REVIEW ARTICLE doi: 10.2176/nmc.ra.2017-0006

Neurol Med Chir (Tokyo) 57, 217–224, 2017 Online April 12, 2017

Review of Past Reports and Current Concepts of Surgical Management for Craniosynostosis

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Abstract

The purposes of surgery for craniosynostosis are to release increased intracranial pressure and to normalize cranial shape. The procedure was developed from a simple strip craniectomy in practice which ranged from the removal of the fused suture before the 1960s to total calvarial remodeling after 1970s and later methods of the 1990s, such as distraction and its modifications. According to its history, craniofacial surgeons might be changing their procedures with more effective, than less invasive ways. Since the late 1990s, when the distraction was applied to the craniofacial surgery, the gradual expansion, in particular of the anterior cranium, common in Japan, has long been controversial until the Caucasians accepted its use for the posterior cranium. Currently, the method may revert to the old procedure because a more sophisticated and better morphological result can be obtained depending on the types of deformity, even if a little more invasive maneuver is required. In other words, if treatment can be performed in optimal time, the procedures that were developed in the last half a century should be altered to each condition.

Key words: craniosynostosis, suturectomy, intracranial pressure, distraction osteogenesis, mental development

Introduction

Craniosynostosis is the premature fusion of one or more sutures in either the cranial vault or anterior skull base, resulting in an abnormal head shape and intracranial pressure increasing in one-third of all cases that may have a mental mal-development.[1–5] This pathologic process occurs in approximately 1 in 2000 to 2500 births in Western countries and less in Orientals.[6] The first surgical treatment was reported in 1890s, with linear craniectomy for opening the fused suture, and had long been done by neurosurgeons, until Tessier started craniofacial surgery in 1960s, with a frontal bone repositioning for the purpose of cranial volume expansion.[7–9] Since this revolutionary medical event, the surgical procedure for the craniosynostosis has been developed in a variety of ways based on several ideas, by surgeons' intrepid and prudent "trial and error" up to the current time.[10–15] We sought to review the historical reports of our predecessors, discussing the transition of concepts and methods in Japan as well as in the rest of the world.

Received January 11, 2017; Accepted February 1, 2017

History of Craniosynostosis: Its Diagnosis, Management and Surgical Procedure

Except Hippocrates' first historical description of craniosynostosis in 100 BC, Sömmerring and Otto initially described in 1800 and in 1830, that premature cranial sutural fusion would result in deformity, and the etiology was thought to be based on either fetal or birth trauma.[16,17] In 1851, Virchow first introduced the term of 'craniosynostosis' and formulated what is today known as Virchow's law: there is a cessation of growth occurs in the direction perpendicular to that of the affected suture while growth proceeds in a parallel direction.[18] Based on the concept, the abnormal calvarial growth due to a premature fusion of the sutures, provided the basis for early operative treatment of craniosynostosis, with removing the offending suture in an attempt to release the constricted brain. In 1890, Lannelongue in Paris described bilateral strip craniectomies for the treatment of craniosynostosis, and Lane followed the intervention two years later in the United States.[19,20] Two years later, Jacobi reported alarming outcomes and high morbidity and mortality in 33, open strip craniectomies on craniosynostosis patients, due to major blood loss. The concept of brain-releasing

from contracted cranium was not wrong, but this surgery had not been performed until 1927, when Faber and Towne reported their successful more extensive craniotomy.[21] Since then the development of anesthetic and blood management over the years, has provided the opportunity for more difficult and advanced craniosynostosis surgery. The modern era of craniofacial surgery started in the 1960s with Tessier, who first established multidisciplinary craniofacial teams in Paris.[8,9] In 1967, he showed a procedure of fronto-orbital advancement with cranial vault remodeling, with reshaped removal bone pieces stabilizing back to the cranium and established new protocols that followed and consisted in Moss's functional matrix theory in 1959, and the concept of compensatory cranial vault growth by Vollmer(1984) and Delashaw(1989).[9,22–25]

