Transsutural distraction osteogenesis for 285 children with craniosynostosis: a single-institution experience

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OBJECTIVE Although distraction osteogenesis (DO) requires a secondary procedure in the surgical correction of craniosynostosis, it is relatively simple, requires less transfusion, results in a shorter intensive care unit stay, and is quite safe. Because of these positive factors, various DO techniques have been developed. However, there is disagreement regarding the superiority of DO. The authors reported on a new DO technique, transsutural DO (TSDO), 6 years ago that was performed in 23 patients over a period of 6 months, and it continues to be used at the present time. In this paper the authors report the results of TSDO performed in 285 patients with craniosynostosis over a period of 6 years at a single institution.

METHODS TSDO consists of a simple suturectomy of the pathological suture followed by direct distraction of the suturectomy site only. Types of TSDO conducted included sagittal TSDO in 95 patients, bicoronal in 14, unilateral coronal in 57, lambdoid in 26, metopic in 13, multiple in 19, syndromic in 33, and secondary in 28. The mean age ($\pm$ SD) of the patients was 19.4 ± 23.0 months, and mean follow-up was 39.5 ± 21.0 months.

RESULTS The mean operating time was 115 ± 43 minutes, and mean anesthesia time was 218 ± 56 minutes. The mean transfusion volume of red blood cell components was 48 ± 58 ml, and mean transfusion volume of fresh-frozen plasma was 19 ± 35 ml. Total transfusion volume was significantly less in infants younger than 12 months of age and in children with lower lumbar puncture pressures ($p < 0.05$). Complications included 1 (0.4%) death from postoperative acute pneumonia after a distractor removal operation and 23 (8%) surgical morbidities comprising 10 revisions (3.5%) and 13 early removals of distractors (4.6%).

CONCLUSIONS TSDO is a simple, effective, and safe method to use for treating all types of craniosynostosis. Some morbidity was experienced in this study, but it may be attributed to the learning curve of the technique.

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KEY WORDS complications; cranial expansion; craniofacial; craniosynostosis; distraction osteogenesis; surgical technique

Craniotomy for correction of craniosynostosis was introduced in the 1890s and has undergone many changes, from linear strip craniectomy to the more complicated total cranial remodeling of the 1960s.¹⁶ Since the 1990s, minimally invasive surgical techniques have become more popular with the advent of endoscopic linear strip craniectomy and the emergence of the distraction osteogenesis (DO) method.^{7,11} After a period of 15 years, the patient age limitation for performance of endoscopic linear strip craniectomy has increased to up to 1 year of age, after the molding helmet was approved for postoperative use. Another limitation of the DO method with re-

gard to the later spring and distractor applications is that to date their use in large numbers of patients has not been reported.^{1,2,4,5,8–10,12,18,19}

The distractor DO technique permits immediate postoperative control of the latency period, the distraction rate, and the activation period. The subsequent control of the distractor activation rate, in turn, allows control of the distractor vector. This advantage over spring DO is offset by the fact that distractor DO entails external exposure of the distractor, which does not occur in the spring DO method. Therefore, distractor DO cannot be deemed definitely superior to the spring DO technique.^{1–3,5,9,10,12,18–20} Despite the

ABBREVIATIONS DO = distraction osteogenesis; ICP = intracranial pressure; LP = lumbar puncture; TSDO = transsutural DO.

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many advantages of both the spring and distractor DO surgical techniques, popular use of these 2 methods remains limited because of their shortfalls. However, recent reports related to the increased incidence of postsurgical complications that accompany spring DO suggest that it may not be clearly advantageous over total cranial remodeling, and therefore further validation via large-scale studies of distractor DO has been suggested.¹³

The authors present the results of 285 patients with craniosynostosis who underwent the most simple distractor DO procedure to date, the transsutural DO (TSDO) method, during the period from the end of 2007 to the end of 2013.

Methods

Selection of Patients

During the 6-year period from November 2007 to October 2013 at Ajou University Hospital, 285 children with craniosynostosis (159 boys, 126 girls) underwent TSDO and were selected for inclusion in this study. The Ajou University institutional review board approved this study. The 285 patients included 95 (33.3%) with sagittal craniosynostosis, 57 (20%) with unilateral coronal craniosynostosis, 14 (4.9%) with bicoronal craniosynostosis, 26 (9.1%) with lambdoid craniosynostosis, 13 (4.6%) with metopic craniosynostosis, 19 (6.7%) with multiple craniosynostosis, 33 (11.6%) with syndromic craniosynostosis, and 28 (9.8%) with secondary craniosynostosis (Table 1). The 33 children with syndromic craniosynostosis were classified as having Crouzon ($n = 19$), Pfeiffer ($n = 2$), Apert ($n = 2$), simultaneous Pfeiffer and Apert ($n = 1$), Meunke ($n = 3$), Beare-Stevenson ($n = 1$), Sathre-Chotzen ($n = 4$), and Jackson-Weiss syndromes ($n = 1$). Lumbar puncture (LP) was performed over continuous intracranial pressure (ICP) monitoring at the request of childrens' parents, because of the lower risks and the simplicity of the method. Because of possible measurement errors and parents' emotional issues that could ensue due to the child's pain or resistance and crying, general anesthesia was chosen for use during the LP procedure in the present study. Therefore, preoperative LP pressure measurements were obtained by monitoring the end tidal pressure of carbon dioxide preoperatively or immediately before surgery after anesthesia

induction. The postoperative LP pressure was measured during distractor removal surgery, either preoperatively or immediately after surgery while the patient was still anesthetized.

