

## ORIGINAL ARTICLE

# Scaphocephaly: a retrospective series examining treatment strategies and long-term follow-up

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## Summary

**Introduction:** Scaphocephaly, the most common form of craniosynostosis, is caused by the premature fusion of the sagittal suture, resulting in an elongated skull shape and constrained cranial growth.

**Material and methods:** This retrospective study included all consecutive patients with sagittal synostosis who were operated at the Clinic of Neurosurgery, UCCS, in the 20-year period. Detailed patient data were obtained from medical records and neuroradiological diagnostics. The follow-up period for these patients ranged from 6 to 20 years. The acquired data were thoroughly statistically analyzed.

**Results:** There were 93 children with a clear male preponderance. In the majority of children, there were no perinatal complications, and the majority of mothers didn't have any chronic illnesses. Three types of surgical treatment were used, with an almost equal distribution. The mean blood loss during surgery varied across the three operative techniques, with the lowest blood loss observed using the TSEO technique. Successful treatment was observed in only 57.1% patients who underwent strip-suturectomy, 96.6% patients treated with the  $\pi$  procedure, and all patients treated with the TSEO technique. Statistical analysis revealed a significantly higher success rate of the TSEO and  $\pi$  procedure compared to strip-suturectomy. Cranial index remained dolichocephalic in 6/28 children treated with strip-suturectomy alone, while in all cases treated with TSEO and  $\pi$  procedure, cranial index normalized. The majority of operated children had no complications.

**Conclusion:** Successful treatment of scaphocephaly requires a case-specific approach that extends beyond the surgical procedure itself. This series emphasizes the need for individualized treatment plans and a multidisciplinary approach throughout the surgical process.

**Keywords:** craniosynostosis surgery, pediatric neurosurgery, sagittal synostosis

## INTRODUCTION

Craniosynostosis is a condition defined by the premature fusion of one or more cranial sutures, resulting in an atypical cranial appearance and constrained cranial growth (1). Scaphocephaly (sagittal synostosis) is the most common form of isolated (unisuture) craniosynostosis with a distinct prevalence in the male population (3.5-4:1) (2, 3). It occurs in 0.2-1% of newborns and is, in most cases, easily recognized immediately at birth by a typical, narrowed, and elongated skull shape (4). Scaphocephaly holds considerable significance owing to its potential impact on a child's aesthetic appearance, neurological development, and psychosocial health. Early diagnosis and treatment are crucial to prevent not only cosmetic issues, but also increased intracranial pressure, impaired vision, developmental delays, and permanent neurological deficits. (5-8) The involvement of a multidisciplinary team, which includes neurosurgeons, plastic surgeons, pediatricians, nurses, physical therapists, and social workers, is vital throughout the treatment process.

## MATERIAL AND METHODS

This retrospective study includes all consecutive patients with sagittal synostosis who were operated at the Clinic of Neurosurgery, University Clinical Center of Serbia, over the period of 20 years, from January 1999 to December 2018. Detailed patient data were obtained from medical records and accompanying neuroradiological diagnostics.

The clinical data included prenatal medical history, family history, pregnancy history, associated congenital anomalies, age, and weight of the child at the time of surgery. Craniometric reference values included occipito-frontal circumference (OFC) and preoperative and postoperative cranial index (CI). Surgical intervention data included the type of surgical technique used, perioperative blood loss, presence of complications, and reoperations.

The outcomes were assessed through direct anthropometric measurements (head growth rate and cranial index), neuroradiological findings, and the Sloan classification systems to evaluate surgical results.

The surgery was indicated in cases when clear premature sagittal synostosis was diagnosed after clinical and neuroradiological examination – closed sagittal suture and its characteristic ridging, accompanied by characteristic head shape and the signs of increased intracranial pressure – “thumb printing” and subarachnoid collections over the frontal lobes.

