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FOCUS SESSION

Cranioplasty after craniectomy in pediatric patients—a systematic review

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Abstract

Introduction Complications following cranioplasty with either autografts or cranial implants are commonly reported in pediatric patients. However, data regarding cranioplasty strategies, complications and long-term outcomes are not well described. This study systematically reviews the literature for an overview of current cranioplasty practice in children.

Methods A systematic review of articles published from inception to July 2018 was performed. Studies were included if they reported the specific use of cranioplasty materials following craniectomy in patients younger than 18 years of age, and had a minimum follow-up of at least 1 year.

Results Twenty-four manuscripts, describing a total of 864 cranioplasty procedures, met the inclusion criteria. The age of patients in this aggregate ranged from 1 month to 20 years and the weighted average was 8.0 years. The follow-up ranged from 0.4 months to 18 years and had a weighted average of 40.4 months. Autologous bone grafts were used in 484 cases (56.0%). Resorption, infection and/or hydrocephalus were the most frequently mentioned complications. In this aggregate group, 61 patients needed a revision cranioplasty. However, in 6/13 (46%) papers studying autologous cranioplasties, no data was provided on resorption, infection and revision cranioplasty rates. Cranial implants were used in 380 cases (44.0%), with custom-made porous hydroxyapatite being the most commonly used material (100/380, 26.3%). Infection and migration/fracturing/loosening were the most frequently documented complications. Eleven revision cranioplasties were reported. Again, no data was reported on infection and revision cranioplasty rates, in 7/16 (44%) and 9/16 (56%) of papers, respectively.

Conclusion Our systematic review illuminates that whether autografts or cranial implants are used, postcranioplasty complications are quite common. Beyond this, the existing literature does not contain well documented and comparable outcome parameters, suggesting that prospective, long-term multicenter cohort studies are needed to be able to optimize cranioplasty strategies in children who will undergo cranioplasty following craniectomy.

Keywords Craniectomy · Cranioplasty · Children · Pediatric neurosurgery

Introduction

Craniectomy is a commonly performed procedure in adult and pediatric patients, mainly suffering from medically refractory elevated intracranial pressure (ICP) in the setting of traumatic brain injury (TBI), vascular pathologies, or various other conditions [6, 45]. With advances in operative and perioperative care, more craniectomy patients survive their initial insult and eventually require subsequent cranioplasty procedures [6, 47].

Cranioplasty is necessary to restore cosmesis and to protect the underlying dura and brain from physical insult. Furthermore, restoration of normal cranial vault geometry contributes to neurological recovery by reversing the abnormalities in cerebral blood flow, cerebrospinal fluid hydrodynamics, and metabolic activity that result from craniectomy [47].

In adults, numerous materials have been used for cranioplasty to repair the cranial defect. Cranial vault reconstruction with the original autologous bone flap has gained

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support over time and is favored for its strength and elasticity, biocompatibility, durability, general accessibility, resistance to infection, and natural recovery of its feature to maintain the cranial contour [7, 22, 45, 46]. In cases with destruction or loss of the original calvarium (particularly for re-do cranioplasty after initial infection), synthetic materials are still frequently used for reconstruction. Methyl methacrylate is the most extensively used cranial implant material in patients above 18 years of age [3], but there are many alternatives such as glass-fiber-reinforced composite, titanium-based plates and mesh, hydroxyapatite, acrylic compounds (polymethyl methacrylate), ceramic compounds (the calcium phosphate-based cements), and plastic polymers such as porous polyethylene [22, 41, 42].

In pediatric patients, the skull is still growing and the bony contour is dynamic, which places specific constraints on materials that can be used for cranial vault reconstruction [15]. Also, thinner scalp and a better long-term survival prognosis further restrict usable bone replacements [10, 15]. Cranioplasty materials to be used in children should be able to grow with the skull, be durable for a long period of time and have a high resistance to late infection [13].

The most commonly used cranioplasty materials in adults do not meet these pediatric criteria [15]. Therefore, autologous bone grafts such as particulate bone graft, split calvarial bone graft, as well as extracranial bone sources, have been historically favored for children over synthetic materials such as acrylic or metals [2, 13, 22, 28, 30, 34, 38, 43]. These autologous grafts have their own set of complications, the most common of which is bone resorption, with reported rates ranging from 21 to over 80% [6, 27, 34, 44]. Autologous bone can be difficult to mold, and the limited amount that can be harvested may not sufficiently cover the whole cranial defect [22].

