are available in Table 1.

Selection Criteria

Two authors (KB & GH) independently screened all titles and abstracts. For inclusion, studies had to 1) relate to short- or long-term outcomes, 2) compare at least two standard techniques, 3) discuss single-suture coronal, metopic, or lambdoid craniosynostosis (sagittal was excluded), and 4) include more than 20 patients in the study. Included studies could not select patients by outcomes. All case reports, technique papers, and reviews were excluded. Studies meeting criteria underwent full-text review. The same criteria were used for inclusion at this stage. Disagreements at both stages were resolved through discussion between the two screening authors.

Data Extraction

Two authors (KB & GH) reviewed the final included papers for relevant study details. Author names, title, study design, study quality, number of study participants, demographics, complications, and outcomes data were recorded on an Excel spreadsheet. Any disagreements between the two authors were resolved by discussion during the extraction phase. Given the extremely heterogeneous study outcomes, qualitative synthesis was performed after extraction.

RESULTS

Description of Studies

The combined PubMed and Embase search strategies yielded a total of 2,348 articles after duplicate removal in EndNote X6 (Figure 1). Manual removal of duplicates yielded 2,035 articles. Both title and abstract review was performed using the selection criteria as mentioned in the methods section. After review and resolution of remaining conflicts, 100 articles remained. After full text review and discussion, 19 articles were selected for inclusion and data extraction (Table 2) [18, 25–42].

Of the 19 included studies, 4 were prospective [25, 26, 30, 33], and the rest were retrospective. Three of the prospective studies utilized a multicenter design [25, 30, 33]. Fourteen studies enrolled fewer than 100 patients, and eight studies enrolled fewer than 50 (Table 2). Quality of evidence as measured by the Grading of Recommendations Assessment, Development, and Evaluation (GRADE) system was “low” or “very low” for all papers except for one, which was deemed “moderate” [25].

Twelve studies compared endoscopic techniques with open techniques, while the other seven compared an open technique with a “microscopic” technique, spring-mediated cranioplasty, distraction osteogenesis, or other minimally invasive techniques. Patients with all types of synostosis were included in eleven studies, whereas four studies focused on metopic synostosis patients, and four concentrated on unicoronal synostosis patients. The remaining studies enrolled varying combinations of synostosis patients. All studies provided short-term outcomes data, but only two studies provided long-term outcomes data (defined as more than two years of follow up). In these two studies, reoperation was the long-term outcome measured, but specific length of follow-up was not required for inclusion in the study. In the first study, the mean follow-up was 7.4 years for the less invasive technique and 7.2 years for

the open technique; the range of follow up in the second study for both groups was 16–45 months (Table 2).

Studies examining all types of synostosis

Four studies examined multiple types of synostosis and compared endoscopic techniques with open techniques [25–27, 29]. All four presented transfusion data. In three studies, the rate of RBC product given was significantly lower in the endoscopic strip craniectomy (ESC) group [25–27]. In the fourth study, the rate of RBC product given was higher for CVR, but statistical analyses were not performed [27]. Both length of stay (LOS) and surgical duration were significantly reduced in the ESC groups in three of the four studies [25, 27, 29]. Two studies reported complications, although the types of complications between the two studies were heterogenous [25, 27].

One study compared microscopic with open techniques [28]. In the microscopic technique, three incisions are made, and suturectomy is performed with a microscope [43]. Mean follow up in this study was greater than seven years for both groups, and the authors reported higher long-term reoperation rates for patients who underwent microscopic techniques, especially in metopic and coronal synostosis patients. Of the nine patients who required reoperation after the microscopic technique, five required total cranial vault reconstruction, despite postoperative helmeting [28]. Another study compared fronto-orbital advancement (FOA,) CVR, and spring cranioplasty [30]; two studies compared minimally invasive techniques with open techniques [31, 33]; another compared FOA to “other techniques” [32]; and another compared distraction osteogenesis (DO) with open cranioplasty [34]. Given the variety of compared techniques and the heterogeneity of outcomes (Table 2), these results could not be compared.

