

Craniofacial Surgery for Craniosynostosis: Challenges in Diagnosis, Management and Long-term Outcome

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Background and Objective: Craniofacial surgery for craniosynostosis is one of the most challenging reconstructive procedures. Restoration of particular functional and anatomic requirements is important for development from infancy to adulthood. The purpose of this study is to present the authors' experience of craniofacial surgery for management of patients with craniosynostosis in Srinagarind Hospital, Khon Kaen, Thailand, addressing the challenges of diagnosis, management and outcomes, which may be adapted in other developing countries.

Material and Method: This paper presents the cranial and associated deformities, diagnosis, radiologic findings, preoperative evaluation, craniofacial and maxillary surgeries and outcome(s) of patients with craniosynostosis. The care team, made up of neurosurgeons, plastic surgeons, radiologists, ophthalmologists and pediatricians, established the Tawanchai Center's protocol for craniosynostosis, to manage the timing of craniofacial procedures from infancy to adulthood.

Results: The physical examination and radiologic findings of three patients, two with sagittal synostosis and one with plagiocephaly are reported. The clinical, craniofacial and maxillofacial surgeries and long-term outcomes of another three patients were studied, one with Apert syndrome and two with Crouzon syndrome. All the latter three patients were lost to follow-up after the initial post-surgical visit. At that time, there were appropriate surgical results vis-à-vis appearance and satisfaction from the perspective of the two patients with Crouzon syndrome and their families. One of the patients with Crouzon syndrome received normal education supported by a successful family, while the other was still continuing her studies at school with good progress. The patient with Apert syndrome continued to live with his parents. Additional reconstructive surgery is recommended for all three patients. Economic problems and lack of adequate information were the main reasons for their discontinuing follow-up appointments.

Discussion and Conclusion: Systematic physical examination and radiologic assessments by the craniofacial team are critically important for diagnosis, evaluation, planning of management and outcome assessment of the patients with craniosynostosis. In Thailand and other developing countries, the challenges in management of these patients are the development of standard craniofacial surgery, craniofacial team management and well-coordinated care, planned surgeries and outcome assessments from infancy to adolescence. A supportive government health system and establishment of a craniofacial center and foundation is needed in order to support and provide proper care for these groups of patients.

Keywords: Craniosynostosis, Craniofacial surgery, Challenges, Diagnosis, Team management, Long-term outcome, Developing countries

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Craniosynostosis is the premature fusion of one or more cranial sutures leading to suture-specific deformity of the cranial vault, cranial base, and facial bones, and can involve any of the cranial sutures, including sagittal, lambdoidal or coronal. Hippocrates⁽¹⁾ described cranial deformities and their relationship with cranial sutures while Galen described the significance of cranial sutures by patients with headache and exophthalmos, with too few sutures⁽²⁾.

The physiology of an abnormal skull shape was first described by Virchow in 1851, who stated that skull growth is arrested in a perpendicular direction to the closed suture which causes compensatory over-expansion to take place⁽³⁾. He also published the first classification correlating site of suture synostosis with the head shape. Craniosynostosis can occur as non-syndromic or syndromic craniosynostosis. Its etiology remains unclear and occurs at an estimated birth incidence of 0.4 per 1,000 births⁽⁴⁾.

Types of craniosynostosis were defined by Cohen in 1986⁽⁵⁾ as simple (coronal synostosis and metopic synostosis), complex (coronal and sagittal synostosis and sagittal and metopic synostosis), primary (coronal synostosis, sagittal synostosis, and coronal and sagittal synostosis), secondary (hypertelorism and rickets), isolated (sagittal synostosis and coronal and sagittal synostosis) and syndromic (Apert, Crouzon and Carpenter syndromes).

The first surgical repair of craniosynostosis was performed by Lannelongue in 1892⁽⁶⁾. The earliest techniques were linear craniectomy and fragmentation of the cranial vault. In 1967, Tessier published the results of craniosynostosis surgery by intracranial approach to correct the recessed forehead and supra-orbital region⁽⁷⁾. Cranial vault reconstruction with fronto-orbital advancement to correct bilateral coronal synostosis was reported by Persing et al⁽⁸⁾, and Cohen et al⁽⁹⁾.