When autograft fails, or is not available or chosen as initial reconstruction method, cranial implant cranioplasty has been reported to be up to 90% efficacious [6], with success attributed to shapeable fit with the cranial contour and unlimited amount. However, these cranial implants may carry a risk of infection and immunological reactions, contamination, pathologic ossification, and fragmentation [22, 36].

While there are several retrospective studies assessing complications of cranioplasty in adults [2, 7, 15, 24, 36, 42, 47], similar pediatric cranioplasty studies remain scarce—primarily focused on the influence of bone flap size, timing of cranioplasty, and patient age on the durability of the bone flap replacement [17, 34, 40, 49]. Although new methods and techniques for the use of cranioplasty in pediatric patients have recently emerged, most manuscripts have only discussed one particular cranioplasty material at a time [8–10, 16, 19, 20, 23, 28, 37, 39, 41, 51, 52]. It is hard to draw significant conclusions from these studies since long-term outcomes are lacking and only small sample sizes were used [8–10, 16, 20, 39, 41, 51, 52]. This systematic review of cranioplasties in the pediatric population aims to synthesize these smaller studies into a coherent aggregate for a better understanding of cranioplasty strategies, complications, and possible long-term outcomes in children.

Methods

This review was performed according to the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) guidelines, an evidence-based set of criteria for reporting in systematic reviews and meta-analyses [35].

Search strategy

To capture as many relevant publications as possible, an elaborate search of the electronic medical databases of PubMed (1950 to 2018), Embase (1981 to 2018), and Medline (1981 to 2018) was performed.

These databases were searched using the key words "cranioplasty" and "children" in conjunction with either "long-term," "materials," or "craniectomy". The reference sections of included studies were also examined to identify relevant papers. Of this total number of studies ($n = 655$), duplicates were removed. Searches were limited to the English language and to clinical studies (Fig. 1).

Study selection

The remaining records ($n = 393$) were screened for "cranioplasty," "decompressive craniectomy," and "materials" in their titles or abstracts. After screening of the citations on these key words, the abstracts of articles of interest were then reviewed in detail by using inclusion and exclusion criteria, specified a priori as follows:

- Pediatric patients were defined as those younger than 18 years of age.
- The minimal follow-up of these patients needed to be more than 1 year.
- To be included, the papers had to share results of the use of specific cranioplasty materials after decompressive craniectomy in pediatric patients. Cranioplasty materials included autologous bone and/or one or more synthetic material(s).
- Retrospective analyses, cohort studies, and clinical trials were all deemed eligible for inclusion.
- Individual case reports and systematic reviews (with no original data) were excluded from this review. When the papers contained results of the use of cranioplasty for treatment of any form of craniostenosis or craniosynostosis, they were also excluded from the review.