Studies examining metopic synostosis

Three studies compared endoscopic and open techniques in metopic synostosis [35–37], and a fourth study compared FOA with endoscopic techniques of varying degrees of invasiveness [38]. Two studies comparing endoscopic and open techniques came from the same group, and it was found that the postoperative interfrontal angle improved equally in both groups, and the angles were also not significantly different from those of controls [35]. The same result was found in the second study, which also found no differences between groups in the interzygomaticofrontal distances (ZF-ZF), the zygomatofrontal glabellar (ZF-G-ZF) angles, and the intercanthal distances (ID) [36]. The third study similarly reported equivalent interzygomaticofrontal distances and interdacryon distances between groups [37]. However, the authors of these studies did not demonstrate a correlation between these measurements and clinically relevant outcomes, such as aesthetics or neurologic function. The remaining outcomes of the three studies were heterogenous. The outcomes of the fourth study included descriptive data on estimated blood loss (EBL), transfusion volume, surgical time, and LOS, all of which tended to be lower in the endoscopic group, but no statistical analysis was performed [38].

Studies examining unicoronal synostosis

All four studies with unicoronal synostosis patients compared endoscopic and open techniques [18, 39–41]. The first two compared temporal measurements and frontofacial symmetry between endoscopic and open groups, demonstrating equivalent temporal outcomes in the two groups and superior facial symmetry in the endoscopic group [18, 39]. The final two studies from the same group compared ophthalmic outcomes between endoscopic and open techniques, finding greater variance in anisotigmatism in the open group [40]. Strabismus surgery was more frequent in the open group in the second study [40] but equivalent in the first [41]. Other ophthalmic outcomes tended to be worse in the open groups in both studies (Table 2).

DISCUSSION

After reviewing the available literature from two separate databases, we found only 19 articles that compared at least two surgical techniques for the treatment of craniosynostosis. Furthermore, these 19 articles did not all compare the same techniques, and they also did not report on the same type of craniosynostosis. Finally, the outcome measures were extremely variable between papers. More specifically, long-term outcomes were lacking, with only two studies reporting reoperation rates. Additionally, length of follow up was not used as an exclusion criterion in either study, resulting in reoperation rates that may reflect procedures performed shortly after the index procedure, or many years later. Due to the heterogeneity of both techniques and outcomes, a meta-analysis could not be performed, and the long-term efficacy of these procedures could not be assessed.

Several less invasive procedures for the treatment of craniosynostosis have been described in recent decades. Endoscopic strip craniectomy, in conjunction with postoperative molding therapy, was touted as an aesthetically viable alternative to open CVR with significantly less operative morbidity [12]. It was initially described in patients with sagittal synostosis and performed at a much younger age, which presumably prevents the worsening of calvarial dysmorphology as the child grows [44]. Since then, the indications for endoscopic strip craniectomy have extended to other types of synostosis, even syndromic bilateral coronal synostosis [45–47]. Ridgway et al. reported long-term maintenance of calvarial shape in patients with sagittal synostosis, although mean follow up in the study was just over 2 years [48]. To add further confusion, the results by Hashim et al. indicate potentially worse long-term neuropsychological outcomes in patients who undergo endoscopic strip craniectomy [49]. Another less invasive technique, spring-mediated cranioplasty, has also been described and developed over the last two decades [20, 50]. Advocates of spring-mediated cranioplasty cite its reduced perioperative morbidity compared with CVR, as well as its ability to facilitate a dynamic reconstruction that maintains itself with the child's growth [51]. This technique was also originally described in patients with sagittal synostosis [50], but has been performed more recently in patients with metopic and coronal craniosynostosis as well [52–54]. Long-term outcomes suggest promising results, but are compromised by single-center designs and no comparison group [55, 56]. Distraction osteogenesis, a commonly described technique for syndromic craniosynostosis [57, 58], has also been used for single-suture synostosis [59–61]. However, the few studies with more long-term results are underpowered

and lack control groups [62–64]. Another less invasive option is the microscopic technique, previously described by Teichgraeber et al., which involves the use of three incisions and a microscope for suturectomy [43]. Based on long-term outcomes data, the authors now only recommend the technique for sagittal and lambdoid synostosis, given the high rate of complications in metopic and unicoronal patients [28]. Finally, while “minimally invasive technique” is often used as a broad term to refer to any technique that is less invasive than CVR, the minimally invasive technique as described by Maugans et al. refers to a specific surgical procedure. It also requires three incisions, but suturectomy is performed using cartilage scissors under direct visualization. This study reported no long-term results [31].