We also studied the mean birth and preoperative body weight and the mean head circumference standard deviations (z score), which are important developmental factors in children.

TSDO Operative Technique

The basic principle of TSDO is dissection and distraction of the pathological suture, which can be applied to all types of craniosynostosis.²² Therefore, surgical techniques for sagittal, coronal, and lambdoid suture craniosynostosis consisted of resection of the pathological suture and placement of the distractor followed by DO (Fig. 1). For patients with metopic craniosynostosis, the surgical technique was modified according to the patients' coronal suture mobility (Fig. 2). For children with multiple, syndromic, and secondary craniosynostosis, various DO methods were applied simultaneously (Fig. 3).

Children with metopic craniosynostosis who were less than 4–6 months of age and had excellent coronal suture mobility by palpation of both frontal bones during surgery received resection of the metopic craniosynostosis only and placement of 1–2 distractors for DO on the metopic suture resection site (Fig. 2A–C). For children 4–9 months of age with slightly good coronal suture mobility, the bicoronal suture and the metopic suture were resected followed by distractor placement and DO (Fig. 2D–F). In those patients older than 6–9 months who had poor coronal suture mobility, distractors were applied to both metopic and bicoronal sutures (Fig. 2G–J). DO provided expansion of the frontal space in the first group of patients with distractor placement only in the metopic suture. Distractors placed on the metopic suture only was followed by bicoronal suture resection in the second group of patients. In the third group of patients where distractors were placed in both the metopic and bicoronal sutures, DO with the bicoronal distractor was performed first. Removal of the bicoronal distractors was performed after widening of the frontal base was confirmed at 2–3 weeks followed by further expansion of the frontal space by DO of the metopic suture distractor.

TABLE 1. Demographic data of 285 children who underwent TSDO*

Type of Synostosis	No. of Patients (M:F)	Mean Gestational Age $\pm$ SD (wks)	Mean Birth Weight $\pm$ SD (g)	Mean Age $\pm$ SD (mos)	Mean Follow-Up Duration $\pm$ SD (mos)
Sagittal	95 (66:29)	38.3 $\pm$ 2.5	3083 $\pm$ 626	22.6 $\pm$ 27.4	40.1 $\pm$ 20.7
Bicoronal	14 (4:10)	37.9 $\pm$ 3.0	2795 $\pm$ 730	5.3 $\pm$ 5.9	33.8 $\pm$ 19.9
Unilateral coronal	57 (22:35)	38.6 $\pm$ 1.9	3116 $\pm$ 464	12.0 $\pm$ 14.0	41.3 $\pm$ 21.3
Lambdoid	26 (15:11)	38.8 $\pm$ 1.5	3347 $\pm$ 521	10.8 $\pm$ 11.5	41.9 $\pm$ 19.3
Metopic	13 (9:4)	36.8 $\pm$ 4.1	2744 $\pm$ 733	8.2 $\pm$ 10.9	33.4 $\pm$ 20.7
Multiple	19 (8:11)	38.7 $\pm$ 2.5	2876 $\pm$ 604	33.2 $\pm$ 28.4	36.9 $\pm$ 20.7
Syndromic	33 (19:14)	38.6 $\pm$ 1.6	3182 $\pm$ 368	20.5 $\pm$ 24.9	41.8 $\pm$ 22.7
Secondary	28 (16:12)	35.6 $\pm$ 5.1	2293 $\pm$ 1030	33.0 $\pm$ 18.3	36.0 $\pm$ 22.9
Total	285 (159:126)	38.1 $\pm$ 2.9	3004 $\pm$ 677	19.4 $\pm$ 23.0	39.5 $\pm$ 21.0

* Boldface values are statistically significant ($p < 0.05$).

FIG. 1. The 3D reconstructed CT images after TSDO according to craniosynostosis types. The pathological suture has been surgically removed by TSDO, and the height of the distracted skull flap is different from that of the adjacent nondistracted skull flap (red arrows). **A:** Bicoronal suture TSDO. **B:** Lambdoid suture TSDO. **C:** Unilateral coronal suture TSDO. **D:** Sagittal suture TSDO. Figure is available in color online only.