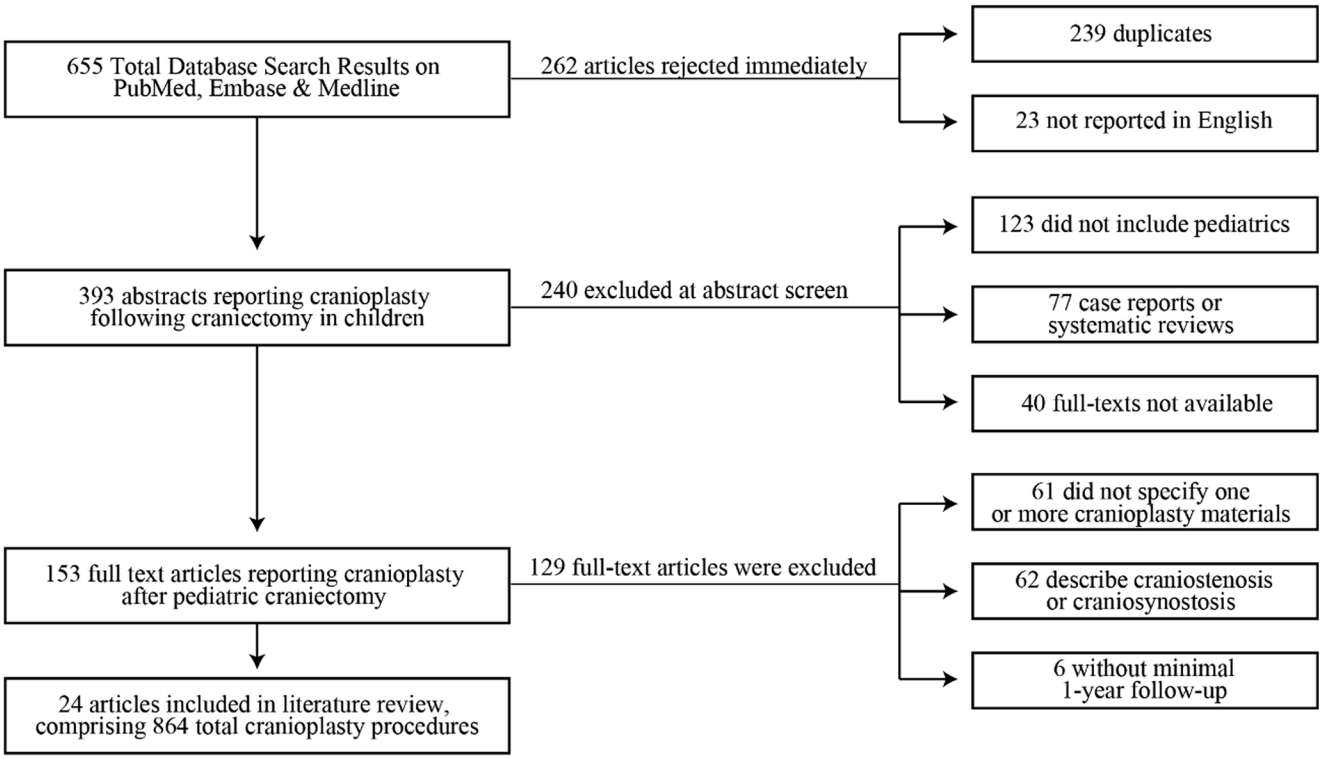

Fig. 1 Flowchart representing results from the PubMed, Embase, and Medline databases

Of the 393 studies, 240 could be excluded after reading the abstract alone, and another 129 could be excluded after reading the full text (illustrated in Fig. 1). Twenty-four articles remained from this screening process, involving 864 cranioplasty procedures, and these were analyzed [5, 6, 8–10, 13, 16, 17, 19, 20, 22, 27–29, 33, 34, 39–41, 43, 48, 50–52].

Statistical analysis was performed using IBM SPSS Statistics, version 25 [IBM Corp., Armonk, NY, USA].

Results

Twenty-four manuscripts describing outcomes and complications following cranioplasty in the pediatric population after decompressive craniectomy met search criteria for this study (Table 1). Several articles also reported information about time until bone flap replacement, patient age, or other potential risk factors [6, 17, 27, 40]. Twenty-one of the 24 included articles met CEBM criteria for level 4 as case series [5, 6, 8–10, 13, 16, 17, 22, 27–29, 33, 39–41, 43, 48, 50–52], and the remaining studies met criteria for level 3 as either a controlled cohort study or post-marketing surveillance studies [19, 20, 34]. Three multicenter studies [19, 20, 33] and one prospective study of eight cases [41] were included. The remainder were all retrospective, single-center studies. All studies collectively described 864 total cranioplasty procedures.

The age of all included patients in the aggregate ranged from 1 month to 20 years [6, 8–10, 13, 16, 17, 19, 20, 22, 27–29, 33, 34, 39–41, 43, 48, 50–52] and the weighted average was 8.0 years [5, 6, 8–10, 16, 19, 20, 22, 27–29, 39, 40, 43, 48, 50–52].

The duration of follow-up ranged from 0.4 months to 18 years [6, 8–10, 13, 16, 20, 22, 27, 28, 33, 34, 39–41, 43, 48, 51] and had a weighted average of 40.4 months [5, 6, 8–10, 16, 19, 20, 22, 27, 28, 33, 34, 39–41, 43, 48, 50, 51].

Autologous bone grafts were used in 484 cases (56.0%) [6, 13, 17, 22, 27–29, 33, 34, 40, 43, 48, 50] and cranial implants in 380 cases (44.0%) [5, 8–10, 16, 19, 20, 22, 29, 33, 34, 39, 41, 50–52].