Surgical reconstruction of craniosynostosis has evolved from simple "suturotomy" to extensive cranial vault, fronto-orbital and mid-face reconstruction. Among the recently advanced techniques are minimally invasive techniques for craniosynostosis repair reported by Vicari⁽¹⁰⁾, and distraction osteogenesis to reconstruct the fronto-orbital and cranial vault abnormalities⁽¹¹⁾. Restoration of particular functional and anatomic requirements is important for optimal intelligence and neurocognitive development. Early surgery is recommended to prevent problems such as cranial vault reconstruction during the first year of life⁽¹²⁾.

The purpose of this study was to present the

authors' experience with craniofacial surgery for management of patients with craniosynostosis at Srinagarind Hospital, Khon Kaen, Thailand, while addressing the challenges in diagnosis, management and outcome and to offer this experience for developing craniofacial surgery in other developing countries.

Material and Method

The protocol of this study has been reviewed and approved by the Ethics Committee of Khon Kaen University, based on the Declaration of Helsinki and written informed consent was obtained for each patient.

Diagnosis and Preoperative Evaluation

The child with craniosynostosis demonstrates classic patterns of cranial deformities which are due to the inability of the skull to expand in a direction perpendicular to the stenosed suture, such as in sagittal synostosis (scaphocephaly), coronal synostosis (brachycephaly), unilateral coronal synostosis (plagiocephaly), metopic synostosis (trigonocephaly) and lambdoid synostosis (plagiocephaly). It is believed that extensive involvement of the cranial base sutures in syndromic craniosynostosis may result in profound mid-face hypoplasia with exorbitism and malocclusion. There may be a varying incidence of an associated increase in intracranial pressure and developmental delay seen more commonly in multiple suture and syndromic craniosynostosis.

After team consultation for a patient with craniosynostosis, a comprehensive and systematic evaluation is performed for appropriate diagnosis and evaluation of associated and related deformities, including differentiation between non-syndromic and syndromic craniosynostosis. Patients who require craniofacial surgery are thoroughly evaluated, including a detailed, pre-operative history and physical examination vis-à-vis function and aesthetics by a plastic surgeon, a neurosurgeon and an ophthalmologist.

The size of the brain triples during the first year of life and there may a disparity between brain size and intracranial volume leading to an increase of intracranial pressure. Fundoscopic examination by an ophthalmologist is performed to identify papilledema, an indicator of increased intracranial pressure. A skull series is used to evaluate the cranial sutures for evidence of sutural fusion and signs of increased intracranial pressure. Ultrasonography, computed tomography (CT) and three-dimensional CT are

essential for assessment of the brain and cranial vault anatomy as well as for checking for signs of hydrocephalus, increased intracranial pressure and other pathologic neuroanatomy. The frequently associated radiologic findings with increased intracranial pressure are “thumb printing” or the beaten-copper appearance of the skull. Information derived from these imaging studies is important for pre-operative consideration.

Craniofacial Surgery for Craniosynostosis

The correction of deformities of a child with craniosynostosis is accomplished through a protocol requiring a series of operations according to the period of growth and development of craniofacial skeleton. Cranial vault remodeling is indicated in craniosynostosis with intracranial hypertension regardless of the patient’s age to prevent ocular damage and other neurologic injuries⁽¹³⁾.

At Srinagarind Hospital, the team care (including neurosurgeons, plastic surgeons, radiologists, ophthalmologists and pediatricians) has established a protocol and system for evaluation, consultation and management of patients with craniosynostosis. Table 1 presents the protocol for dealing with cleft lip-palate and craniofacial deformities (*i.e.*, craniosynostosis) at The Tawanchai Center at Khon Kaen University.

Intraoperative Management

After assessment, evaluation, pre-operative management and anesthesia, craniofacial surgery is performed by a plastic surgeon who dissects the scalp soft tissue and provides exposure of the calvarium. Then a neurosurgeon performs a craniotomy and further extradural dissection. Adequate exposure is required for fronto-orbital advancement, orbital exploration and temporo-parietal barrel-staving. Traction sutures are used to affix the dura to advance the bone fragment. The bone flaps are replaced after re-modeling and re-

positioning. Wirings or absorbable plates and screws are preferred for bony fixation.