Patients with secondary and multiple craniosynostosis included those with sagittal and lambdoid DO, and coronal sagittal lambdoid DO. For the combined application of simultaneous sagittal lambdoid DO, the bicoronal, sagittal, and bilambdoid sutures underwent craniectomy followed by distractor placements at the sagittal and lambdoid sutures. For the combined application of coronal sagittal lambdoid DO, craniectomy was performed at the bicoronal suture, sagittal suture, and bilambdoid suture, followed by distractor placement at each of these suture sites. Initially, the sagittal lambdoid DO method was conducted, which resulted in relatively inadequate coronal suture expansion, after which coronal sagittal lambdoid DO was conducted to induce wider and more generalized expansion of the whole cranium (Fig. 3).

After surgery, activation was performed depending on the ICP, the severity of cranial asymmetry, or Chiari malformation. After a latency period of 0–10 days, depending on the patient's age, ICP, volume of transfusion, general

postoperative condition, postoperative scalp tension, and subdural space size, the distractors were activated 0.15–1.0 mm/day, for an activation period of 15–74 days. Three-dimensional reconstructed CT was conducted every 1–3 weeks after activation, depending on daily activation distance, subdural space volume, and patient age to confirm adequate advancement of the skull and underlying brain expansion without increased subdural space widening, after which activation was stopped followed by a 1–4 month consolidation period. Thereafter, the plates, screws, and distractors used in the distraction and fixation were all removed.

Satisfaction after surgery was investigated by telephone survey 1 year after surgery; 178/285 (62.5%) responded to our questionnaire. A nurse contacted the parents of the patients, and the question and answers were sorted according to the 4 Whitaker classification categories (Table 2)²⁷ and the 7 Sloan classification categories (Table 3).²⁵

Results

Demographics

Among the total 285 children, the male to female ratio was 159:126, showing a greater proportion of male patients. This ratio tendency was the same for sagittal, lambdoid, and metopic craniosynostosis but was reversed for unicoronal and bicoronal craniosynostosis. The mean gestational age of the patients was 38.1 ± 2.9 weeks. This was similar for all groups except patients with secondary craniosynostosis, which had a statistically lower mean age of 35.6 ± 5.1 weeks. The mean birth weight of the children was 3004 ± 677 grams, which was significantly greater in the lambdoid craniosynostosis group, and significantly less in the secondary craniosynostosis group. The mean age of the patients at the time of surgery was 19.4 ± 23.0 months, which was significantly lower in the bicoronal, unicoronal, metopic, and lambdoid craniosynostosis groups. The mean follow-up duration was 39.5 ± 21.0 months, which was not significantly different among the study population. The demographic data of patients according to craniosynostosis type are shown in Table 1. The mean standard deviation (z score) for head circumference at birth was -0.4 ± 1.6 , and the mean standard deviation (z score) for preoperative body weight was -0.95 ± 1.4 .

Operative Description

The mean operative time was 115 ± 43 minutes, and the mean anesthesia time was 218 ± 56 minutes. Surgery duration and anesthesia time were significantly shorter for lambdoid and sagittal craniosynostosis surgery. The mean packed red blood cell infusion volume was 48 ± 58 ml,

TABLE 2. Four-category classification of operative results introduced by Whitaker et al.²⁷

Category	Definition
I	Patients in whom no surgical revisions were considered advisable or necessary by the surgeon, patient, or family.
II	Soft tissue or minor bone contouring revisions were desirable, whether or not they were actually performed.
III	Patients in whom major secondary osteotomies or bone grafting procedures were needed or performed.
IV	Patients in whom a major craniofacial procedure, duplicating or exceeding the extent of the original surgery, was or would be necessary.

FIG. 2. The 3D reconstructed CT images before and after TSDO for metopic suture craniosynostosis. **A–C:** Method for metopic suture distraction with distractors after metopic craniosynostosis resection only in a child with metopic craniosynostosis less than 4–6 months of age with good coronal suture mobility. **D–F:** Method for metopic suture distraction with 1 distractor after metopic craniosynostosis resection as well as bicoronal suture resection in a child with metopic craniosynostosis 4–9 months of age with slightly good coronal suture mobility. **G–J:** Method for metopic suture distraction of both metopic and bicoronal sutures in a child with metopic craniosynostosis greater than 6–9 months of age with poor coronal suture mobility. Figure is available in color online only.

mean fresh-frozen plasma infusion volume was 19 ± 35 ml, and mean platelet-concentrated plasma infusion volume was 6 ± 17 ml. The patients with lambdoid craniosynostosis received significantly less infusion of all components. Intraoperative details of patients according to the

subtype of craniosynostosis are shown in Table 4. A comparison between children less than 12 months of age and those greater than 12 months of age with regard to mean operation time was 104.9 ± 47.0 minutes and 128.9 ± 45.5 minutes, respectively, which is statistically different ($p <$

TABLE 3. Classification of surgical results after reconstruction for craniosynostosis by Sloan et al.²⁵