On the other hand, similar groups of the craniosynostosis had been reported and classified in terms of each characteristic problem over the years. In 1906, although brachycephalic craniosynostosis with syndactyly had already been reported toward the end of the nineteenth century, the French pediatrician Apert is generally credited with describing the condition.[26,27] In 1912, Crouzon, a neurologist, reported the condition that is named after him.[27,28] Those syndromic diseases were suffered the skull base sutures' fusion besides cranium, ended with a facial retrocession or jaw deformities, that needs craniofacial surgeons to manage the middle third of the face after an operation of cranium in infants. As multidisciplinary teams developed all over the world, clinical geneticists became involved and studied inheritance patterns and syndromic features. In 1993 the first genetic lesion, a specific missense mutation in the MSX2 gene, was identified by Melville et al.[29] in a large family with autosomal dominant craniosynostosis, known as Boston type. This discovery launched molecular diagnostics by identifying a key gene in calvarial development. And in the late 1990s, some craniofacial anomalies, like Crouzon or Pfeiffer syndromes, have been elucidated to be caused by a mutation of FGFR gene, but its phenotype does not correspond to one location by one, so that prenatal gene analysis may not lead a definite diagnosis. Furthermore, other responsible genes have been found out such as Saethre-Chotzen syndrome by TWIST. In the near future, these studies of responsible gene location or clone hopefully reveals each minute pathology and prognosis, and be a foundation and application of gene therapy.[30]

Since early craniofacial neurosurgery had focused on cranial sutures, leading to suturectomies, the new idea by surgeons of the new generation has led more successful reconstruction not only for cranial form, but also orbit or midface from the needs (Fig. 1).[31–36] And so the patients with craniosynostosis or craniofacial dysostosis syndromes, who have gained more stable results, over the ensuring years, have been required better cosmetic results, mainly facial part.[37,38] Namely technological development by craniofacial surgeons has reported respectively, monobloc advancement (fronto-facial advancement), RED(rigid external distraction) frames and those modifications for the facial reconstruction over the past decade.[39–41]

In the 1990s, several minimally invasive methods had been introduced. Those methods had a common concept of not separating a cranial bone from a dura mater.

In 1992, McCarthy introduced Illizarov bone expansion into the craniofacial area, with his first

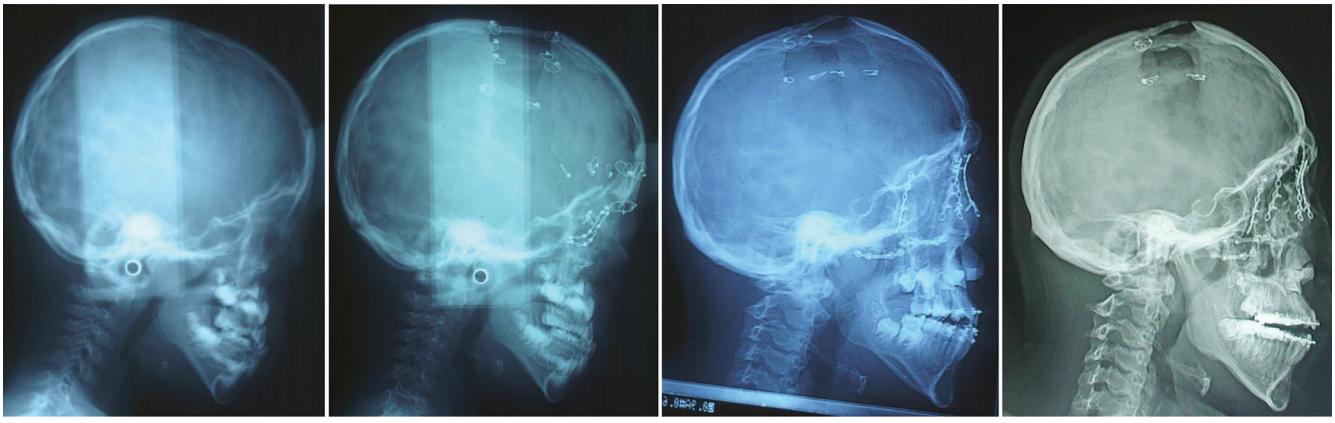

Fig. 1 Representative case treated with conventional primary surgery and later mid facial correction; Crouzon disease female underwent a conventional forehead advancement (moved anteriorly in 2.1 cm) in 1 year and Le Fort III mid-face advancement in 10 years of age. X-ray of 1(pre-operatively), 4(3 years after forehead advancement), 12(2 years after mid-face advancement), 20 years of age (right to left in order).

