The Dura Split Technique in the Treatment of Craniosynostosis - is it still an option?

ABSTRACT

Background: The aim of this study was to report the outcome and the complications for patients operated on for craniosynostosis using the dura split technique. Specifically, we aimed to evaluate the safety of this technique, which is currently not in use, and to determine whether it is still useable.

Methods: The data was collected from the hospital patient records of all children surgically treated for craniosynostosis using the dura split technique in Turku University Hospital during the period 1975-2015. The data was analyzed to determine the clinical and radiological outcomes of the surgical procedure, the need for reoperations, and the rate of complications.

Results: During the study period, the dura split technique was used in the surgery of 65 patients. The outcome was either good or acceptable in most patients and reoperation was needed in only two patients (3.1%). Surgical complications included significant blood loss (26.2%), lesions on the inner layer of the dura (21.5%), leakage of cerebrospinal fluid (13.8%), and persistent bone defects (15.4% on palpation and 63.1% radiologically).

Conclusion: While the outcome of surgery for craniosynostosis using the dura split technique was mostly acceptable and the need for reoperations rare, the technique cannot, however, be recommended in the future due to high rates of bone defects, frequent problems with lesions on the inner layer of the dura, and consequent perioperative leakage of cerebrospinal fluid.

INTRODUCTION

The reported incidence of craniosynostoses varies between 2.8 and 6.4 per 10,000 live births [1, 2]. Several techniques have been used for the surgical treatment of these conditions with the objective to find a technique with acceptable morbidity and also good neurocognitive and cosmetic results [3, 4]. Attempts have been made to minimize the invasiveness of the operations to reduce morbidity and perioperative blood loss [3]. However, early re-ossification after minimally invasive procedures has sometimes led to a need for reoperation. Thus, different techniques have been developed to prevent too rapid fusion of the osteotomy site [3, 5].

Foreign material interposition, such as polyethylene or silicone, has been used to prevent too rapid re-ossification of the osteotomy site after surgery for craniosynostosis [5]. To avoid the use of foreign material and to minimize the associated risk of infection, van Der Werf described his technique in the 1970s. He suggested splitting the outer layer of the dura from the inner layer, turning it over the edge of the bone at the osteotomy site, and suturing it to the periosteum [5, 6]. This `dura split` technique was developed in the hope of delaying the ossification of the osteotomy site before adequate reshaping of the skull is achieved. Van der Werf observed that the craniotomy lines remained open for a long time after the primary surgical procedure, thereby enabling adequate molding and growth of the skull. It was estimated that the use of this technique avoided the need for reoperation and reduced the risk of infection compared to the artificial materials used to inhibit early ossification [5]. For a while, the technique was widely used. However, suspicion of an increased rate of complications reduced the popularity of this technique, and currently many consider it too risky.

To the best of our knowledge, no studies have so far been published on the outcomes and complications of the dura split technique. Our objective was to report the surgical outcomes, complications, and reoperations for patients treated for craniosynostosis in Turku University Hospital using the dura split technique. We hypothesized that complications such as leakage of cerebrospinal fluid and significant blood loss during surgery would be common and that there might be an elevated risk for permanent bone defects in the skull.

MATERIALS AND METHODS

Patients and data collection

In this study, we included all patients surgically treated for craniosynostosis using the dura split technique in Turku University Hospital in the period 1975-2015. We collected and analyzed data retrieved from the patient records, the surgical reports, photographs, skull x-rays, head magnetic resonance images (MRI) and computer tomography scans (CT) including the radiological reports. To search for data from the hospital archives, we used ICD-8, ICD-9 and ICD-10 (International Statistical Classification of Diseases and Related Problems 8^{th}, 9^{th}, and10^{th} Revisions) codes for craniosynostoses. In addition, the codes for surgeries for craniosynostosis of the Nordic Medico-Statistical Committee (NOMESCO) Classification of Surgical Procedures Finnish national version (NCSP-F, that has been used since 1997) were used to retrieve data from the hospital archive and since 2000 also from hospital's electronic registers.