Spiral computed tomography was performed from the skull base to the vertex using a Siemens SOMATOM Definition AS 128-slice Computed Tomography System (Siemens AG, Munich, Germany). All patients were scanned in a supine position in the axial plane. For the majority of children, neither general anesthesia nor se-

dation was used during imaging. The technical scanning parameters included a fast scan mode, wider collimation, rapid rotation time (0.5 seconds), and exposure parameters of 80-100 kV and 60-150 mA. The image was reconstructed with 1 mm-thick slices, and a 3D model was generated using Radiant DICOM Viewer (Medixant).

The follow-up period for these patients ranged from 6 to 20 years.

All statistical analyses were performed using IBM SPSS Statistics, version 20.0 (IBM Corp., Armonk, NY, USA). Descriptive statistics were used to summarize patient demographics, surgical details, and outcomes. The Kruskal-Wallis test was used to compare perioperative blood loss across the three surgical techniques, due to a violation of normality in one group (assessed using the Shapiro-Wilk test). Post hoc pairwise comparisons were conducted using the Mann-Whitney U test, with Bonferroni correction for multiple comparisons. Differences in ordinal outcomes, such as the Sloan classification, were analyzed using the Kruskal-Wallis test with post hoc Bonferroni adjustment. Comparisons of categorical data between groups were performed using the Chi-square test or Fisher's exact test, as appropriate. All patients included in the study had complete follow-up and relevant outcome data; there was no loss to follow-up. All tests were two-tailed, and a p-value < 0.05 was considered statistically significant. All statistical analyses were performed on anonymized patient data, ensuring that individual identities could not be disclosed or compromised.

This study has been conducted in full accordance with national and international ethical guidelines and standards relevant to this type of study.

## RESULTS

There were 93 children with sagittal craniosynostosis surgically treated at the Clinic of Neurosurgery, UCCS, in the period from January 1999 to December 2018. There was a clear male preponderance, since there were 71.0% (66) boys and 29.0% (27) girls. The median age at the time of the surgery was 6 months, with the youngest child being only 2 months old and the oldest 3.5 years old. The minimal weight of an operated child was 4450 g, with high variability, since the oldest child was 3.5 years old and weighing 18 kg.

In the perinatal period, 8.6% of children experienced asphyxia during delivery, and 1.1% had intracranial hemorrhage. In the remaining 90.3% there were no perinatal complications. The majority of children (98.9%) were born to pregnancies conceived naturally. Over 90% of the mothers were non-smokers, did not consume caffeine, and did not suffer from chronic illnesses. The majority of pregnancies were full-term, 82.8%, while only 16.1% were premature, and only 1.1% were post-term. There were no associated anomalies in 94.6% of children. How-

**Table 1.** Compared variables between the groups according to the type of surgical treatment

PROCEDURE	STRIP-CRANIECTOMY	$\pi$ PROCEDURE	TSEO
Blood loss	197.5 ml (IQR 171)	240.0 ml (IQR 105)	127.5 ml (IQR 90)
Sloan Classification – Good outcome (1,2)	16 (57.1%)	32 (97.0%)	32 (100%)
Postoperative OFC lower than normal (N)	6	0	0
CI < 75 (N)	6	0	0
Complications	3	2	2
Restenosis	3	1	0
N	28	33	32

ever, there was only one case of cleft palate and one case of congenital blindness. The youngest father was 20 and the oldest 41, with the mean age of 30.5 years. The youngest mother was 18 and the oldest was 39, with the mean age of 26.7 years.

Three types of surgical treatment were used. In 30.1% of cases, only a simple strip-suturectomy was performed, although the majority of these were cases done before 2010. A little more than a third (35.5%) were treated using the standard  $\pi$  procedure, and the remaining 34.4% were treated with the surgical technique created and modified in our institution in 2013 (Table 1) (9). The median blood loss during surgery varied across the three operative techniques. Patients who underwent strip-suturectomy had a median blood loss of 197.5 ml (IQR 171), compared to 240.0 ml (IQR 105) for the  $\pi$  procedure and 127.5 ml (IQR 90) for TSEO (Table 1).