Before the reintroduction of strip craniectomy and the description of other less invasive techniques, CVR was considered the gold standard for the treatment of craniosynostosis [65, 66]. Unlike less invasive techniques, CVR allows for the immediate correction of calvarial shape at the time of surgery, without relying on any postoperative molding or additional procedures [8]. However, the operation is considerably morbid, in that blood loss is high, transfusions are frequently required, and patients are admitted to the intensive care unit postoperatively [25]. Hospital length of stay is routinely longer than for less invasive techniques, and cost is higher, even when helmeting therapy and follow up are considered [29]. However, the procedure gives the surgeon control over the final postoperative head shape, allowing the surgeon to address both the hypoplastic areas as well as the areas subject to compensatory growth [8]. Long-term results in some centers are favorable [65, 67, 68], but others report significantly high rates of reoperation [69–71]. This variation is likely secondary to technical differences in the performance of the procedure.

While a shorter, less invasive operation is certainly less morbid in the short term, the question of long-term efficacy remains unanswered, especially in the era of postoperative orthotic molding. Strip craniectomies were previously abandoned, as these procedures could not achieve equivalent aesthetic results [11]. The critical modification in recent decades is the addition of helmet therapy. Of the 16 papers that reported using postoperative helmeting, only six included length of helmeting protocols, with five of these lasting until one year of age [27–29, 36, 37, 39]. This places a considerable burden on families and requires a high level of compliance, which may not be achieved despite the best intentions. While the burden may be an acceptable tradeoff for some families, we still have no high-quality studies that compare long-term outcomes between open techniques and less invasive techniques with helmeting. Future investigation is especially important, in light of anecdotally higher frequencies of complete re-do CVRs after endoscopic strip craniectomy compared with primary CVR [72].

Furthermore, we have also failed to establish a standardized, reproducible outcome measure. While reoperation rates are easy to measure, reoperation rates may reflect variation in surgeon preferences and shared decision-making much more than they reflect recapitulation of deformity [68]. To determine which operations maintain normal calvarial shape long-term, we need prospective, multicenter data with standardized, measurable outcomes that adequately capture aesthetics. These outcomes should also include patient and family-reported outcomes, since it is the patient and the family who live with the aesthetic result.

Additionally, the exact nature of “minimally invasive procedures” must be defined and agreed upon by investigators, such that we know exactly what is being compared to CVR.

Our review was characterized by several limitations. Given the heterogenous nature of both the procedure types studied and outcomes measured, it is impossible to make any definitive conclusions regarding the safest and most efficacious procedure for the correction of non-sagittal, single suture craniosynostosis. Additionally, the review was subject to publication bias, as negative findings related to any procedure type are often not published. However, the goal of the review was to summarize the current state of the literature surrounding this topic, and it reveals substantial deficiency with regard to prior methodology and lacks consistency in reported outcomes.

In conclusion, short-term outcomes indicate that less invasive techniques may be superior to CVR for the treatment of non-sagittal, single-suture craniosynostosis, but measured outcomes were of such a heterogenous nature as to preclude data pooling for meta-analysis. Currently, there are no studies with long-term follow up as inclusion criteria that compare long-term outcomes between procedure types. Moving forward, multicenter studies should be undertaken to adequately determine the safety and long-term efficacy of these procedures. However, measurable and meaningful outcomes must first be established, and “minimally invasive procedures” must be adequately defined before such an undertaking is possible.

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Figure 1.
PRISMA 2009 flow diagram.

Table 1.

Search strategies.