Altogether 164 patients were identified in the initial database search. The charts for eight patients identified during the search could not be located from the hospital archives. The surgery report was missing for six patients. Three patients had not been operated on due to other morbidity or mildness of the condition. One patient had been operated on at another hospital. Four patients did not have craniosynostoses at all. Out of these 142 patients, 73 patients were excluded because other surgical techniques were preferred for their respective surgical procedures and a further four because they had syndromic craniosynostosis.

The complications of surgery were defined as significant blood loss during surgery, postoperative wound infection, lesion on the inner layer of the dura, cerebrospinal fluid leakage during or after surgery, bulging or herniation of the brain at the osteotomy site, and delayed ossification.

Significant blood loss during surgery was defined in our study as a mention in the surgery report of profuse blood loss during surgery, a mention of measured blood loss of ≥200ml during surgery, or a mention of ≥30% blood loss of estimated total blood volume. Bone defects were regarded as delayed or missing ossification if bone gaps were present more than 12 months postoperatively. Delayed ossification of the osteotomy site was recorded separately for a mention of bone defects on palpation and a mention of bone gaps in the radiological reports or bone gaps seen in the skull x-rays, head CT, or MRI.

Reoperations for craniosynostoses or for complications of the primary surgery for craniosynostoses were recorded. The surgical outcome was also assessed according to patient records using scoring by Aryan et al. [7]. This is a scoring method which classifies the outcome of surgical procedures into four categories (see Supplemental Table 1) and is a modification of a slightly simpler classification originally published by Whitaker et al. [8].

Ethics

The study was approved by Turku University Hospital Clinical Research Services Research Center (permissions no: T135/2018). This was a retrospective register study and the participants were not contacted. Thus, no informed consent or ethics committee approval was required. The legal basis for processing of personal data is public interest and scientific research (EU General Data Protection Regulation 2016/679 (GDPR), Article 6(1)(e) and Article 9(2)(j); Data Protection Act, Sections 4 and 6).

RESULTS

The dura split technique was used in 65 patients with craniosynostosis during the study period. Mean age at primary surgery was 3.7 months (median 3, SD 2.92, range 0.2-13 months). Most of the patients were treated for sagittal synostoses and 75.4% were male. The background characteristics of the study patients are shown in Supplemental Table 2. The mean surgical outcome score was 2.02 points (median 2, SD 0.63, range 1-4) (see Supplemental Table 3). None of the study patients died during surgery or in the immediate postoperative period.

An intraoperative photograph of the dura split technique is shown in Figure 1A.

Complications

Intra- and postoperative complications are listed in Supplemental Table 4. Significant blood loss was common during surgery (26.2% of the patients). In addition to the patients reported as having significant blood loss, one patient was reported to have had a small hole in the sinus sagittalis that was repaired with sutures.

Lesions on the inner layer of the dura were reported in 21.5% of cases. These lesions were mostly caused during the splitting of the dura and during the release of bone from the dura. The lesions were mostly less than 2 cm in size, and they were repaired using sutures, the pericranium (galea aponeurotica), periosteum, or commercial products, or combinations of these.

Delayed ossification was apparent in ten patients on palpation and in 41 patients radiologically ≥12 months after surgery. Bone defects were situated lateral to the sagittal suture and were mostly narrow. However, some defects were large, up to 10 cm in length. Examples of typical bone defects are shown in Figure 1B-D. Data on palpation was missing for 11 and on radiological findings for 21. Most of those with missing data were historical cases with a short follow up of less than 12 months after surgery. Average follow-up time for all patients from the primary surgery to the last clinical follow up and head

palpation was 4.9 years (min 0.08 - max 15.1) and to the last radiological checkup 5.3 years (min 0.08 - max 36.8).

No postoperative infections were reported in the patient records after dura split procedure and reoperations for complications were rare.

Reoperations

No reoperations were needed for immediate complications in the postoperative period. However, two patients with primary surgery for sagittal synostoses at the age of one month were reoperated on at a later stage. The dura split method was not used in the redo surgeries. In addition, reoperation was recommended at 18 years of age for a sagittal synostosis patient with large postoperative bone defects (up to 10 cm in length) lateral to the sinus sagittalis, but the patient declined.