Blood loss was compared among the three surgical technique groups using the Kruskal-Wallis test, due to a violation of normality in one group. Kruskal-Wallis test revealed a significant difference in intraoperative blood loss among the three surgical techniques ( $p < 0.001$ ). Subsequent pairwise comparisons were conducted using the Mann-Whitney U test with Bonferroni correction for multiple comparisons (adjusted significance threshold  $p < 0.017$ ) and showed that the blood loss was statistically significantly higher only in the  $\pi$ -procedure group compared to the TSEO group ( $p < 0.001$ ). Although the median blood loss appeared lower in the TSEO group compared to the strip-suturectomy group, this difference did not reach statistical significance in the pairwise analysis ( $p = 0.065$ ), as shown in Table 2.

The success of surgical treatment for sagittal synostosis was evaluated using the Sloan classification and by comparing the cranial index (CI) before and after surgery. According to the Sloan classification, classes 1 and 2 are considered successful treatment, and this was achieved in 86.0% of cases. When analyzed according to the surgical

technique used, successful treatment was observed in only 57.1% patients who underwent strip-suturectomy, 97.0% patients treated with the  $\pi$  procedure, and all patients treated with the TSEO technique (Table 1). Statistical analysis revealed a significant difference between groups (Kruskal-Wallis test,  $p < 0.001$ ). Pairwise comparisons using the Mann-Whitney U test, with Bonferroni correction applied (significance threshold  $p < 0.017$ ), demonstrated statistically significant differences between all surgical techniques. Specifically, the outcome scores differed significantly between strip-suturectomy and  $\pi$  procedure ( $p < 0.001$ ), strip-suturectomy and TSEO ( $p < 0.001$ ), as well as between the  $\pi$  procedure and TSEO ( $p = 0.006$ ). These findings indicate that each surgical technique was associated with significantly different treatment outcomes.

The rate of successful treatment (Sloan 1 or 2) was significantly higher in the TSEO (32/32, 100%) and  $\pi$  procedure (32/33, 97.0%) groups compared to the strip-suturectomy group ( $p < 0.001$ ). Post hoc pairwise Fisher's exact tests with Bonferroni correction confirmed that both the TSEO and  $\pi$  procedure groups had significantly higher success rates than strip-suturectomy ( $p < 0.001$  for both comparisons). In contrast, the difference between TSEO and the  $\pi$  procedure was not statistically significant ( $p = 0.508$ ) as shown in Table 3.

The proportion of patients with an abnormal postoperative cranial index (CI < 75) differed significantly between surgical techniques. Among children treated using strip-suturectomy, 6/28 children had a persistently abnormal CI, whereas all patients in the TSEO and  $\pi$  procedure groups achieved normalization of the cranial index (Table 1). This difference was statistically significant ( $p < 0.001$ ). The occipito-frontal circumference (OFC) in these 6 cases also remained below the appropriate value for age and gender (Table 1). Preoperative and immediate postoperative shape of the skull in a patient with sagittal synostosis using the TSEO technique in our series is presented in Figure 1.

**Table 2.** Comparison of blood loss among surgical techniques

Comparison	Median Blood Loss (ml)	p-value (Kruskal-Wallis)	Pairwise p-values (Mann-Whitney, Bonferroni adj.)
Strip-suturectomy	197.5		strip-suturectomy vs $\pi$ procedure: 0.090
$\pi$ procedure	240.0	<0.001	strip-suturectomy vs TSEO: 0.065
TSEO	127.5		$\pi$ procedure vs TSEO: <0.001

Table 3. Comparison of good outcome (Sloan 1 and 2)

Comparison	Good Outcome	p-value (Chi-square/Fisher's exact)	Pairwise p-values (Fisher's exact test)
Strip-suturectomy	16/28 (57.1%)	<0.001	strip-suturectomy vs $\pi$ procedure: <0.001
$\pi$ procedure	32/33 (97.0%)		strip-suturectomy vs TSEO: <0.001
TSEO	32/32 (100%)		$\pi$ procedure vs TSEO: 0.508

The majority of operated children had no complications (92.5%); however, there were 4 cases of wound infection, two respiratory infections, and one case of liquor-rhea. Three children operated using strip-suturectomy and one using the  $\pi$  procedure were reoperated because of restenosis. All cases of wound infection were mild and resolved after antibiotic treatment.