PubMed	((spring-assisted[tiab] OR spring-mediated[tiab] OR strip craniectomy[tiab] OR endoscopic craniectomy[tiab] OR endoscope-assisted[tiab] OR cranial vault reconstruction[tiab] OR cranial vault remodeling[tiab] OR fronto-orbital advancement[tiab] OR fronto-orbital advancement[tiab] OR (Craniosynostoses[mesh]) OR (Craniosynostoses/surgery[mh]) AND english[la] NOT (animals[mh] NOT humans[mh]))
Embase	((spring-assisted:ti,ab OR spring-mediated:ti,ab OR strip craniectomy:ti,ab OR endoscopic craniectomy:ti,ab OR endoscope-assisted:ti,ab OR cranial vault reconstruction:ti,ab OR cranial vault remodeling:ti,ab OR fronto-orbital advancement:ti,ab OR fronto-orbital advancement:ti,ab) AND (craniosynostosis:ti,ab OR synostosis:ti,ab OR craniosynostoses:ti,ab OR synostoses:ti,ab OR "synostosis"/exp OR "craniofacial synostosis"/exp OR "complex craniosynostosis"/exp OR "cranial suture"/exp) OR ("fronto-orbital advancement"/exp OR "cranial vault reconstruction surgery"/exp OR "cranial vault reconstruction"/exp OR "spring assisted cranial vault expansion"/exp OR "spring assisted cranioplasty"/exp OR "endoscopic strip craniectomy"/exp) AND (english/[lim] AND [humans]/lim)

Table 2.

Summary of included full text articles.

Reference	Study design	Study participants	GRADE ^d of evidence	Techniques compared	Synostosis type(s)	Short Term Outcomes	Long Term Outcomes	Results
<i>All types of synostosis</i>								
Thompson et al., 2017	Prospective, multicenter cohort	933	Moderate	ESC ^b , open	Sagittal, metopic, unicoronal, bicoronal, lambdoid, multiple sutures, unknown	Transfusion data, coagulation products, intraoperative blood loss, use of antifibrinolytics, postoperative intubations, length of ICU ^c stay, hospital LOS ^d , anesthesia time, surgical time, outcomes by case volume, complications	None	Reduced blood products, blood donor exposure, ICU stay, hospital LOS, anesthesia time, surgical duration in ESC group; higher incidence of postop intubation in open group
Meier et al., 2016	Prospective, single-center cohort	118	Low	ESC, open	All types	Transfusion data	None	CVR ^e associated with transfusion
Han et al., 2016	Retrospective cohort	295	Low	ESC, open	Sagittal, metopic, unicoronal, bicoronal, lambdoid, squamosal, multisuture	EBL ^f , EBV ^g , RBC ^h product given, postop transfusions, steroids, TXA ⁱ , surgical complications, durotomies, medical complications, 30-day readmission rate, reoperation rates, LOS, surgical time,	None	Reduced EBL, EBV, RBC products, postop transfusions, steroid use, TXA use, LOS, and surgical time in ESC; surgical complications, medical complications, durotomies, and 30-day readmission were equivalent between groups; reoperation 2.1% in ESC and 6.5% in open
Teichgraber et al., 2014	Retrospective cohort	180	Low	Microscopic, open	All types	Reoperation, complications	Reoperation rates	Reoperation rate 14.9% for micro, 7.1% for open; no complications in non-sagittal patients undergoing micro operation; major complications in 13.4% micro; minor complications in 5.3% of open, major complications in 1.8% of open
Chan et al., 2013	Retrospective cohort	57	Low	ESC, FOA ^j	All types	EBL, EBV, rate RBCs given, transfusion volume, LOS, surgical time, cost	None	Reduced EBL, EBV, rate of RBCs given, transfusion volume, LOS, surgical time, cost in ESC group
Howe et al., 2012	Prospective multicenter cohort	127	Low	FOA, CVR, spring mediated cranioplasty	All types	EBV, RBC product, blood donor exposure, invasive monitoring, surgical complications, surgical transfusions by case volume	None	Reduced EBV, RBC product, blood donor exposure, invasive monitoring, surgical complications, surgical time, and transfusions by case volume in spring group, but no statistical analysis performed

