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Endoscopically assisted craniosynostosis surgery (EACS): The craniofacial team Nijmegen experience

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A B S T R A C T

Introduction: An evaluation of our first 111 consecutive cases of non-syndromic endoscopically assisted craniosynostosis surgery (EACS) followed by helmet therapy.
Methods: Retrospective analysis of a prospective registration database was performed. Age, duration of surgery, length of hospital stay, blood loss, transfusion rate, cephalic index and duration of helmet therapy were evaluated. An online questionnaire was used to evaluate the burden of the helmet therapy for the child and parents.
Results: 111 EAC procedures were performed: 64 for scaphocephaly, 34 for trigonocephaly and13 for anterior plagiocephaly. The mean age at the time of surgery was 3.9 (±1) months, mean surgical time was 58 (±18) minutes, mean blood loss was 34 (±28) ml, transfusion rate was 22% (n = 26), mean duration of postoperative helmet therapy was 10 (±2.5) months, mean preoperative and postoperative CI were respectively 0.67(±0.057) and 0.72 (±0.062) in scaphocephalic patients and the mean length of hospital stay was 2.6 (±1) days. The burden of the helmet therapy for the child and his family was deemed very low.
Conclusion: EACS for non-syndromic patients shows low morbidity rates, short surgical time, short length of hospital stay, little blood loss and low need for blood transfusion and is associated with satisfying cosmetic results.

1. Introduction

Craniosynostosis occurs in 1:2000–2500 living births and is defined by the premature closure of one or more cranial sutures, causing typical head shapes depending on the affected suture. Several genetic disturbances are identified in syndromic cases of craniosynostosis, but the etiology of single suture craniosynostosis remains largely unknown (Morriss-Kay and Wilkie, 2005, Senarath-Yapa et al., 2012).

Since the first operative treatment of craniosynostosis in 1890 by Lannelongue (Lannelongue, 1890), surgical methods to treat craniosynostosis have evolved from a relatively simple strip craniectomy to a diverse spectrum of partial or complete cranial vault remodeling (Mehta et al., 2010). More complex procedures, such as the pi-procedure, have shown excellent results yet often with high comorbidity (Kanev and Lo, 1995).

Therefore, minimally invasive craniosynostosis surgery has been explored in the last few decades. The main goal of minimally invasive craniosynostosis surgery is to reduce the morbidity and invasiveness of classical surgical procedures, with equal long term results both functional as cosmetic (Sanger et al., 2014).

Emphasis is placed on early operations through less invasive procedures in order to halt the associated morbidity and limit

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compensatory growth mechanisms of the skull. With the introduction of endoscopic surgical approaches, a clear drop in blood loss, operative time, complications and morbidity was reported for surgical treatment of craniosynostosis (Jimenez and Barone, 1998, 2000; Jimenez et al., 2002). To reach these goals in our centre, we adopted endoscopy-assisted suturectomy supplemented with helmet molding therapy in 2005, as described by Jimenez et al. (Jimenez and Barone, 2000, 2012a, 2012b, 2012c, 2013; Jimenez et al., 2002).

In this study the results of endoscopically assisted craniosynostosis (EAC) surgery followed by postoperative helmet therapy at Radboudumc, Nijmegen, The Netherlands have been evaluated, including the burden of the helmet therapy. We present the results of our first 111 consecutive non-syndromic cases.

2. Materials and methods

This is a retrospective study of a well-defined patient cohort in an Academic Hospital performed in accordance to local rules from the institutional board review. We performed a retrospective analysis of a prospective database in which all neurosurgical pediatric procedures, complications and follow up are included since 2004 (van Lindert et al., 2014). From this database we retrieved demographic information, diagnosis, type of procedure and intra- and post-operative complications. The following variables were evaluated: duration of surgery, length of hospital stay, estimated blood loss, transfusion rate, age at time of surgery, duration of helmet therapy, surgical results including cephalic index for scaphocephalic patients, re-operation rate and occurrence of papilledema. These variables were both evaluated by diagnosis and for the total cohort of patients. Besides single suture craniosynostosis, we also treated multisutural and syndromic cases. Because of the different treatment goal and excessive morbidity these cases will be presented in another study.

Postoperative skull shape change was evaluated using 3D stereophotogrammetry. Preoperative and postoperative 3D-photographs were acquired using a 5-point 3D stereophotogrammetical camera set up and processed using 3dMD patient software (3dMDCranial™, 3dMD Patient, 3dMD ALL, Atlanta, USA).