Class	Definition
1	Good to excellent correction, w/ no visible or palpable irregularity.
2	Good to excellent correction w/ palpable but not visible irregularity (e.g., a palpable, but not visible, surgical wire, plate, or bony irregularity), not requiring reoperation.
3	Good to excellent correction w/ visible irregularity (e.g., a visible prominence from a surgical wire or plate, or a visible bony spicule or defect that does not compromise the overall correction), not requiring reoperation.
4	Good to excellent correction w/ visible or palpable irregularity requiring reoperation (e.g., a surgical plate requiring removal).
5	Compromised overall correction, but not severe enough to require reoperation (e.g., slight forehead asymmetry).
6	Compromised overall correction requiring reoperation.
7	Compromised overall correction, believed to require reoperation by the surgeon, but family declines further surgery.

FIG. 3. The 3D reconstructed CT images before and after TSDO for sagittal and lambdoid suture distraction, and sagittal, lambdoid, and coronal distraction. **A and B:** For combined application of sagittal DO and bilateral lambdoid DO, simultaneous bicoronal suture, sagittal suture, and bilambdoid suture craniectomy are performed, followed by sagittal and lambdoid suture TSDO. **C and D:** For combined application of bilateral coronal DO, sagittal DO, and bilateral lambdoid DO, simultaneous bicoronal suture, sagittal suture, and bilambdoid suture craniectomy is performed followed by coronal, sagittal, and lambdoid suture distractor placement. Preoperative images show unfused sutures (blue arrows) and fused sutures (red arrows). Figure is available in color online only.

0.05). Total transfusion volume was also statistically different in these age groups (59.7 ± 65.8 ml and 87.7 ± 113.7 ml, respectively, $p < 0.05$).

An average of 3.3 ± 0.8 distractors were applied, with a mean latency period of 3.2 ± 1.4 days, mean activation rate of 0.5 ± 0.3 mm/day, mean activation period of 33.6 ± 25.3 days, mean total distraction length of 15.2 ± 7.4 mm, and a

mean consolidation period of 49.6 ± 20.4 days. The mean activation period was statistically shorter for the children with lambdoid and metopic craniosynostosis, while it was significantly longer in patients with secondary craniosynostosis. The mean consolidation period was significantly longer in those patients with secondary craniosynostosis (Table 5).

LP Pressure, Transfusion Volume, and Head Circumference

The mean preoperative LP pressure was 22.3 ± 8.0 cmH₂O, and the mean postoperative LP pressure was reduced to 17.5 ± 5.3 cmH₂O with a significant statistical difference ($p < 0.001$). There was no significant difference in the mean preoperative LP pressure among the subgroups, but the mean postoperative LP pressure was significantly lower in patients with secondary craniosynostosis.

Although we did not observe any difference in operation time according to LP pressure, the mean total transfusion volume was significantly less (56.6 ± 78.8 ml) in the low LP pressure group with less than 20 cmH₂O compared with 82.6 ± 97.0 ml in the high LP pressure group with greater than 20 cmH₂O ($p < 0.05$). The mean preoperative head circumference z score was -0.2 ± 2.0 and the mean postoperative head circumference z score increased to 0.4 ± 2.1 with a significant statistical difference ($p < 0.001$). The mean postoperative head circumference z scores were statistically increased in all craniosynostosis subgroups (Table 6).

Surgical Outcomes

Immediate correction of abnormal head contours was observed in all patients after distraction. Complications included 1 death (0.4%), 10 revisions (3.5%), and 13 early removals of distractors (4.6%). The child who died was a patient with Crouzon syndrome with multiple craniosynostoses who experienced minimal infection that was managed by distractor removal, wound curettage, and irrigation. The surgery duration for this patient was 70 minutes, anesthesia duration was 210 minutes, no transfusions were administered, and there were no other distinguishing operative characteristics. However, the patient unfortunately suffered severe postoperative acid-base imbalance and re-

TABLE 4. TSDO surgeries in 285 children*

Type of Synostosis	Operative Time (min)	Anesthetic Time (min)	Volume of Transfusion (ml)		
			P-RBC	FFP	P-Conc
Sagittal	99 ± 41	197 ± 45	48 ± 70	19 ± 38	6 ± 18
Bicoronal	145 ± 26	260 ± 32	61 ± 60	21 ± 43	2 ± 8
Unilateral coronal	113 ± 33	218 ± 44	33 ± 31	14 ± 29	4 ± 16
Lambdoid	69 ± 20	166 ± 43	15 ± 25	6 ± 15	1 ± 6
Metopic	159 ± 45	270 ± 52	89 ± 52	25 ± 41	12 ± 20
Multiple	137 ± 53	237 ± 64	64 ± 55	22 ± 32	7 ± 15
Syndromic	154 ± 61	268 ± 55	66 ± 58	33 ± 44	10 ± 20
Secondary	121 ± 40	219 ± 53	50 ± 63	19 ± 29	10 ± 25
Total	115 ± 43	218 ± 56	48 ± 58	19 ± 35	6 ± 17

FFP = fresh-frozen plasma; P-Con = platelet-concentrated; P-RBC = packed red blood cell.