Autologous cranioplasty

Autologous cranioplasty was used in 484 cases (56.0%, Table 2). Time until bone flap replacement was reported in 220 (45.5%) of these, ranging from 1 week to 17 months [6, 22, 27, 34, 40, 50] with a weighted average of 4.7 months [6, 22, 27, 40, 50]. The follow-up period ranged from 0.4 to 216 months [6, 13, 22, 27, 28, 33, 34, 40, 43, 48] and had a weighted average of 43.9 months [6, 22, 27, 28, 33, 34, 40, 43, 48, 50].

An autologous bone flap replacement using the original bone flap was used in 359 of 484 cases (74.2%) [6, 17, 27, 29, 33, 34, 40, 43, 50]. In the remaining autologous grafts, 70 split calvarial bone grafts (14.4%) [13, 22, 29, 33], 42

Table 1 Included pediatric cranioplasty studies

Author and year	CEBM level of evidence	Age range, min-max (mean age) in months	Follow-up range, min-max (mean follow-up) in months	Number of autografts	Number of cranial implants	Total cranioplasties	Resorption (%)	Infection (%)		Need for replacement (%)	
								Auto	Implant	Auto	Implant
Edwards et al. [13], 1987	4	12–204 (−)	8–84 (−)	19	0	19	1 (5.3)	–	N/A	–	N/A
Posnick et al. [43], 1993	4	12–198 (100.8)	12–66 (31.4)	27	0	27	2 (7.4)	0	N/A	0	N/A
Choi et al. [9], 1998	4	4–228 (72)	1–43 (26)	0	19	19	N/A	N/A	1 (5.3)	N/A	–
Taggard et al. [48], 2001	4	11–240 (76.8)	2–48 (27)	13	0	13	–	–	N/A	–	N/A
Cohen et al. [10], 2004	4	11–216 (88.8)	3–60 (24.8)	0	34	34	N/A	N/A	0	N/A	–
Grant et al. [27], 2004	4	1.4–228 (111.6)	6–72 (57.6)	40	0	40	20 (50)	0	N/A	20 (50)	N/A
Pang et al. [39], 2005	4	24–114 (66)	24–50.4 (34.8)	0	15	15	N/A	N/A	0	N/A	–
Josan et al. [29], 2005	4	1.4–180 (97.2)	–	24	4	28	–	3 (12.5)	–	3 (12.5)	–
Greene et al. [28], 2008	4	36–240 (96)	6–216 (73.2)	38	0	38	–	1 (2.6)	N/A	1 (2.6)	N/A
Chao et al. [8], 2009	4	25.2–58.8 (43.2)	13.4–41.8 (29.3)	0	13	13	N/A	N/A	–	N/A	–
Piedra et al. [40], 2012	4	32.8–195.1 (112.9)	2–124 (24)	61	0	61	18 (29.5)	4 (6.6)	N/A	–	N/A
Frassanito et al. [17], 2012	4	1–216 (−)	–	23	0	23	4 (17.4)	–	N/A	1 (4.4)	N/A
Bowers et al. [6], 2013	4	6–192 (74.4)	1.5–168 (38)	54	0	54	27 (50)	9 (16.7)	N/A	26 (48)	N/A
Martin et al. [34], 2014	3	1–204 (−)	18–130 (83)	23	4	27	8 (34.8)	2 (8.7)	–	10 (44)	0
Piittulainen et al. [41], 2015	4	30–192 (−)	22–53 (35.1)	0	8	8	N/A	N/A	2 (25)	N/A	2 (25)
Bowers et al. [5], 2015	4	− (98.4)	− (33.3)	0	69	69	N/A	N/A	9 (13)	N/A	–
Frassanito et al. [20], 2015	3	30–72 (58.8)	13–80 (40.1)	0	24	24	N/A	N/A	2 (8.3)	N/A	3 (12.5)
Williams et al. [51], 2016	4	74–213 (153)	15–105 (55)	0	22	22	N/A	N/A	–	N/A	0
Fu et al. [22], 2016	4	12–228 (88.8)	1.2–108 (27.9)	11	30	41	–	–	0	–	0
Fiaschi et al. [16], 2016	4	20–155 (84.3)	24–96 (55.7)	0	12	12	N/A	N/A	–	N/A	1 (8.3)
Waqas et al. [50], 2017	4	24–144 (85.2)	− (18.2)	31	5	36	–	–	–	–	–
Zaccaria et al. [52], 2017	4	84–168 (115.2)	–	0	6	6	N/A	N/A	–	N/A	–
Ma et al. [33], 2018	4	19–216 (−)	0.4–81.6 (48)	120	39	159	–	–	2 (5.1)	–	–
Frassanito et al. [19], 2018	3	84–160.8 (120.4)	− (36.1)	0	76	76	N/A	N/A	4 (5.3)	N/A	5 (6.6)
Total		1–240 (96.0)	0.4–216 (40.4)	484	380	864	80	19	20	61	11