Both patients undergoing reoperation suffered a lesion on the inner layer of the dura during the primary surgery. In the first patient, postoperative cerebrospinal fluid leakage was noted and treated conservatively. However, the patient later developed a 1 x 3 cm bone defect at the site of the previous leakage with elevated bony edges. This bothered the patient, occasionally causing pain when accidentally bumping the head. Reoperation was performed at six years of age. In the second patient, no cerebrospinal fluid leakage was reported despite the lesion on the inner layer of the dura. The patient later developed a 3 x 7 cm bone defect on the left side lateral to the sinus sagittalis and a bony prominence on top of the sinus sagittalis with skin problems. Reoperation was performed at 4.5 years of age.

The dura split method was not used in the redo surgeries. In both patients, the bony prominences were evened out. In the first patient, bone from the site of the prominence was used as autograft to the unossified site. This patient also had bulging of the brain into the bony defect. In the second patient, the dura was scarred and a new lesion on the inner layer of the dura was caused during the second craniotomy at the release of bone from dura. Periosteum was used to cover the bony defect to avoid prominence. However, in both patients, bony defects were still apparent at later follow-up visits. No new reoperations were scheduled.

DISCUSSION

In this retrospective register study, we reported the use of the dura split technique in 65 surgeries for craniosynostoses in Turku University Hospital during the period 1975 to 2015. The outcome was good or acceptable in most patients and reoperations were needed in only 3.1% of the cases. However, significant blood loss (26.2%), unintentional lesions on the inner layer of the dura (21.5%), and leakage of cerebrospinal fluid (13.8%) were reported in several cases during surgery and delays in ossification were common (15.4% on palpation and 63.1% radiologically).

To the best of our knowledge, this is the only long-term follow-up study on the outcome of surgery for craniosynostoses using the dura split technique. Due to the retrospective nature of this study, we did not have a control group treated for craniosynostoses using other surgical techniques.

Due to the historical nature of the data, there were some inconsistencies in the reports of intraoperative blood loss. In some cases, we had to rely on the surgical reports and on other patient records in the assessment of blood loss instead of anesthetic reports with exact quantifications of blood loss in milliliters. In addition, we were not able to analyze the number of postoperative transfusions because this data was no longer available on patients treated before the introduction of electronic patient records. In interpreting the results concerning intraoperative blood loss, one must bear in mind that methods of managing intraoperative blood loss have developed considerably from the 1970s to the 21st century [9, 10]. Also, it has previously been reported that significant blood loss and the need for blood transfusions are common in the surgical treatment of craniosynostoses. Park and associates reported an average estimated volume red cell loss of 77% of the circulating volume during craniosynostosis surgery and 90% of the patients operated on for craniosynostoses received blood transfusion perioperatively [11].

Earlier studies have reported incomplete re-ossification to vary between 0.5% and 18.2% after surgery for craniosynostosis using different surgical techniques [12, 13]. However, there has been considerable variation in the follow-up time and diagnostic method (i.e., palpation, radiology, or both) in these earlier reports. As far as we know, there is no existing data available on the rate of delayed ossification related to the dura split technique. In the present study, delayed ossification was seen in most of the patients with data on radiological investigations performed more than 12 months after surgery and felt on palpation for 15.4%. However, only two patients needed reoperation due to delayed ossification and associated problems.

It therefore seems that the overall results in regard of reshaping of the head are reasonable. However, the dura split technique carries a risk for lesions on the inner layer of the dura, leakage of cerebrospinal fluid, and bleeding. Nevertheless, the need for reoperations is relatively low. We showed high rates of bone gaps on palpation and especially radiologically, indicating that the dura split technique carries a particular risk for delays in ossification of the osteotomy site.

CONCLUSION

While the outcomes of surgery for craniosynostosis using the dura split technique were mostly acceptable and the need for reoperations was rare, we do not think that the dura split technique has a role in modern craniosynostosis surgery due to high rates of bone defects, frequent problems with lesions on the inner layer of the dura, and perioperative leakage of cerebrospinal fluid.