* All values given as means ± SD. Boldface values are statistically significant ($p < 0.05$).

TABLE 5. Distraction procedures in 285 cases with TSDO

Type of Synostosis	No. of Distractors	Latency Period (days)	Activation Rate (mm/day)	Activation Period (days)	Total Distraction Length (mm)	Consolidation Period (days)
Sagittal	3.3 ± 0.7	3.1 ± 1.1	0.6 ± 0.3	32.3 ± 11.6	16.8 ± 6.1	49.0 ± 17.8
Bicoronal	4.0 ± 0.0	3.7 ± 2.0	0.4 ± 0.1	31.6 ± 16.7	11.0 ± 5.3	43.0 ± 12.5
Unilateral coronal	2.7 ± 0.6	3.0 ± 0.5	0.4 ± 0.3	32.9 ± 11.4	12.8 ± 5.6	49.1 ± 14.2
Lambdoid	2.7 ± 0.7	3.3 ± 1.5	0.5 ± 0.1	24.3 ± 7.5	12.7 ± 4.3	50.2 ± 9.9
Metopic	3.5 ± 1.1	3.6 ± 1.4	0.5 ± 0.2	15.0 ± 6.7	7.9 ± 4.1	31.5 ± 31.2
Multiple	3.7 ± 0.7	3.2 ± 0.9	0.7 ± 0.6	31.3 ± 14.5	19.4 ± 16.2	57.2 ± 18.5
Syndromic	3.7 ± 0.8	3.1 ± 1.1	0.6 ± 0.3	35.3 ± 14.9	17.8 ± 7.1	48.4 ± 31.6
Secondary	3.6 ± 0.6	3.9 ± 2.7	0.5 ± 0.3	43.3 ± 20.1	16.9 ± 5.2	59.1 ± 23.1
Total	3.3 ± 0.8	3.2 ± 1.4	0.5 ± 0.3	33.6 ± 25.3	15.2 ± 7.4	49.6 ± 20.4

* All values are means ± SDs. Boldface values are statistically significant ($p < 0.05$).

spiratory distress syndrome, and subsequently died 1 day after surgery.

Ten procedure revisions were necessary and included 4 distractor malfunctions, 2 distractor breakages, 2 wound debridements and irrigations for pus discharge, 1 wound dehiscence, and 1 CSF leakage. Thirteen early distractor removals consisted of 9 wound problems with pus discharge, 2 transient sixth cranial nerve palsies that were relieved completely, 1 wound dehiscence, and 1 large incidental traumatic subgaleal hematoma that developed from an injury due to a fall during the consolidation period. The 14 children with minor problems who did not require further surgical intervention included 8 with minimal pus discharge, 3 with distractor malfunction, 2 with wound pain managed with medication, and 1 with an immediate postoperative seizure that developed 2 hours after surgery in the recovery room (Table 7).

All 178 telephone survey responders were Whitaker Category I, and according to the 7 Sloan classifications, there were 123 (69%) of 178 Category 1, 52 (29.2%) Category 2, 2 (1.1%) Category 3, and 1 (0.6%) Category 4 patients.

Discussion

Advantages and Limitations of the TSDO Operative Technique

The advantages of DO are presently well known. This

technique permits expansion not only of the cranial bone but also of the scalp, which results in a larger expansion volume, and also allows control and regulation of the distractor DO direction, distance, and distraction speed.^{1-3,5,9,10,12,13,18-22} Other noteworthy merits of TSDO are the simplicity of the surgical procedure as in the spring DO, the ability of the procedure to resemble as closely as possible normal cranial growth that ensues from resection only of the affected sutures.²² The spring DO procedure results in expansion of the cranium immediately after surgery, and therefore there is no latency period. Also, the distraction direction vector cannot be adjusted, and thus there is an increased risk of complications, such as bleeding, CSF leakage, subdural space widening, and unwarranted less expansion or overexpansion of the cranium with undesirable cosmetic consequences.^{12,13} At present, no studies or information exist on the proper and safe expansion distance in any of the DO surgical techniques, including TSDO, so that determination of this factor is difficult.

