Comparative outcomes of craniectomy versus cranial remodelling in the human infant with isolated sagittal synostosis

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ABSTRACT

BACKGROUND

Craniosynostosis is a congenital condition characterised by the premature closure of one or more cranial sutures. The sagittal suture is the most common site, comprising 40-60% of cases. Premature fusion of this suture can cause scaphocephaly, seen morphologically as a narrow elongated skull with a decreased cephalic index. Diagnosis is made clinically and/or radiologically. The goals of surgical correction and the techniques used have evolved over time. Whilst there has been a general move from limited craniectomy to calvarial remodelling, in recent times there has been a return towards less invasive methods.

OBJECTIVES

The objectives were to identify and synthesize the best available evidence on the morphological, functional and neurological outcomes of craniectomy compared to cranial vault remodelling and compare this to existing results.

METHODS

A systematic review of the literature was conducted using the Joanna Briggs Institute methodology. The review considered studies of infants with primary isolated sagittal synostosis operated on before a mean of two years of age. The intervention of interest
was sagittal craniectomy; this was compared to cranial vault remodelling. Morphological, functional and neurological outcomes were included. Mortality, complications and aesthetic outcome were included as tertiary outcomes. A comprehensive search was undertaken across major databases. Retrieved studies were assessed by two independent reviewers for methodological validity. Data was extracted and where possible, pooled in statistical meta-analysis. Where this was not possible, findings were presented in narrative form.

RESULTS

Based on critical appraisal 27 studies, all descriptive in nature, were of suitable quality for inclusion. Meta-analysis was only possible for the primary morphological outcome (mean change in cephalic index post-operatively) based on two studies. This showed that at one year post-operative follow-up remodelling offers an advantage over craniectomy (Z = 4.16, P<0.0001). Narrative synthesis suggests that improvements of cephalic index to varying degrees were seen in patients receiving either procedure; whilst the mean change appears to peak early in patients who have undergone remodelling procedures, the trend suggests it may improve in the longer term after craniectomy.

Whilst global IQ scores may be comparable to an age-matched population, narrative review suggests that patients with sagittal synostosis who have undergone a surgical correction of any type may have discrepancies in specific domains and may be at risk of developing learning disorders. There is insufficient primary research with inter-
procedure comparison of pre-operative and post-operative cognitive or neurological outcome.

CONCLUSIONS

At one year follow-up, remodelling is superior to craniectomy in terms of mean change in cephalic index. However both procedures were seen to give improvements in the short, medium and long term. Neither procedure offers a distinct sustained advantage; longer follow-up is required to assess the comparative improvement over time.

There is insufficient evidence whether craniectomy or remodelling procedures offer superior functional or neurological outcome. Patients who have had surgical repair (any type) may have deficiencies in different subdomains and be at risk of learning disorders, whilst maintaining an age-appropriate global IQ and school performance. It is unknown if either surgery impart any restorative or protective benefit.
DECLARATION

I certify that this work contains no material which has been accepted for the award of any other degree or diploma in my name, in any university or other tertiary institution and, to the best of my knowledge and belief, contains no material previously published or written by another person, except where due reference has been made in the text. In addition, I certify that no part of this work will, in the future, be used in a submission in my name, for any other degree or diploma in any university or other tertiary institution without the prior approval of the University of Adelaide and where applicable, any partner institution responsible for the joint-award of this degree.

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May Thwin
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DEDICATION

To my father.