CEBM, Centre for Evidence-Based Medicine; −, not reported; *N/A*, not applicable

All studies were single-institute studies, except for Frassanito et al. 2015 and 2018 and Ma et al.

All studies were retrospective reviews, except for Martin et al. (controlled cohort study) and Frassanito et al. 2015 and 2018 (post-marketing surveillance studies)

particulate bone grafts (8.7%) [22, 28], and 13 rib grafts (2.7%) [48] were used. In six out of 13 (46%) papers that studied autologous grafts, no data on resorption, infection, and revision cranioplasty rates were reported.

Reported complications were associated with 32.6% of all autologous cranioplasties. Out of the 484 autologous cases, 80 (16.5%) documented resorption as a complication [6, 13, 17, 27, 34, 40, 43], with infection in 19 (3.9%) [6, 27–29, 34, 40, 43], and hydrocephalus in 35 (7.2%) [6, 17, 27, 34, 40]. Five patients did not achieve an acceptable cosmetic outcome (1.0%) [22, 29, 43] and three had problems with migration, fracturing, or loosening (0.6%) [17]. Seven patients developed subdural hygroma (1.4%) [17, 34]. Other postcranioplasty outcomes included existence of residual bone defects (0.6%), shunt infections (0.4%), behavioral issues (0.2%), sterile wound dehiscence (0.2%), seroanguinous sterile wound discharge (0.2%), and non-ossification of the graft (0.2%) [22, 28, 29, 34]. Sixty-one patients (12.6% of autologous grafts) required revision cranioplasty [6, 17, 27–29, 34, 43].

Cranial implants

Cranial implants were used in 380 cases (44.0%, Table 2). The interval between craniectomy and cranioplasty was reported in 51 of these, ranging from 15 days to 12.6 months [34, 50] with a weighted average of 31.1 months [16, 22, 50]. The duration of follow-up ranged from 0.4 to 130 months [8–10, 16, 20, 22, 33, 34, 39, 41, 51] and had a weighted average of 36.6 months [5, 8–10, 16, 19, 20, 22, 33, 34, 39, 41, 50, 51].

Custom-made porous hydroxyapatite (CustomBone Service) was the most common used cranial implant, used in 100 cases (26.3%) [19, 20]. Titanium was used for 60 cases (15.8%) [22, 29, 33, 50, 51], polymethyl methacrylate/polyhydroxyethyl methacrylate for 41 cases (10.8%) [5], hydroxyapatite for 40 cases (10.5) [9, 39, 52], polyetheretherketone for 36 cases (9.5%) [5, 22, 33], carbonated apatite cement for 34 cases (8.9%) [10], polymethyl methacrylate for 31 cases (8.2%) [16, 22, 34], demineralized bone matrix for 13 cases (3.4%) [8], porous polyethylene (MedPor) for 9 cases (2.4%) [22], glass-fiber reinforced composite in 8 cases (2.1%) [41], and methyl methacrylate for 7 cases (1.8%) [29, 50]. One cranioplasty was performed with an unspecified cranial implant (0.3%) [22].

From the 16 papers that addressed cranial implants, seven of those (44%) did not report on infection rate and nine (56%) papers failed to provide data on revision cranioplasty rates.