Patient Reports

Patient 1 - Sagittal Synostosis

Fig. 1 shows the patient with sagittal craniosynostosis and a scapho-cephalic head shape with long antero-posterior diameter, bitemporal narrowing and prominent occiput.

Patient 2 - Sagittal Synostosis

Photos and plain films of another patient with sagittal craniosynostosis demonstrate a scapho-cephalic head shape (Fig. 2).

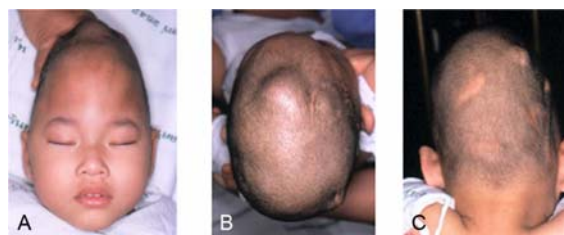


Fig. 1 Patient with sagittal craniosynostosis demonstrating a scapho-cephalic head shape

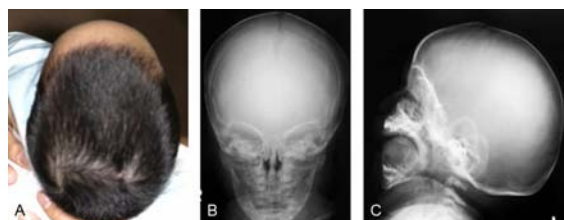


Fig. 2 Patient with sagittal craniosynostosis (A) shows the scapho-cephalic head shape. Plain films of skull in frontal (B) and lateral (C) projections show premature synostosis of the sagittal suture with elongation of the calvarium in the occipital-frontal direction and decreased transverse axis of the calvarium

Table 1. The Tawanchai Center’s protocol for craniofacial procedures, particularly for patients with craniosynostosis

Procedures	Timing (Age)
Ventriculo-peritoneal shunt, strip craniectomy, fronto-orbital advancement, cranial vault remodeling	Infancy (younger than 1 year)
Secondary fronto-orbital advancement/ cranial vault remodeling, LeFort III osteotomy and mid-face advancement	Early childhood (younger than 6 years)
Monobloc fronto-facial advancement, mid-face distraction	Late Childhood (younger than 12 years)
Orthognathic surgery, rhinoplasty	Adulthood (18 years or older)



Fig. 3 Patient with unilateral coronal synostosis and bilateral lambdoid synostosis

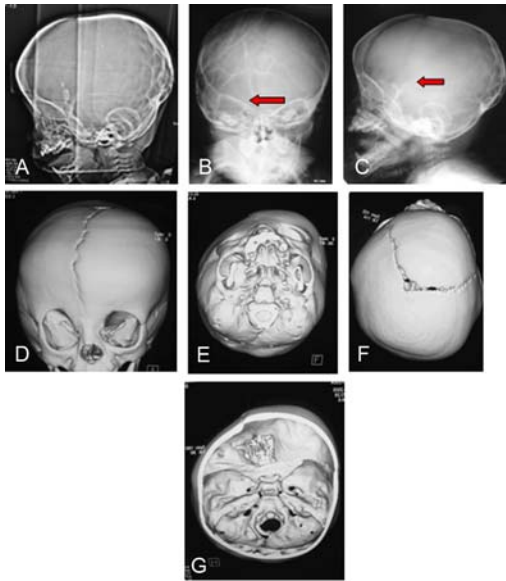


Fig. 4 Plain films (A, B, C) and CT scans (D, E, F, G) of a patient with unilateral right coronal synostosis and bilateral lambdoid synostosis

Patient 3 - Plagiocephaly

Plagiocephaly is the common term for cranial asymmetry. Photos (Fig. 3) and radiologic findings (Fig. 4) of a patient with unilateral right coronal synostosis and bilateral lambdoid synostosis demonstrate frontal flattening of the right side and the right ear pulled toward the affected right coronal suture with bilateral



