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Cranial distraction osteogenesis for syndromic craniosynostosis: Long-term follow-up and effect on postoperative cranial growth

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Summary *Background:* Although cranial distraction osteogenesis (CDO) is beneficial, few studies have reported on detailed operative procedures and postoperative cranial growth. Herein, we demonstrated the objective effectiveness of CDO in younger infants.
Methods: The study included infants who underwent primary cranial distraction for craniosynostosis. Infants who had hydrocephalus were excluded and those who underwent additional osteotomy surgeries were analysed before the subsequent procedures. The infants' computed tomography data were analysed using Mimics® software (Materialise, Leuven, Belgium) to calculate the cranial volumes and compare them with the Abbott curve for a normal population. We defined cranial growth gap as the difference between the subject data and normal infant data to demonstrate the perioperative effects on cranial growth.
Results: CDO was performed in 10 infants. The mean infant age at the time of surgery was 6.4 months (range, 24—61 months) and the mean duration of postoperative follow-up was 38.9 months (range, 24—61 months). Five infants with Crouzon syndrome and five with Apert's syndrome were included. All infants showed postoperative cranial growth, but cranial growth gap showed postoperative declines for a certain period, indicating cranial growth suppression immediately following expansion. At the last follow-up, all cases were within ±2 standard

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deviation (SD) compared with the normal population. We derived a formula to predict the CDO target volume using the declining cranial growth gap curve.
Conclusions: CDO was applicable and suitable for younger infants requiring aggressive cranial expansion.

The ultimate goals of treatment for craniosynostosis are relieving cranial constriction to allow functional brain development and improving or maintaining cosmetic appearance. Various surgical techniques such as fronto-orbital advancement (FOA) or total calvarial reconstruction have been described for treating craniosynostosis, which are standard procedures for craniosynostosis because of their postoperative effects on the cranial vault.[1–4] However, compared to conventional FOA, cranial distraction osteogenesis (CDO) allows gradual bone tissue regeneration in the distracted area and coincident soft-tissue expansion, indicating that all bones are totally vascularised from the attached dura. Additionally compared with FOA or total calvarial reconstruction patients, CDO patients had significantly less intra-operative bleeding and shorter operating times.[5] Considering these merits, we suggest CDO as a standard procedure for cranioplasty, especially for younger syndromic infants who often show severe cranial deformities and require aggressive cranial enlargement.[5,6] To prove these potential merits, we conducted the present study on young infants with syndromic craniosynostosis and also describe our CDO procedure along with long-term postoperative changes in cranial volume. Furthermore, we derived a formula to determine the target volume based on the operative age of the infant.

Materials and methods

Patient cohort

The study included infants who underwent primary cranial distraction for craniosynostosis at Osaka City General Hospital between the years 2000 and 2008. CDO was indicated for infants who were diagnosed with at least one synostosis on computed tomography (CT) with clinical symptoms or signs such as papilloedema or digital markings on skull radiographs. All surgeries were performed in close collaboration between the departments of Pediatric Neurosurgery and Plastic and Reconstructive Surgery. Infants were excluded from this study if their operative age was >1 year; if they had non-syndromic craniosynostosis, hydrocephalus or pansynostosis; if the scheduled elongation was not completed; or if no CT data were available. Infants who subsequently underwent additional osteotomy surgeries such as secondary or more cranial remodellings and facial osteotomy were analysed before the secondary procedures. Our protocol was approved by the Institutional Review Board.

Surgical technique

A skin incision was made in a zigzag or lazy S-shaped fashion followed by dissection of the pericranial layer. As simulated with a three-dimensional (3D) cranial model before surgery, osteotomy was initiated by drilling small cranial burr holes, continued through the lateral orbital rim and the orbital roof, and was completed above the nasion bilaterally. If there were severe temporal protrusions or sphenoid ridges, they were resected sufficiently to inhibit early re-osteogenesis. The distraction device (Medical U&A, Osaka, Japan) comprises three parts: one movable bar and two handles. The handles are U-shaped, which allow grasping the bone edges, and have two screws to secure the fixation. Four titanium devices were applied: two to the parietal

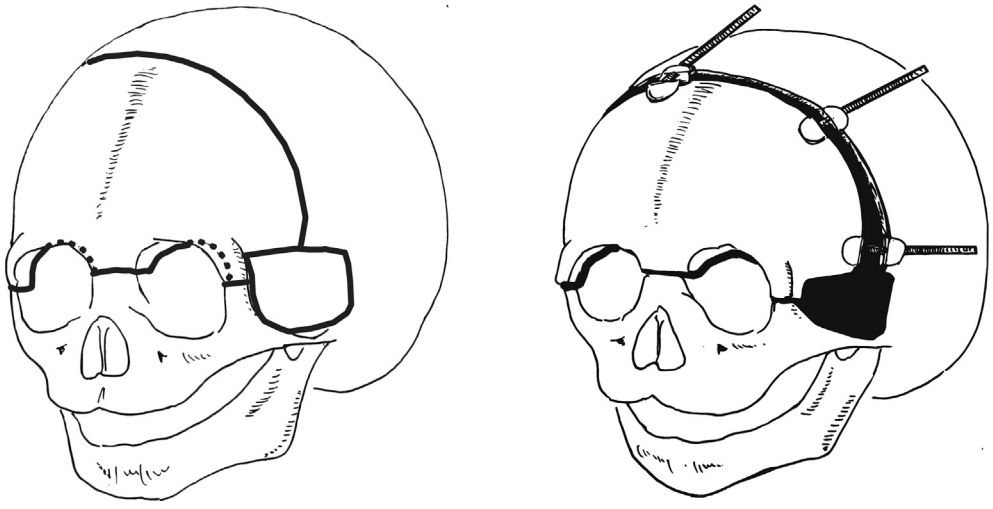

Figure 1 Cranial distraction osteotomy design. If severe temporal bulging and sphenoid ridges are present, they should be resected.