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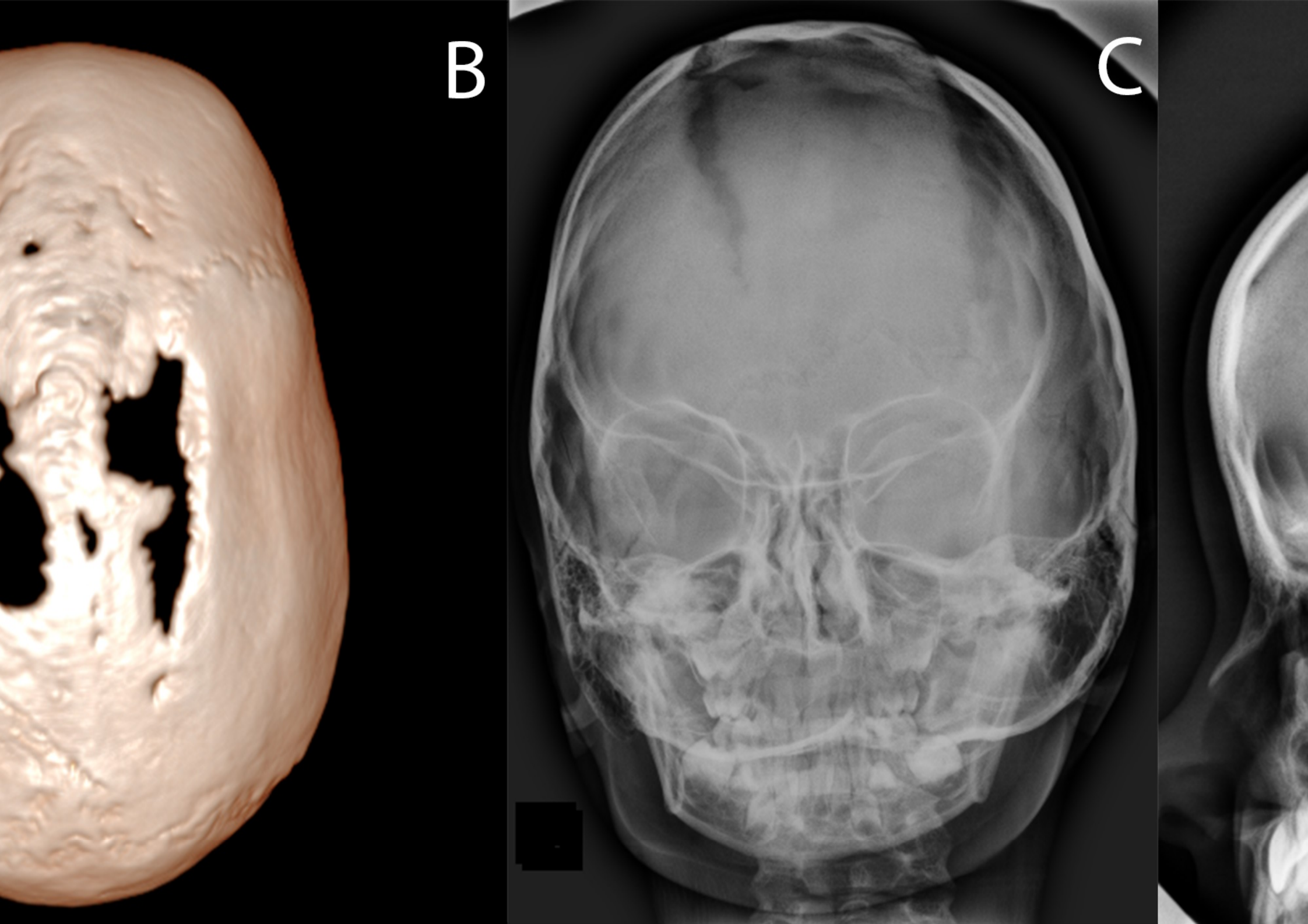
FIGURE LEGENDS

Figure 1. (A) An intraoperative photograph from a corrective surgery of a sagittal synostosis using the dura split technique showing the sinus sagittalis (blue arrow) and the outer layer of the dura sutured over the edge of the osteotomy site (yellow arrows). (B-D) Typical postoperative parasagittal bone defects after correction of sagittal synostosis using the dura split technique; (B) Severe bone defects 16.5 years postoperatively in computer tomography and (C-D) milder ossification defects in a patient 3.5 years postoperatively in x-ray.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