Fig. 5 Patient with Apert syndrome demonstrates a turribrachycephalic calvarial deformity, exorbitism, maxillary hypoplasia (A, B, C), papilledema (D) and complex syndactyly of both hands and feet (E, F, G)

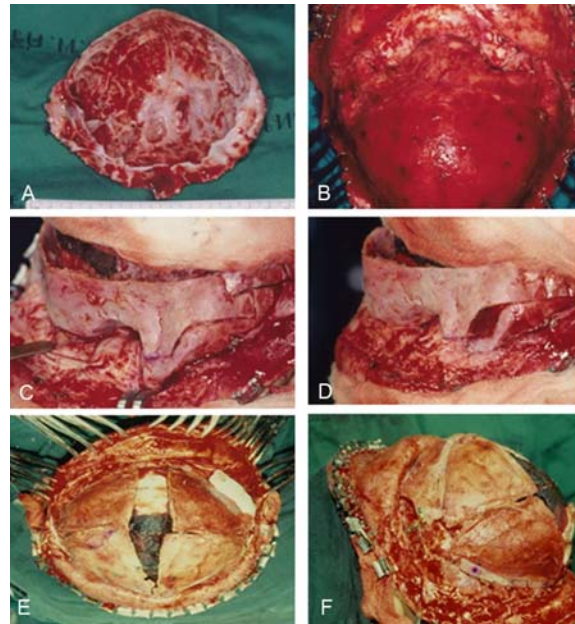


Fig. 6 Patient with Apert syndrome show the shape of the skull during the operation (A, B) which indicated increased intracranial pressure present on the inner

occipital flattening. Plain films and CT scans demonstrate the presence of: 1) increased convolutional markings (or thumb-printing) of the inner table of the calvarium; 2) the unilateral harlequin sign; and, 3) the

cranial base deviating toward the affected side.

Fig. 4, plain films of the skull in frontal (B) and lateral (A and C) projections, show: 1) premature closure and ridging of the bilateral lambdoid sutures; 2) flattening of the occipital squamosa; and, 3) a squamosa narrower than normal. Premature closure of the right limb of the coronal suture caused lifting of the roof of the right orbit into a more oblique position (harlequin appearance). There is an increased height of the skull caudad to the cephalad and a decreased antero-posterior dimension. Increased convolitional markings (thumb-printing) indicate long-standing increased intracranial pressure.

Frontal (D), apex (E), basal (F) and skull base (G) views of the three-dimensional CT reveal characteristic deformities of plagiocephaly: flattening of the ipsilateral fronto-parietal region, elevation of the ipsilateral sphenoid wing and tilting of the right orbit, and a cranial base deviated toward the right side. Small anterior cranial fossa and relative enlargement of middle cranial fossa are also seen.

Patient 4 - Apert Syndrome

A male patient, living in Khon Kaen Province, first presented at the age of 9 months in 1993 with a turribrachycephalic calvarial deformity, severe exorbitism, maxillary hypoplasia and complex syndactyly of both hands and feet. Papilledema was also demonstrated on fundoscopic examination (Fig.



Fig. 7 Early results at age 2 in 1993 (A, B, C) and follow-up at age 18 (D, E, F). Hands and feet left untreated (G, H, I)

5).

Fronto-orbital advancement with total cranial vault remodeling through a coronal incision was performed at the age of 9 months. The sign of increased intracranial pressure was also seen on the inner side of the skull during the cranial vault remodeling (Fig. 5).

Due to the family's poverty, the patient could only be taken for a few follow-up visits when he was young and so did not contact with the hospital until he was 18 (in the year 2010), not having had any additional surgery or other management. Fig. 7 shows early and late results.

He was the second child of his mother and lived with his grandmother because both of his parents worked in another province. He studied until he finished primary school but could not continue his education because of: 1) scholastic limitations; 2) teasing from peers at school; and 3) economic and



Fig. 8 Patient with Apert syndrome at his house with his grandmother in 1995 and during The Tawanchai Foundation team visit in 2010



Fig. 9 Patient with Crouzon syndrome with turribrachycephalic calvarial deformity, shallow orbits, exorbitism, strabismus, mid-face hypoplasia and malocclusion with anterior open bite

transportation problems.