Comparison of the 3 surgical methods (distraction including TSDO, strip craniectomy with molding helmet, and classical total calvarial remodeling with absorbable plate fixation) showed that the costs of all 3 types of surgery and admission are relatively low in our country (Korea) compared with that in other countries. The costs are approximately US \$600 for each type of surgery with total costs for all 3 surgeries ranging from US \$5000 to \$10,000, because of Korean national health insurance cov-

TABLE 6. Preoperative and postoperative changes of LP and head size in 285 children with craniosynostosis

Type of Synostosis	Mean LP Pressure ± SD (cmH ₂ O)			Mean Head Size ± SD (z scores)		
	Preop	Postop	p Value	Preop	Postop	p Value
Sagittal	23.7 ± 8.1	17.0 ± 4.9	<0.001	1.1 ± 1.5	1.5 ± 1.5	<0.001
Bicoronal	21.7 ± 6.6	19.2 ± 6.9	0.13	-1.2 ± 1.4	-0.2 ± 1.0	0.003
Unilateral coronal	21.7 ± 6.3	19.2 ± 4.8	0.02	-0.4 ± 1.3	0.2 ± 1.3	<0.001
Lambdoid	25.8 ± 0.7	17.5 ± 5.7	<0.001	0.6 ± 1.0	1.3 ± 1.0	<0.001
Metopic	20.8 ± 6.5	15.7 ± 2.9	0.02	-0.5 ± 1.2	0.1 ± 1.1	0.008
Multiple	24.2 ± 8.9	16.1 ± 4.9	0.007	-0.2 ± 1.3	0.5 ± 1.3	0.01
Syndromic	25.4 ± 9.2	20.8 ± 5.7	0.008	-0.7 ± 1.5	0.4 ± 1.7	<0.001
Secondary	21.7 ± 9.6	13.4 ± 4.0	<0.001	-3.6 ± 2.7	-3.0 ± 3.0	<0.001
Total	22.3 ± 8.0	17.5 ± 5.3	<0.001	-0.2 ± 2.0	0.4 ± 2.1	<0.001

TABLE 7. Complications of TSDO operations in 285 patients with craniosynostosis

Complication	No. of Patients (%)
Death	
Acute fatal pneumonia	1 (0.4)
Morbidity	
Wound revision	10 (3.5)
Distractor malfunction	4
Distractor breakage	2
Pus discharge managed w/ wound debridement & irrigation	2
Wound dehiscence	1
CSF leakage	1
Early removal of distractor	13 (4.6)
Pus discharge from wound	9
Sixth cranial nerve palsy (transient)	2
Wound dehiscence	1
Incidental traumatic subgaleal hematoma	1
Nonoperative care	14 (4.9)
Minimal pus discharge	8
Distractor malfunction	3
Wound pain	2
Postoperative seizure	1

erage. The cost of materials and equipment is high; the cost of 1–4 distractors is approximately US \$800–\$3000, molding helmet US \$3000, and absorbable implants US \$10,000–\$15,000. On average, the cost of the distraction method is the lowest but is similar to that of strip craniectomy with a molding helmet, while the cost of the classic total calvarial remodeling with absorbable plate fixation is the most expensive. However, all these costs may differ from country to country and in different regions.

At our institution, various methods were used to correct craniosynostosis until 2008, but from 2009 onwards the majority of patients with craniosynostosis have undergone TSDO surgery because of its safety, effectiveness, and aesthetics, with the exception of those less than 2 months of age and those who have incomplete bone fusion after primary nondistractor surgery.

Results of the TSDO Operative Technique in 285 Children With Craniosynostosis

While our results agree with results from previous series that the proportion of children with bicoronal craniosynostosis is relatively low (5.3%), we were not able to discern the reason for the disparity from the 9.3%–25% incidence reported by other investigators.^{9,12,22} The male to female ratio showed a higher proportion of females with bicoronal and unilateral coronal craniosynostosis, which is similar to results in past studies.¹² Our patients with secondary craniosynostosis had lower mean gestational ages and mean birth weights, which we presume is a natural phenomenon due to overall brain and systemic underdevelopment.

The mean age of the children who underwent surgery

was 19.4 ± 23.0 months, which is earlier than the 30 ± 25 months of Selo et al.²⁴ and the 59 months of Nonaka et al.¹⁹ However, it is similar to the 14.42 months of Kim et al.,⁹ the 16 months of Cho et al.,² and the 16.3 ± 8.7 months of Akai et al.¹ It is later than the 6 months of the national study conducted by Nguyen et al.¹⁷ in the US. These differences may be due to the structure of the medical patient transfer system in each country. The mean age of the patients was lower in bicoronal, unilateral coronal, and metopic craniosynostosis, and we attribute this difference to the abnormal head appearance and thus early detection of disease.