DISCUSSION

Preoperative considerations for scaphocephaly include accurate diagnosis to differentiate between synostotic and nonsynostotic cases, as the treatment varies (10, 11). An absolute contraindication for neurosurgical intervention is microcephaly, in which the skull sutures close secondarily, due to the absence of normal brain development and expansion (12). The patient's age, severity of the deformity, and associated risks such as elevated intracranial pressure must be assessed. Surgical options range from limited-access cranial vault remodeling for younger patients with less severe deformities to total cranial vault remodeling for older patients (12). Understanding the

potential for blood loss and the length of hospital stay is crucial for planning and optimizing patient outcomes. In our series, particularly with the specifically developed TSEO technique, blood loss was lower than in both strip-suturectomy and  $\pi$  procedure. However, statistical significance was observed only in comparison with the  $\pi$  procedure.

Most studies suggest that the best time to perform surgery is between 3 and 6 months of age. This period is considered ideal due to the flexibility of the skull at this age, which minimizes the risks of complications such as reossification or restenosis after surgery (12-14). In our series, this period is highly variable, varying from 2 months to 3.5 years; however, no significant difference was found, since the majority of cases achieved excellent surgical outcomes.

There are various surgical choices and views on the ideal treatment, but clear guidelines are still not available, and there is insufficient research to endorse a specific surgical method (12). While early intervention is generally recommended, the choice of surgical technique and timing should be tailored to the individual patient's condition and the surgeon's expertise (12-14).