His parents were afraid of his undergoing a high-risk operation, not being aware of the qualified team of surgeons at Srinagarind Hospital. When the team visited his home (Fig. 7), he still had his hospital appointment card because he still hoped for surgical correction if there ever was financial support.

Patient 5 - Crouzon Syndrome

A female patient, from Mahasarakam Province, presented in 1993 at 8 years of age with turribrachycephalic calvarial deformity, shallow orbits, exorbitism, strabismus, mid-face hypoplasia, malocclusion with anterior open bite (Fig. 9). Fronto-orbital advancement was performed at 8 years of age (Fig. 10).

She underwent LeFort III mid-face advancement a year later, but was lost to follow-up until the age of 25. At the time of the team visit in 2010,

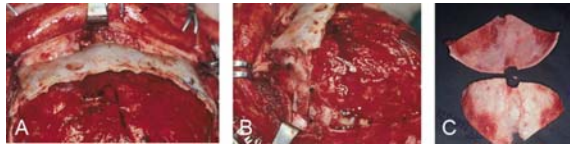


Fig. 10 Fronto-orbital advancement in the patient with Crouzon syndrome. She underwent LeFort III mid-face advancement a year later, but was lost to follow-up until the age of 25. At the time of the team visit in 2010, she still had mild headaches and problems with her left eye.



Fig. 11 Patient with Crouzon syndrome at age 3 (A, B, C) and during the last visit at age 25 (D, E, F)

she still had mild headaches and problems with her left eye.

Owing to poverty, the patient stayed in a small hut with her mother. Later, she received support from a donation and the government for a new house (Fig. 12). She did well in school finishing secondary school. At the time of this report, she was married with two normal sons. Her reasons for refusing to go for follow-up and further surgery were: 1) fear of the high-risk surgery; and, 2) worry over who would take care of her sons if she died during the surgery. After the team visit, however, she decided to go to see the doctors at Srinagarind Hospital for additional treatment.

Patient 6 - Crouzon syndrome

A female patient, living in Mukdaharn Province, visited Srinagarind Hospital in 1999 at the age of 3. She presented with turribrachycephalic calvarial deformity, shallow orbits, exorbitism and mid-face hypoplasia (Fig. 13).

The patient received fronto-orbital advancement in 1999 and schedule for maxillary surgery was planned but she was lost to follow-up because of problems getting to treatment and poverty. She lived with her parents and had a normal brother and a normal sister. The household income was unstable. The parents were also taking care of their grandchild and did not have the time to take her to hospital for continuity of treatment and follow-up.

During The Tawanchai Foundation visit, her mother said that she hoped for additional surgery. In



Fig. 12 Patient with Crouzon syndrome. A and B taken in 1995 while the patient was at school and with her mother in the old house. C and D taken in 2010, show the new house constructed by the government, and the visit from The Tawanchai Foundation Team

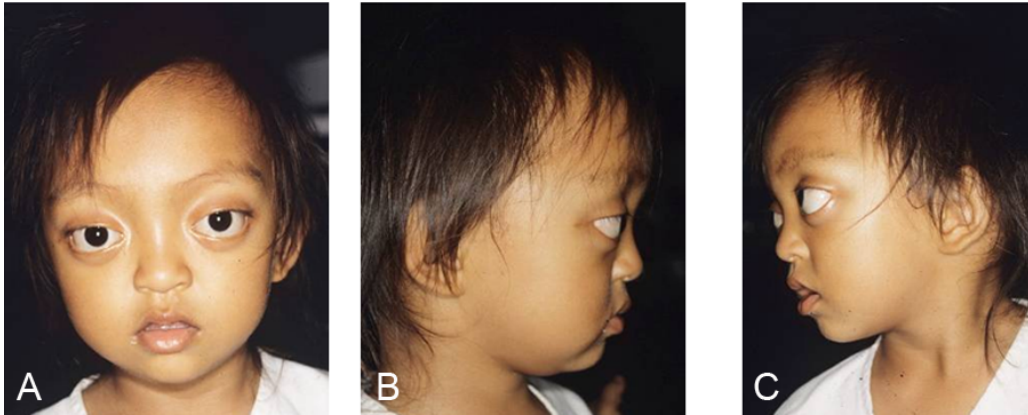


Fig. 13 Patient with Crouzon syndrome presenting with turribrachycephalic calvarial deformity, shallow orbits, exorbitism and mid-face hypoplasia