The overall reported complication rate in the cranial implant group was 14.2%. Documented complications were infection in 20 cases (5.3%) [5, 9, 19, 20, 33, 41], migration, fracturing, or loosening in 10 cases (2.6%) [16, 19, 20] and cerebrospinal fluid leakage in 2 cases (0.5%) [5, 41]. Furthermore, behavioral issues were reported in 2 cases (0.5%), headaches in 5 cases (1.3%), and increased sensitivity of the ear and lateral rectus muscle impingement both in 1 case (0.3% each) [22]. The authors did not report which of their various cranial implants let to the occurrence of these complications.

Other postcranioplasty outcomes included existence of residual bone defects (0.5%), postoperative implant adjustments (0.5%), subgaleal hematomas (0.5%), recurrent subgaleal fluid collections (0.3%), seroma (0.3%), intracranial hypotension (0.3%), EEG alterations (0.3%), granuloma formation (0.3%), left central retinal occlusion (0.3%), and skin retraction (0.3%) [5, 8, 9, 16, 19, 33, 51, 52]. Eleven patients (2.9% of cranial implants) required revision cranioplasty [16, 19, 20, 41].

Five studies [22, 29, 33, 34, 50] compared autologous bone to synthetic material cranioplasty. The remainder only included patients who had all undergone cranioplasty using either an autologous bone graft or a cranial implant.

Fu et al. documented a complication rate of 27.3% (3/11) in the autologous group and a 30.0% (9/30) complication rate in the cranial implant group [22].

Josan et al. documented an autologous complication rate of 20.8% (5/24) and an cranial implant complication rate of zero [29].

Ma et al. did not report any complications in the autologous group or in the group of patients with a synthetic PEEK implant, since there was a primary focus on outcomes following titanium cranioplasty. A complication rate of 9.1% (3/33) was documented in the titanium group [33].

Martin et al. documented an autologous complication rate of 73.9% (17/23). No complications were reported in the cranial implant group [34].

Waqas et al. reported on the major postcranioplasty complications (infection, cosmetic problems, wound dehiscence, and cerebrospinal fluid leak) noted in their cohort, but did not specify which material caused which complication. However, there was no significant difference in complication rates between autologous or cranial implant cranioplasty [50].

In this systematic review, an infectious complication was reported in 19 out of 484 cases in the autologous group and in 20 out of 380 cases in the cranial implant group, a difference that did not reach statistical significance ($p = 0.56$).

There was a need for cranioplasty replacement in 61 out of 484 autologous cases and 11 out of 380 cranial implant cases, but the difference was not statistically significant ($p = 0.12$).

Discussion

In the reviewed literature of pediatric cranioplasties, autologous cranioplasty was used in 484 cases (56.0%) in which a complication rate of 32.6% was experienced in a weighted mean follow-up period of 43.9 months.

Table 2 Summary of reported postcranioplasty complications

Implant material	Number of cranioplasties (%)	Postcranioplasty complications							
		Resorption (%)	Infection (%)	Hydrocephalus (%)	Cosmetic problems (%)	Migration/ fracturing/ loosening (%)	CSF leakage/ subdural hygroma (%)	Other (%)	Need for replacement (%)
Original bone flap	359 (74.2)	79	18	35	4	3	7	3	60
Split calvarial	70 (14.4)	1	–	–	–▲	–	–	1▲	–
Particulate bone	42 (8.7)	–	1	–	–▲	–	–	3▲	1
Rib grafts	13 (2.7)	–	–	–	–	–	–	–	–
Total for autografts	484	80 (16.5)	19 (3.9)	35 (7.2)	4 + 1 (1.0)▲	3 (0.6)	7 (1.4)	7 + 2 (1.9)▲	61 (12.6)
Methyl methacrylate	7 (1.8)	N/A	–	–	–	–	–	–	–
Polymethyl methacrylate	31 (8.2)	N/A	–	–	–	2	–	3■	1
PMMA/PHEMA	41 (10.8)	N/A	5	–	–	–	1	2	–
Hydroxyapatite	40 (10.5)	N/A	1	–	–	–	–	2	–
Custom-made porous hydroxyapatite	100 (26.3)	N/A	6	–	–	8	–	2	8
Carbonated apatite cement + PLA plates	34 (8.9)	N/A	0	–	–	0	–	–	–
DBX + resorbable mesh bilaminate	13 (3.4)	N/A	–	–	0	0	–	2	–
Polyetheretherketone	36 (9.5)	N/A	4	–	–	–	–	–■	–
Porous polyethylene	9 (2.4)	N/A	0	–	–	–	–	–■	0
Titanium	60 (15.8)	N/A	2	–	–	–	–	2■	–
Glass-fiber reinforced composite	8 (2.1)	N/A	2	–	0	0	1	0	2
Unspecified	1 (0.3)	N/A	0	–	–	–	–	–■	0
Total for cranial implants	380	N/A	20 (5.3)	–	–	10 (2.6)	2 (0.5)	13 + 9 (5.8)■	11 (2.9)