Comparison of previous mean operative times shows that while Lauritzen et al.¹² reported a mean of 97–215 minutes for spring DO, Kim et al.,⁹ Park et al.,²¹ and Park et al.²² observed similar mean times of 248 minutes, 251 minutes, and 144 minutes, respectively, for the distractor DO procedure. The current study shows the shorter mean operative time of 115 minutes. Although Kim et al.⁹ showed that there was no difference in operative time according to the different types of craniosynostosis, we were able to demonstrate significantly shorter surgical times for patients with sagittal and lambdoid craniosynostoses. In contrast to the 178 ml (Kim et al.⁹) and 330 ml (Park et al.²¹) mean transfusion volumes in previous studies, our study showed a markedly decreased mean volume of 72 ml. This volume was especially low in children who had undergone TSDO for lambdoid craniosynostosis. In addition, the shorter operative time and lower total transfusion volume in our study patients who were less than 12 months of age might be attributed to the smaller field of surgery in this study. Our results agree with results from past series that have suggested that craniosynostosis surgical duration is proportional to blood loss volume, and thus may be considered a factor in decreasing transfusion volume.²³

While there was no statistical difference in postoperative distraction variables pertaining to the latency period for each type of craniosynostosis, the DO activation rate for bicoronal and unilateral coronal craniosynostosis was slower due to their particular structural anatomical characteristics. The activation period for the early surgery lambdoid and metopic craniosynostosis was relatively shorter as a consequence of the short distraction length. In children with secondary craniosynostosis, the DO was prolonged and had subsequent prolonged consolidation periods.

According to Sloan et al.,²⁵ among the 250 patients with craniosynostosis who underwent surgery, 115 were surveyed 1 year after surgery, and 38 (33%) were above Category 5, while 14 (12.2%) who required reconstructive plastic surgery were Category 6. In another report by Esparza et al.⁶ of a series of 283 patients, 95 (33.6%) were above Whitaker classification Category 2 requiring cosmetic surgery, while our results did not show any patients with Category 5 or higher after more than 1 year of follow-up, demonstrating better outcomes than those of previous authors.^{6,27}

Perioperative LP Pressure and Head Size

With the exception of patients with bicoronal craniosynostosis, all craniosynostosis subtypes demonstrated a

statistically significant decrease in postoperative LP pressure, with no difference between the different types of craniosynostosis. LP pressure was shown to be particularly lower in children with secondary craniosynostosis after surgery.^{21,22} In the present study, the subgroup of patients with LP pressures less than 20 cmH₂O demonstrated no significant difference in operative time but significantly less mean total transfusion volume ($p < 0.05$) compared with those with greater than 20 cmH₂O of LP pressure. We believe the reason for this difference in total transfusion volume is due to the increased volume of drainage via the extracranial pathway, which results from increased ICP, so that greater blood loss occurs when incising the scalp and removing the fused skull suture.

The head size was significantly increased in all patients after TSDO in our series. Children with sagittal and lambdoid craniosynostoses had larger preoperative mean cranial sizes compared with other types of craniosynostosis, while patients with secondary craniosynostosis had smaller craniums before surgery as well as postoperatively. We interpret these results as a consequence of the absence of LP pressure correlation between preoperative head size in sagittal and lambdoid craniosynostosis patients and others so that the same increased ICP will result in relatively greater head size for patients with sagittal and lambdoid craniosynostoses for protection reaction roles. Our observation of the development among our patients has also shown that those with sagittal and lambdoid craniosynostosis demonstrated more favorable outcomes compared with outcomes for other patients.

Furthermore, the early efficacy of craniosynostosis surgery has not yet been substantiated and is therefore still subject to scrutiny, but most cases of craniosynostosis surgery result in statistical lowering of the LP pressure and increase in head size, suggesting that the surgical procedure has a beneficial effect on craniosynostosis.

Complications of TSDO Operations

The medical literature reports that mortality after craniosynostosis surgery is approximately 1%, but as yet no mortality report exists pertaining to DO.¹⁷ In our experience, we did not observe any patient deaths directly attributable to the distractor procedure but rather a death after secondary surgery for distractor removal in a child with Crouzon syndrome and multiple craniosynostoses. This patient had undergone tracheostomy for respiratory distress; the intermittent aspiration leading to mild pneumonia that had continued from birth did not improve with time and led to distractor removal surgery. Intraoperative minimal distractor infection and the following wound curettage and irrigation resulted in slightly prolonged operative time and anesthesia duration but without any adverse events or transfusions. However, severe acid-base imbalance and rapid arterial oxygen pressure (PaO₂) level decline ensued after surgery with consequent severe pneumonia and death within 1 day postoperatively. We attribute this death probably to the surgical and anesthetic stress that caused severe aggravation of the already present pneumonia, and finally acute respiratory distress syndrome.

Past reports regarding complications of craniotomy or total calvarial remodeling for craniosynostosis correction

have consisted of intraoperative complications, such as extensive bleeding and air embolism, and postoperative complications that include epidural or subdural hemorrhages, dural defects or dural tearing, osteomyelitis, sepsis, and implant infections.^{5,6,13} In contrast, complications related to DO that have been reported are CSF leakage, distractor breakage, distractor malfunction, spring dislodgement, overcorrection, undercorrection, and subgaleal hemorrhage from delayed sinus tears.^{1-3,5,9,10,12,14,15,18-22} For spring DO, Lauritzen et al.¹² reported a study of 100 patients in 2008, and David et al.³ reported 75 patients in 2010. Investigations to date reporting distractor DO in more than 5 patients are few: 19 by Yonehara et al.³⁰ in 2003, 7 by Nonaka et al.¹⁹ and 6 by Cho et al.² in 2004, 5 by Komuro et al.¹⁰ in 2005, 9 by Akai et al.¹ and 11 by Nishimoto et al.¹⁸ in 2006, 14 by Kim et al.⁹ in 2007, 26 by Esparza et al.⁵ in 2008, 9 by Park et al.²⁰ and 6 by White et al.²⁸ in 2009, 23 by Park et al.²¹ in 2010, 10 by Serlo et al.,²⁴ 8 by Steinbacher et al.,²⁶ and 26 by Winston et al.²⁹ in 2011, with no series including more than 50 patients.