Reference	Study design	Study participants	GRADE ^a of evidence	Techniques compared	Synostosis type(s)	Short Term Outcomes	Long Term Outcomes	Results
Maugans et al., 2011	Retrospective single center cohort	56	Low	MI, open	All types	EBL, rate RBCs given, transfusion volume by receipt of TXA	None	TXA reduced EBL in MI group but not open group
Goyal et al., 2011	Retrospective single center cohort	95	Very low	FOA, other techniques	All types	LOS, EBL	None	Increased LOS and EBL associated with FOA, compared with "other" techniques
Kim et al., 2008	Prospective multicenter cohort	47	Low	MI, open	All types	Parenting stress index	None	Lower total stress score in MI group
Akai et al., 2006	Retrospective cohort	24	Very low	DO, open	All types	EBL, rate of RBC product given, complications, surgical time	None	Reduced EBL and rate of RBC product given in DO; reduced surgical time in DO but required two additional operations
Park et al., 2010	Retrospective single center cohort	38	Low	RDO, cranioplasty and remodeling	Unicoronal, sagittal, lambdoid, microcephaly	Transfusion volume, complications, reoperation, anesthesia time, surgical time, distraction data	Reoperation	Transfusion volume equivalent; four complications in RDO and two in CR; no reoperations in median follow up period (33 months); anesthesia and surgical time reduced in RDO
<i>Metopic</i>								
Furber et al., 2017	Retrospective cohort	36	Very low	ESC, FOA	Metopic	Frontal width, interfrontal angle	None	Postop interfrontal angle equivalent between ESC and FOA
Nguyen et al., 2014	Retrospective cohort	56	Very low	ESC, open	Metopic	ZF distance, ZF-G-ZF ^P angle, interfrontal angle, intercanthal distance	None	Postop interfrontal angle, ZF-ZF distance, ZF-G-ZF angle, intercanthal distance equivalent between ESC and open groups
Huang et al., 2013	Retrospective cohort	30	Very low	ESC, open	Metopic	ZF distance, interdacryon distance, FT-G-FT angle	None	Postop ZF distance, interdacryon distance, and FT-G-FT angles equivalent between groups
Keshavarzi et al., 2009	Retrospective cohort	33	Low	Endoscopic (suturectomy alone, suturectomy +FOA, or suturectomy + cranial reconstruction), FOA	Metopic	EBL, transfusion volume, LOS, surgical time	None	Reduced EBL, transfusion volume, LOS, surgical time with endoscopic techniques, but no statistical analysis performed
<i>Unicoronal</i>								
Masserano et al., 2018	Retrospective cohort	66	Low	ESC, open	Unicoronal	Posterior temporal width, anterior temporal width, orbital width, anterior cranial fossa area	None	Procedure type had no effect on symmetric improvement of any measurement
Tan et al., 2013	Retrospective cohort	22	Very low	ESC, open	Unicoronal	Facial deviation, nasal tip deviation, middle facial depth, lower facial depth, supraorbital projection	None	Reduced facial deviation, nasal tip deviation, middle facial depth in ESC group

Reference	Study design	Study participants	GRADE ^a of evidence	Techniques compared	Synostosis type(s)	Short Term Outcomes	Long Term Outcomes	Results
Mackinnon et al., 2013	Retrospective cohort	43	Very low	ESC, FOA	Unicoronal	Anisostigmatism, overelevation in adduction, ocular head tilt, strabismus surgery, amblyopia	None	Standard deviation of anisostigmatism greater for FOA (determined by bootstrapping); ocular head tilt, strabismus surgery, and amblyopia increased in FOA
Mackinnon et al., 2009	Retrospective cohort	37	Very low	ESC, FOA	Unicoronal	Anisostigmatism, strabismus surgery, severity of V pattern strabismus, exocyclotorsion	None	Standard deviation of anisostigmatism greater for FOA (determined by bootstrapping); no difference in strabismus surgery; greater severity of V pattern strabismus in FOA; more frequent postop exocyclotorsion in FOA

^aGrading of Recommendations Assessment, Development, and Evaluation

^bEndoscopic strip craniectomy

^cIntensive care unit

^dLength of stay

^eCranial vault reconstruction

^fEstimated blood loss

^gEstimated blood volume

^hRed blood cells

ⁱTranexamic acid

^jFronto-orbital advancement

^kMinimally invasive

^lDistraction osteogenesis

^mRotating distraction osteogenesis

ⁿCranial reconstruction

^oInterzygomaticofrontal distance

^pZygomaticofrontal suture glabellar angle

Frontotemporal angle

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