In order to examine patient satisfaction and to evaluate the EAC surgery including the benefit and burden of therapy, we created an on-line questionnaire, covering social impact of the treatment, experiences with the helmet therapy and general satisfaction. To assure anonymity, no identifying data were gathered. Likert items were used to measure the level of agreement on presented statements. The questionnaire was edited in Dutch, an English translation is presented in Appendix A for readers' convenience.

All data were expressed in mean and standard deviation. Paired t-test was used to determine the level of significance in paired data. The cutoff for significance was $p < 0.05$.

3. Results

From August 2005 until December 2014, 111 EAC procedures were performed. Most patients suffered from scaphocephaly ($n = 64$, 57%) and trigonocephaly ($n = 34$, 30%) as shown in Table 1.

The mean age at the time of surgery was 3.9 (±1) months. Scaphocephalic patients were the oldest at time of surgery, with a mean of 4 (±1) months.

Overall, the mean surgical time of EAC surgery in this study was 58 (±18) minutes. Scaphocephaly procedures were somewhat longer (62 ± 20 min) compared to trigonocephaly (55 ± 15) and anterior plagiocephaly procedures (49 ± 12 min). We noticed a learning curve with a clear reduction in operative times with growing experience (Fig. 1).

The mean length of hospital stay was 2.6 (±1) days, mainly consisting of one preoperative day, the day of surgery and one postoperative day. Again, the learning curve shows a decline in length of hospital stay in time (Fig. 2). This reflects the prudence taken by the surgical team regarding the new technique at the start of our experience. In time, we evolved to a postoperative stay of 24 h, which we maintain today. Likewise, the first three patients receiving EACS stayed in the ICU for two days, but since then, none of the patients receiving EACS stayed in the ICU.

The mean estimated blood loss was 34 (±28) ml. One blood transfusions was needed during surgery and the postoperative blood transfusion rate was 22% ($n = 24$). Transfusion rate was highest in scaphocephalic patients (30%), as shown in Table 1. However blood loss was highest in trigonocephalic patients, 39 (±25) ml. There was no clear learning curve concerning blood loss.

3D stereo-photogrammetry was used for follow up allowing a clear visualization of skull shape change in time. Overall, esthetic results were satisfying for parents and the craniofacial team (Figs. 3–5). In trigonocephaly cases, we noticed a persistent flattening of the front just above the lateral brow at the time helmet therapy was stopped. This normalized very slowly in time. Therefore, we added small wedge-shaped osteotomies pointed towards the upper lateral orbital edges to our treatment protocol in 2012. No long term follow up data is yet available to evaluate whether these osteotomies assist in the outbending of the flat area just above the lateral brow.

For scaphocephalic patients, we calculated cephalic index values. However, being a retrospective study, we could only retrieve in 54 out of the 64 scaphocephalic patients preoperative and postoperative cephalic index values, measured from 3D photography since the photos of the other ten patients were of low quality. Postoperative CI was measured at least more than 6 months after surgery in order to get a clear view of the progression. The mean preoperative cephalic index was 0.67 (±0.057) and the mean postoperative cephalic index was 0.72 (±0.062) (paired *t*-test: $p < 0.001$).

Re-operation rate was very low (2.7%, $n = 3$). One patient treated with EACS for anterior plagiocephaly underwent a fronto-orbital advancement at a later age in another centre, for cosmetic reasons. Two other patients, both treated for plagiocephaly, displayed a persistent craniectomy gap with outbending of the bony edges, suggesting raised intracranial pressure although no clinical signs or symptoms were present. These patients underwent fronto-orbital remodeling.

Standard ophthalmological control for papilledema took place pre-operatively, at the age of 2 y.o. and 4 y.o. None of the single suture craniosynostosis cases developed papilledema, with a minimal follow up time of 1 year.

Table 1
Shown are the number of patients, age at the time of surgery, the mean duration of surgery (DOS), mean length of hospital stay (LOS), mean blood loss (BL), transfusion rate (TR) and the mean duration of the helmet therapy (HT) per diagnosis.

Diagnosis	Number	Age	DOS	LOS	BL	TR	HT
Scaphocephaly	64	4.0 (±1.1)	62 (±20)	3.0 (±1.1)	32 (±30)	19 (30%)	9.8 (±2.7)
Trigonocephaly	34	3.7 (±0.9)	55 (±15)	2.2 (±0.52)	39 (±25)	6 (18%)	11 (±2.0)
Anterior Plagiocephaly	13	3.9 (±1.1)	47 (±11)	2.2 (±0.60)	34 (±26)	0	11 (±2.6)
Total	111	3.9 (±1.0)	58 (±18)	2.7 (±1.0)	34 (±29)	25 (23%)	10 (±2.5)