Moreover, studies that have reported DO complications in more than 10 patients are even fewer, and are summarized in Table 8. Among the studies that included more than 50 patients, complications consisted of repeat surgery due to mechanical problems such as distractor breakage, distractor malfunction, spring dislodgement in 1.3%–4% of patients who underwent spring DO, and this figure was 2.1% in DO patients treated by the authors, showing no difference between the spring DO and distractor DO techniques.^{3-5,9,12,18,21,29,30} Distractors that were initially used were thin and prone to breakage; however, after thick distractors were developed and introduced into surgery, breakage no longer occurred.^{14,21,22}

With regard to infection, considered the most serious complication of distractor DO, repeat surgery due to infection or a wound after spring DO has been shown to be 0%–4%, and early distractor removal for the same reasons has been reported as 0%–3.3%.^{3-6,9,12,18,21,29,30} In contrast, our incidence of repeat surgery for infection or a wound after distractor DO was 1.1%, and the incidence of early distractor removal for the above reasons was 3.9% in this study. Therefore, we hypothesize that there is no significant difference between distractor DO and spring DO in terms of the incidence of infection.

In the present study, 1 child (0.4%) had CSF leakage and 2 (0.7%) had temporary sixth cranial nerve palsy after distractor DO, but no cranial nerve palsy has been reported in the literature after spring or distractor DO.^{3,12} Temporary cranial nerve palsy that occurred in our study was a consequence of adjusting the initial distraction rate of 0.5–1.5 mm/day and total distraction length of 30 mm/day, which did not occur again after the distraction rate and length were reduced from less than 0.5 mm/day and less than 15 mm/day, respectively.

Conclusions

The TSDO surgical technique proposed by the current authors was not accompanied by significant infectious complications and overcame mechanical limitations such as distractor breakage through additional device develop-

TABLE 8. Postoperative complications reported in the literature regarding patients who underwent cranial DO for craniosynostosis (≥ 10 operations)

Authors & Year	Type of Surgery (no. of patients)	Major Morbidity: Revision (%)					Minor Morbidity: Early Removal of Distracters (%)			
		Additional Surgery	CSF Leakage	Distractor or Spring Reposition	Infection or Pus Discharge	Wound Problem	TCNP	Infection or Pus Discharge	Wound Problem Except Pus	
Yonehara et al., 2003	Distractor DO (19)	1 (5.3)	0	0	0	0	0	0	0	
David et al., 2004	Spring DO (15)	0	0	0	0	0	0	0	0	
Nishimoto et al., 2006	Distractor DO (11)	0	0	2 (18.2)	0	0	0	2 (18.2)	0	
Kim et al., 2007	Distractor DO (14)	0	0	0	0	0	0	0	0	
Esparza et al., 2008	Distractor DO (26)	0	1 (3.8)	0	1 (3.8)	1 (3.8)	0	3	0	
Lauritzen et al., 2008	Spring DO (100)	6 (6)	0	4 (4)	0	0	0	0	2 (2)	
David et al., 2010	Spring DO (75)	0	0	1 (1.3)	1 (1.3)	2 (2.7)	0	1 (1.3)	0	
Park et al., 2010	Distractor DO (23)	0	0	2 (8.7)	0	0	0	0	0	
Winston et al., 2011	Distractor DO (26)	0	0	2 (7.7)	0	0	0	1 (3.8)	0	
Present study	Distractor DO (285)	1 (0.4)	1 (0.4)	6 (2.1)	2 (0.7)	1 (0.4)	2 (0.7)	9 (3.2)	2 (0.7)	

TCNP = temporary cranial nerve palsy.

ment research. Also, the above procedure was effective in producing cranial expansion and reducing ICP. Operative times and transfusion volumes were relatively reduced so that TSDO could be applied safely and effectively as a surgical method for the correction of craniosynostosis.

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Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: both authors. Acquisition of data: both authors. Analysis and interpretation of data: both authors. Drafting the article: both authors. Critically revising the article: both authors. Reviewed submitted version of manuscript: both authors. Approved the final version of the manuscript on behalf of both authors: Yoon. Statistical analysis: both authors. Administrative/technical/material support: both authors. Study supervision: both authors.

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