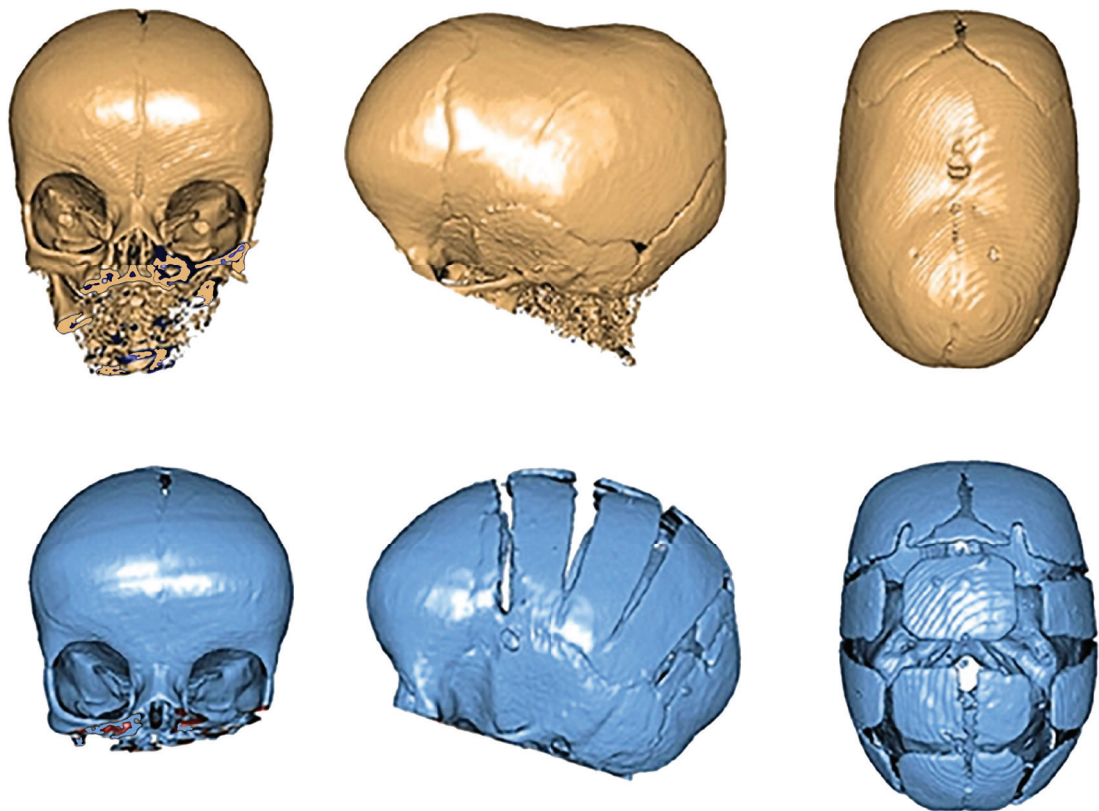


Figure 1. Preoperative (top row) and immediate postoperative (bottom row) shape of the skull in a patient with sagittal synostosis using the TSEO technique in our series

There are many available surgical options. The initial treatment, in the early years of craniosynostosis surgeries, consisted of a simple sagittal suturectomy; however, as is also shown in our series, the aesthetic outcome and the achieved cranial decompression were poor.

Then, the  $\pi$  procedure was developed, which involves creating a specific bone incision pattern that resembles the letter “ $\pi$ ” on the skull (15). A subgaleal flap is raised to visualize the skull from the front fontanel area and the coronal sutures to the lambdoid sutures. A 4-cm strip of bone is carefully taken out from the bregma to the lambda along the superior sagittal sinus. The temporal/parietal bone flaps are cut or greenstick-fractured at their bases while still connected to a piece of temporal muscle, allowing for widening of the temporal/parietal area. The anterior–posterior length is reduced immediately; the midline bone flap is shortened to fit this new distance from bregma to lambda. Then, this midline bone flap is divided into two parts that are placed back and fixed with transosseous sutures to cover the sagittal sinus.

In our institution, we modified this technique to reduce blood loss, enhance the esthetic results, and achieve better decompression. Therefore, the TSEO technique was introduced to protect the superior sagittal sinus during cranial vault reconstruction and to extend the distance between osteotomes, thus reducing the rate of restenosis. It also involves remodeling of the frontal bone or occipital bone, depending on whether there is frontal bossing or occipital prominence (9).

In the last 5 years, throughout many craniosynostosis centers, the endoscopic-assisted craniosynostosis surgery (EACS) has been frequently used with helmet therapy. This procedure is claimed to be less invasive and leads to reduced blood loss and shorter hospital stays than open cranial vault reconstruction (16, 17). Infants are typically operated on at an early age, ideally before 3 months, to take advantage of rapid brain growth and skull plasticity. A small skin incision measuring approximately 2.5–3 cm is made behind the hairline, subsequently followed by a strip craniectomy along the fused sagittal suture. It provides comparable craniometric outcomes at 48 months after the surgery (16, 17).

Common complications of craniosynostosis surgery include CSF leaks, infection risk, blood loss, and temperature dysregulation (18–20). In our series, we have found a very low rate of complications (7.5%) and a very low volume of blood loss during surgery in the last 10 years, especially with the TSEO technique.

The intermediate postoperative phase, spanning from 1–6 months post-surgery, requires monthly clinical evaluations to assess skull growth patterns and monitor for potential relapse. Long-term follow-up continues until skeletal maturity, with annual assessments of head shape, growth parameters, cognitive development, visual function, and social adaptation. Psychological support for

both family and patient, integration with pediatric therapy services when needed, educational support planning, and ongoing monitoring of aesthetic outcomes all contribute to comprehensive care (21). Healthcare providers must offer detailed explanations of the surgical process and expected outcomes, provide guidance on head positioning and handling, ensure families can recognize potential complications, and connect them with appropriate support groups and resources. Family support and education represent crucial elements of successful treatment.