application being a 5-year-old boy's mandible of hemifacial macrosomia.[42] And this technique was applied to the forehead advancement by Sugawara and Hirabayashi 5 years later, and widely spread mainly in Orientals (Fig. 2).[43–47] In addition to the surgery itself, a postoperative cranial molding orthosis is an alternative. In 1999, Jimenez and Barone presented an endoscopic strip craniectomy under a small incision.[48] That provides a significant reduction in blood loss and avoids transfusion. In 1998, Lauritzen introduced the use of internal spring distractors to widen the fused suture.[49]

In 2009, White reported the posterior carvarial expansion that has been now widely accepted for a syndromic craniosynostosis, such as Apert syndrome and Crouzon disease, but it requires more cranial space expansion, otherwise they may require polysurgery due to relapse and additional volume necessity.[47,50] Where is craniofacial surgery heading, when all the procedures and concepts has been appeared out? Practical gene therapy or endoscopy-assisted skull base suturectomy in infancy hopefully will take place.

Surgical management of craniosynostosis in Japan

The surgery for the craniosynostosis in Japan may divide into 4 eras, since the management has been practically started with wide-suturectomy, mainly done by neurosurgeons, as follows;

Period I (dawn era of surgery, since late 1960s)

Because of the low incidence of the disease, the linear suturectomy had been started generally in Japan since around 1980. But its advent actually goes back to more than a decade, the author had noticed the tiny pair of wires migrated in the calvarium of patient who suffered Apert syndrome that I operated on more than 25 years ago, and have long been wondered who left those steel wires in her head; I found the technique and the surgeon by chance from

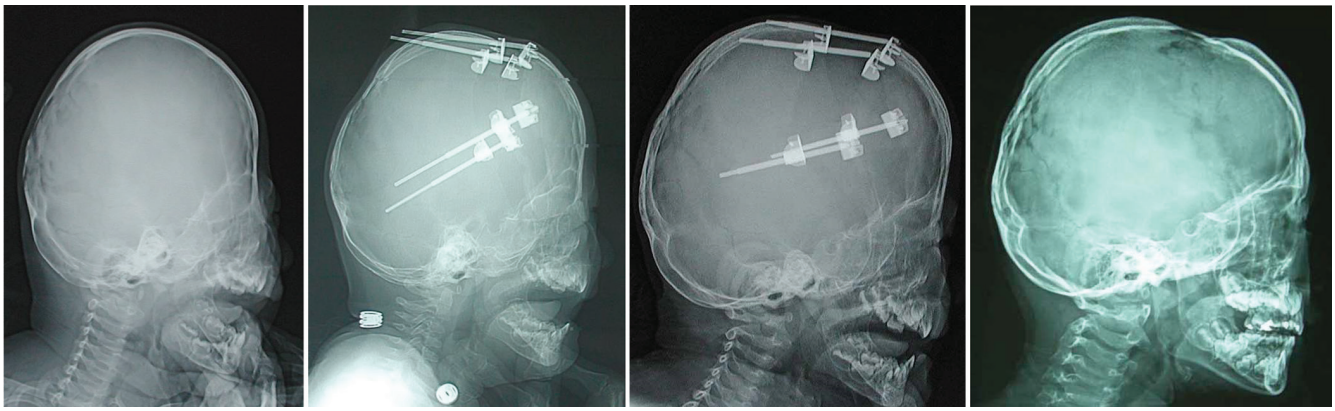

Fig. 2 Representative case treated with distraction; A 8 month-old female with brachycephaly underwent a forehead advancement with gradual distraction. X-ray of pre-ope, during distraction, full expansion (2.3 cm anteriorly) and 10 years after (right to left in order).