–, not reported; *N/A*, not applicable; *PMMA/PHEMA*, polymethyl methacrylate/polyhydroxyethyl methacrylate; *PLA*, polylactic acid; *DBX*, demineralized bone matrix

▲ Fu et al. did not report whether a split calvarial bone graft or a particulate bone graft led to cosmetic problems in one case and other complications in two cases

■ Fu et al. did not report which of these cranial implants let to the occurrence of other complications in nine cases

Cranial implants were used in 380 cases (44.0%) with a total complication rate of 14.2% in a weighted mean follow-up period of 36.6 months.

Autologous cranioplasty

Autologous bone graft has been generally advocated as the gold standard of calvarial reconstruction in pediatric patients, due to its ability to remodel and become integrated with the growing pediatric skull over time [30].

However, it is not without its problems. As shown, the main complication following autologous cranioplasty is bone flap resorption (80/484, 16.5%). This resorption rate is reasonably high, but lower than what might have been expected from (individually) reported resorption rates in the literature, ranging from 21 to over 80% [6, 27, 34, 44]. This difference might be due to the lack of age stratification in the reported literature, since young age is considered to be a risk factor for bone flap resorption [6, 17, 18, 34, 40, 44]. However, not all included studies inquired the correlation between young age and the risk of bone flap resorption [13, 27, 43].

In addition, a considerable number of papers that studied autologous grafts did not report data on resorption (6/13, 46%). This missing data could also have contributed to the relatively low resorption rate.

Other, less-common, complications include infection and hydrocephalus, unacceptable cosmetic outcome, or a mechanical risk of migration, fracture, or loosening of the bone flap.

Autologous cranial bone is often available only in limited quantities and the harvest of extracranial sources of bone such as rib can be complicated by pain, pneumothorax, and chest wall deformity [31]. When an autograft is used for bone flap replacement, it is more likely that surgical time, wound size, and amount of soft tissue dissection will increase [4, 22, 29].

Cranial implants

Over time, effort has been devoted to the development of bone substitutes that can become integrated with the growing pediatric skull [1, 4, 16, 32, 39, 41, 52].

In adults, methyl methacrylate has been generally accepted as the preferred material for bone flap replacement. However, synthetic material is less frequently used in the pediatric population because of concern for growth restriction and/or implant instability.

It is believed that, because it is non-expansible, methyl methacrylate does not remodel to and integrate with the pediatric cranium. Thus, it has been suggested that this material should be avoided in pediatric patients less than 5 years of age [29]. However, Fu et al. found that there was no clinical evidence of growth restriction when cranial implants (such as methyl methacrylate) were used for pediatric cranioplasty [22].

Other synthetic implant materials include titanium plates and meshes, hydroxyapatite, porous polyethylene, polyetheretherketone, and glass-fiber reinforced composite. Although titanium is a strong material and provides resistance to infection, it is difficult to contour to a child's natural skull shape, and it creates large artifacts in CT scans [1, 27, 29].

Hydroxyapatite is the principal component in the bone and is thus considered as a near-ideal bone substitute [12, 14, 26]. However, the hand-made bone cement appears to be unsuitable for the growing pediatric skull, can experience loss of structural integrity when exposed to cerebrospinal fluid or blood, and has high rates of infection and erosive material exposure [11, 12, 14, 26, 27, 36]. To overcome these problems, custom-made porous hydroxyapatite implants have been developed. These implants showed few complications and appeared to be an effective cranial reconstruction material in pediatric patients [19–21].