A key limitation of this retrospective study is the potential for unmeasured confounding factors, such as variations in patient age, baseline severity, or comorbidities, which may have influenced surgical outcomes. Although multivariate regression analysis was considered, the sample size within each surgical group limited the feasibility of adjusting for multiple covariates. Future studies with larger cohorts may benefit from multivariate approaches to better account for potential confounders.

## CONCLUSION

Successful treatment of scaphocephaly requires a case-specific, tailored approach that extends well beyond the surgical procedure itself. In our study, both the  $\pi$  procedure and TSEO were associated with better surgical outcomes compared to strip-suturectomy, particularly in terms of aesthetic results and normalization of the cranial index. Careful attention to both presurgical planning and postsurgical care, combined with comprehensive family support and long-term follow-up, provides the best foundation for optimal outcomes. As medical techniques continue to evolve and our understanding of craniosynostosis improves, treatment protocols will likely continue to be refined, potentially leading to even better outcomes for patients affected by this condition. Ongoing research and clinical experience will continue to improve surgical approaches, aiming for enhanced safety and efficacy in managing this condition.

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**Ethical approval:** This study has been conducted in full accordance with national and international ethical guidelines and standards relevant to this type of study.

**Informed consent statement:** Informed consent was obtained from all subjects involved in the study.

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# SKAFOCEFALIJA: RETROSPEKTIVNA ANALIZA TERAPIJSKIH PRISTUPA I DUGOROČNOG PRAĆENJA

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## Sažetak

**Uvod:** Skafocefalija, najčešći oblik kraniosinostoze, uzrokovana je prevremenim srastanjem sagitalne suture, što dovodi do elongiranog oblika lobanje i ograničenja njenog rasta.

**Metod:** Ova retrospektivna studija obuhvatila je sve uzastopne pacijente sa sagitalnom sinostozom koji su operisani u Klinici za neurohirurgiju, UCCS, u periodu od 20 godina. Detaljni podaci o pacijentima prikupljeni su iz medicinske dokumentacije i neuroradiološke dijagnostike. Period praćenja pacijenata kretao se od 6 do 20 godina. Prikupljeni podaci su detaljno statistički analizirani.

**Rezultati:** Operisano je 93 dece sa jasnom prevalencijom muškog pola. Kod većine dece nije bilo perinatalnih komplikacija, a kod većine majki nije bilo hroničnih bolesti. Korišćene su tri vrste hirurškog lečenja, gotovo ravnomerno raspoređene. Prosečan gubitak krvi tokom operacije varirao je između tri hirurške tehnike, sa naj-

manjim gubitkom krvi kod TSEO tehnike. Uspešno lečenje zabeleženo je kod samo 57,1% pacijenata koji su podvrgnuti strip-suturektomiji, kod 96,6% pacijenata tretiranih  $\pi$  procedurom i kod svih pacijenata tretiranih TSEO tehnikom. Statistička analiza je pokazala značajno viši stepen uspeha TSEO i  $\pi$  procedure u poređenju sa strip-suturektomijom. Kranijalni indeks ostao je u opsegu dolichocefalije kod 6/28 dece tretirane isključivo strip-suturektomijom, dok je kod svih slučajeva tretiranih TSEO i  $\pi$  procedurom kranijalni indeks normalizovan. Većina operisane dece nije imala komplikacije.

**Zaključak:** Uspešno lečenje skafocefalije zahteva specifičan pristup za svakog pacijenta koji prevazilazi samo poznavanje hirurške procedure. Ova serija pacijenata ističe potrebu za individualizovanim planom lečenja i multidisciplinarnim pristupom celokupnog hirurškog procesa.

**Glavne reči:** hirurgija kraniosinostoze, pedijatrijska neurohirurgija, sagitalna sinostoza

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