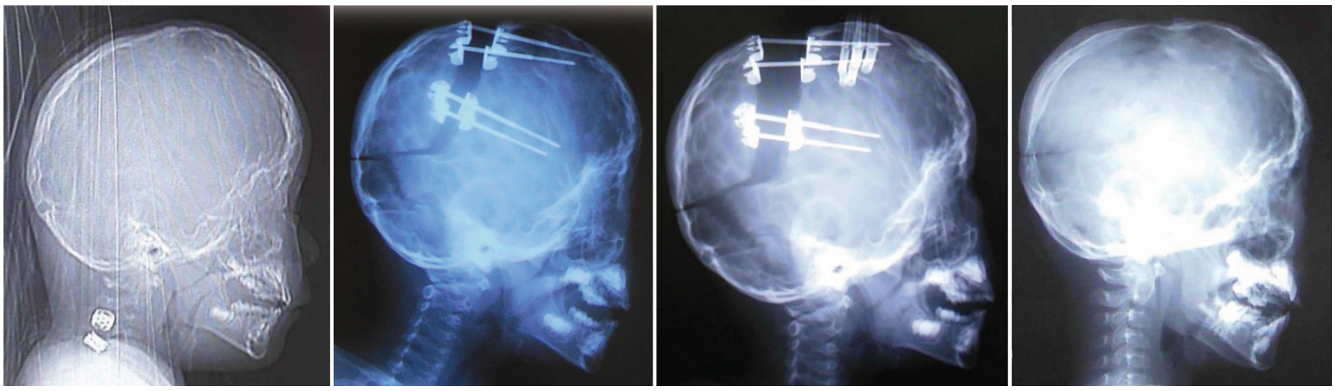

Fig. 3 Illustrative case with posterior distraction; Follow-up conditions of before to 2 years after of posterior expansion applied to a 4 year-old male with oxycephaly who underwent a frontal advancement in 1 year. X-ray of pre-ope, during distraction, full-expansion (2.3 cm posteiorly) and 2 years after (right to left in order).

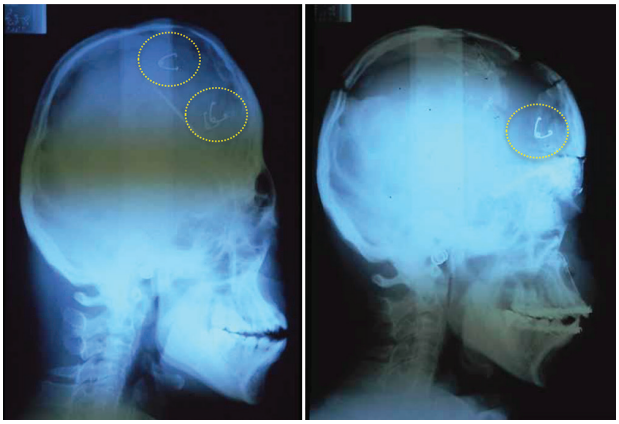

Fig. 4 A proof of the earliest Japanese spring therapy; A 17 year-old female with Apert syndrome underwent a forehead and mid-face advancement in 1989. Pre and post-operative X-ray shows springs (*yellow dot circle*) on the calvarium.

a Japanese article from 1967 (Fig. 4). Thirty-one years before Lauritzen,[49] Uejima had reported the linear craniectomy with pairs of spring that maintained the gap and prevented re-fusing.[51] He applied this operation to 60 cases and compared them with 64 cases with simple suturectomy, and concluded the most effective results. Removal of the fused suture had been done to release the constricted brain in this era.[12,52]

Period II (development of craniofacial surgery, 1983-)

After Tessier, the world's first craniofacial surgeon developed an extensive and more whole cranial work. In 1978, Marchac reported a frontal advancement procedure and followed and established the method as a common treatment for the craniosynostosis in all over the world as well as in Japan. Plastic surgeons had started this work in association with neurosurgeons during this period, when the international society of Craniofacial Surgery was founded by Tessier and his disciples in 1983.[53]