Porous polyethylene has been used extensively in adult cranial reconstruction since it theoretically has the capacity for fibrous ingrowth and revascularization [22]. Long-term data on the use of porous polyethylene in reconstructing pediatric calvarial defects are lacking, although one study reported successful early outcomes without documentation of growth disturbance [32]. Inert, polyetheretherketone implants, are durable and useful when a large defect with a complex contour has to be restored; they are easily removable should a recurrence of intracranial access be needed, but lack any osteoconductive or inductive properties [22, 25].

Early cranioplasty outcomes with the glass fiber-reinforced composite were found to be promising. Piittulainen et al. reported two patients that had their implant removed due to an underlying surgical site infection (out of 8 implants), but after adequate treatment, good and safe outcomes were achieved, both functionally and in terms of cosmetic appearance of the patients [41].

The results of this systematic review show that there is a substantial lack of equal and uniform outcome parameters regarding potential postcranioplasty complications following either autologous or cranial implant cranioplasty in the pediatric population.

Five years ago, Rocque et al. [45] performed a systematic review on potential risk factors that could predispose pediatric cranioplasty patients to postoperative complications and also concluded there is a significant lack of uniform data regarding this important issue. Since then, 12 additional papers appeared reviewing the outcomes of pediatric patients undergoing cranial reconstruction [5, 16, 19, 20, 22, 33, 34, 41, 44, 50–52]. Although all of them have contributed valuable data on this topic, it remains difficult to draw significant conclusions that could definitely guide clinical practice.

One of these papers concerns a recently published study [44]. This manuscript did not meet the inclusion criteria for this review (since it did not specifically list the used implant materials), but it does have pertinent results.

This North-American multicenter retrospective study presented data on a large cohort of 359 patients to determine risk factors for resorption and infection after pediatric cranioplasty [44]. In this cohort, an infection rate of 10.5% was reported. Bone resorption occurred in 21.7% out of the 240 patients who underwent autologous cranioplasty.

Important risk factors regarding perioperative management that led to these postcranioplasty complications were assessed. Young age and the use of external ventricular drains and lumbar shunts were important predictors of bone resorption, while ventilator dependence and the presence of a VP shunt or gastrostomy were significant risk factors for infection. The material used for cranioplasty was not found to be associated with infection. However, it remains unclear how cranioplasty material may affect more long-term complication risks like growth restriction, fracturing, loosening, and the onset of late infection.

It is essential to obtain a better understanding of more long-term outcomes following cranioplasty in the pediatric population since potential complications like a lack of osteointegration or insufficient growth capacity could occur after a long period of time. We would encourage that all patients should be monitored and should continue clinical reassessment regularly at least during the period of skull growth, but preferably over a longer period of time. Follow-up visits should include assessment of wound healing, stability or migration, ossification or resorption of the graft, as well as symmetry and skull shape. While migration, ossification, or resorption can be assessed by CT-scans, medical (2D and/or 3D) photography, sufficient cosmetic outcome can be determined by parents or patients via surveys.

This follow-up should be significantly longer than the existing follow-up duration of the most recent paper [44] and of the papers included in this systematic review, i.e., 32 and 40.4 months respectively.

It has not been possible to perform a meaningful comparison and/or meta-analysis on the association between cranioplasty material and postcranioplasty complications. There was lack of data and scarcity exists on uniform outcome parameters. Also, almost all studies were retrospective and single-center studies with a small number of patients. Therefore, based on the results of this systematic review, it is difficult to draw any meaningful conclusion to help guide clinical practice in the decision on when to opt for which type of cranioplasty.

While a randomized controlled trial would be the gold standard to analyze how cranioplasty material may effect postcranioplasty complication risk and outcomes, this is, ethically and logistically, very difficult to implement. A large-scale prospective multicenter study involving clear outcome parameters followed over a long period of time can overcome the issues discussed before.

Conclusion

Whether autografts or cranial implants are used, postcranioplasty complications and need for revision cranioplasty are quite common.

Beyond this, our systematic review illuminates that the existing literature does not contain well documented and comparable outcome parameters, suggesting that prospective, long-term multicenter cohort studies are needed to be able to optimize cranioplasty strategies in children who will undergo cranioplasty following craniectomy.

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Compliance with ethical standards

Conflict of interest With the submission of this manuscript, I would like to state that the authors report no conflict of interest concerning the materials and methods used or the findings specified in this paper. Furthermore, the authors declare that this study was performed in accordance with the research ethical guidelines.

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