Period III (evolution by distraction 1998-)

Five years after McCarthy's introduction of the Illizarov technique into a craniofacial surgery, [42] Japanese craniofacial surgeons, Sugawara and Hirabayashi published their preliminary successful application to the calvarium and pushed many Japanese craniofacial surgeons to follow along.[43–45,47] Up to 2009, 312 cases had been combinedly reported and showed on the advantages of a distraction for the craniosynostosis from Japan,[46] since it is less invasiveness, without a bone flap separation from a dura, with less bleeding and smaller relapse, compared with conventional procedure, length, speed, direction of expansion valuably regulated. But the Western surgeons still seemed not to accept a distraction to the calvarium, probably because of low bleeding tendency or other differences between Japanese and Westerns.[54]

Period IV (reflection and revolution 2009-)

But White's report in 2009 of the posterior calvarial expansion has been rapidly accepted by Western surgeons; it is interesting that cranial distraction the Caucasians did not accept for decades has been finally started by a report from England.[50] The method which is thought to be a good indication for the syndromic craniosynostosis, because of the amount of cranial expansion is much more effective than frontal distraction advancement or conventional procedure.[45,55] And in this era, the procedure may look back to the calvarial remodeling, in terms of more sophisticated way with less invasiveness rather than one direction elongation. That[51] is to say; the distraction method should not be applied to severely distorted clinocephaly or asymmetrical supra-orbital area which required morphologically satisfied results.[35,56,57,58] Nowadays operation has been safer with the development of technique of an anesthesia with an auto-transfusion or other equipment. And the surgical management is needless to say preferable when lesser invasive and shorter duration.[15,36,46,59]

Timing of the surgery and selection of surgical procedures

"Earlier surgical treatment will have better results with functional and morphological improvement by means of releasing of brain contraction" is the well-known rule among surgeons, except for a few objections.[60–62] But, how early and which procedure to choose for every type of craniosynostosis?

"The early surgery"defines when? Less than sixth months[59,63] may give more alternatives for surgeons, but some earlier reports of a retrospective study recommend[64,65] early surgery, for craniosynostosis less than 9 to 18 months of age, could improve morphology and prevent functional disturbances, with low perioperative complications and no mortality, when pan synostosis and craniofacial dysostosis syndromes have gotten worse results with recurrence or cranial vault mal development that needed major secondary operations. It should be kept in mind that the authors of those papers chose calvarial remodeling.[66,67] In particular to the suturectomy for a scaphocephaly, some reports recommend that it be done at less than 3 month old and the Pi procedure (Greek letter π shaped ostectomy) in less than 2 month old,[7,68] and comparative study with cranial remodeling proved to be worse, if operated later than 1 year.[13,56]

Table 1 Treatment for craniosynostosis - Japan and Western

			1890	Lannelongue	craniectomy
			1948	Ingraham	midline craniectomy
1967	Uejima	suturectomy with spring	1968	Shillito	strip craniectomy
1978	Yamaguchi	suturectomy	1976	Venes	sagittal synostectomy
			1977	Stein	extensive craniectomy
			1978	Jane	π-procedure
			1982	Epstein	total vertex craniectomy
			1982	Marchac	total calvarial reconstruction
1985	Mori	suturectomy	1986	Olds	extensive craniectomy
1985	Nakajima	fan technique	1987	Raimondi	strip suturectomy
1985	Kamiishi	floating sagittal bar technique	1989	Persing	total calvarial remodeling
1987	Mori	suturectomy + temporal osteotomy	1989	Hendel	'Gore pattern' cranial remodeling
1988	Kawakami	π-procedure	1991	Marsh	total vertex craniectomy
1988	Nakajima	bamboo-ware technique	1993	Hudgines	total calvarial reconstruction
1992	Mori	expanding cranioplasty	1993	Sutton	total cranial vault reconstruction
			1996	Pollack	total calvarial reconstruction
1998	Sugawara	gradual cranial vault expansion	1998	Jimenez	endoscopic craniectomy
1998	Hirabayashi	gradual cranial vault expansion	1998	Lauritzen	dynamic cranial reshaping
1998	Aida	modified π-procedure			
1999	Imai	3D model simulated surgery	1999	Panchul	suturectomy vs remodeling
2000	Nakajima	gradual distraction	2002	McCarthy	hung span method
2001	Imai	cranial remodeling by DOG			
2009	Komuro	posterior expansion	2009	White	posterior expansion
			2015	Wink	FOA vs PVDO

DOG: distraction osteo-genesis, FOA: fronto-orbital advancement, PVDO: posterior cranial vault distraction osteo-genesis.

To summarize timing, in recent years, most of the Western surgeons roughly do cranial remodeling at less than one year of age, suturectomy with helmet at less than 3 month old and cranial distraction at less than 6 month old, as a first surgery.[66,67] Regarding the types of deformity of the cranium, Marchac recommends 2 to 4 months operation for scaphocephaly, brachycephaly, and syndromic synostosis, and 6 to 9 months for plagiocephaly and trigonocephaly with calvarial remodeling.[69,70] Even in Japan where the fronto-orbital distraction has been common since early 2000, some surgeons still choose a conventional method for plagiocephaly and trigonocephaly, that requires symmetrical supraorbital remodeling as well as, frontal advancement; the authors agree with this adaption.[14,32,46] And the synostosis, such as brachycephaly, scaphocephaly, is good indication for distraction, because one direction expansion can resolve those conditions, if performed in early age. Another advantage of the distraction method is its tiny adjustment contrary to on-time conventional repair; the elongation could stop anytime you like according to an extra-dural space or releasing the increased intracranial pressure. The authors have applied subtle expansion for a mild form trigonocephaly with increased ICP, indicated for surgery with over 15 mmHg (mean) in an extra dural monitouring.[71–73] Khechoyan reported the characteristic distortion of scaphocephaly of frontal bossing could be avoided, if the Pi procedure is done at less than 3 months.[74] Conversely older scaphocephaly should not be treated by one direction distraction, because narrow frontal bossing would remain and anterior-posterior elongation is not treatable. The older the patient, whose cranium is the harder.[75–77] For the scaphocephaly, a[59] variable strategy is needed, based on its age or severity.[31,56,78–80]

Syndromic craniosynostosis usually requires more operations than simple one, because the cranium needs more space and tends to relapse, and the mid-face advancement will surely be required, moreover hand surgery as well; first time operation for the craniofacial dysostosis syndrome should be treated with reliably effective cranial expansion until the second surgery is performed. In 2015, Derderianet et al. reported a comparative study between fronto-

orbital advancement and posterior cranial vault distraction; volumetric change by posterior distraction is approximately twice as big as the former method, and concluded the posterior expansion gains highly significant volume[55] (Fig. 3). In the last decade, the procedure has been used generally in Western and Japan and additional contributions seemed to be produced, such as improvement of Chiari malformation or fronto-facial impression.

Summarizing above, the authors' policy for the optimal timing of the surgery is 3 to 6 months of age, and the procedure would be varied by the type of craniosynostosis; scaphocephaly may be treated with suturectomy, before 3 months, the distraction could be helpful, if not, as bracycephaly, plagiocephaly and trigonocephaly is desirable to choose conventional fronto-orbital advancement, for syndromic cases, posterior calvarial expansion should be applied at first.

Summary

Looking back on around two centuries of history on craniosynostosis, its character, symptoms and surgical treatments are listed according to the eras, divided by breakthrough developments: craniofacial surgeons seem to be changing methods with more sophisticated, than less invasive ways.

Acknowledgement

The authors thank Dr. H. Kamiishi, Dr. Y. Yamanouchi, Dr. S. Kajimoto and Dr. K. Ueda for their excellent instruction and cooperation.

Conflicts of Interest Disclosure

The authors declare no conflict of interest regarding this review article.

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