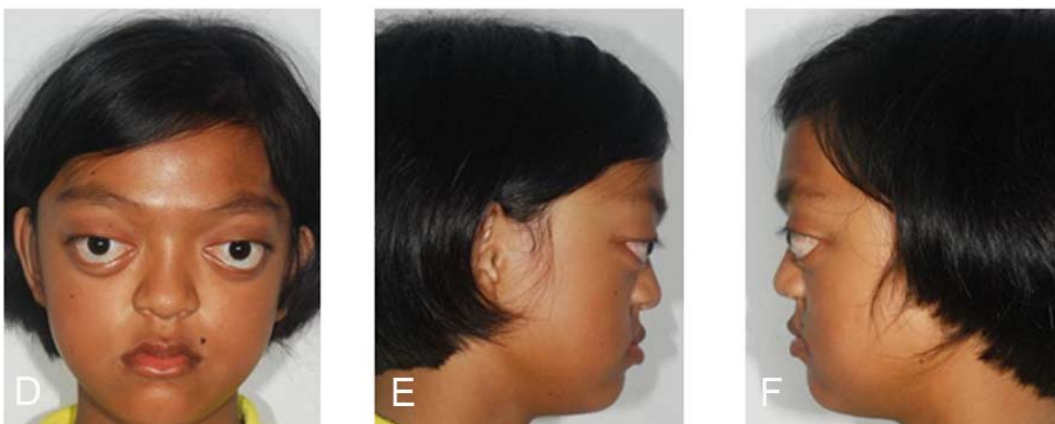


Fig. 14 Patient with Crouzon syndrome after fronto-orbital advancement at the age of 4 years (A, B, C) and during the follow-up visit at 14 years of age (D, E, F)

2010, the patient was 14 and doing well in school at secondary level (top 10 in her class) and was not being teased about her abnormalities.

Discussion and Conclusion

In 1996, the Team Standards Committee of the American Cleft Palate-Craniofacial Association recommended the standards for a Craniofacial Team (CFT). The goal of such a team is to provide coordinated, properly sequenced evaluations and treatments, and with comprehensive clinical records, keeping in mind the framework of the patient's overall development, medical and psychological needs⁽¹⁴⁾. The complex nature of many types of craniofacial anomalies often necessitates multiple operative procedures at the different stages of growth and development.

Longitudinal follow-up is therefore necessary until at least 21 years of age when craniofacial and jaw growth is expected to have ceased. Specific components of the peri-operative evaluation of craniofacial surgery should be based on type of anomaly and the craniofacial zones affected. Reconstructive surgery of the craniofacial region may include soft and hard tissue remodeling, reconstruction, grafting, distraction and implantation. A secondary procedure should be planned for dealing with any persistent, residual deformities that were not corrected or that have resulted from a previous surgery, as well as for evaluating organ function, effects of growth and attainment of maturity⁽¹⁵⁾.

The problems of infant patients with craniosynostosis may include increased intracranial pressure, hydrocephalus, mental retardation and visual abnormalities. Later problems may include craniofacial disproportion, dental problems, dental malocclusion, psychosocial stigma, as well as disturbed growth and development. Systematic physical examination and radiologic assessment by a craniofacial team are critically important for diagnosis, evaluation, planning of management and outcome assessment of these groups having severe craniofacial malformations.

In Thailand, and other developing countries, the challenges of the management of craniosynostosis include the development of standard craniofacial surgery with appropriate craniofacial team composition and well-coordinated care. The proper protocol should be adapted but is still comparable to the universal standard. The challenges of management during infancy are proper diagnosis, evaluation and management of urgent problems, while the challenges of management during craniofacial growth are

opportunities to follow-up and an evaluation of these patients at the critical period of the development.

Besides including neurosurgeons, plastic surgeons, ophthalmologists and pediatric craniofacial anesthesiologists, the authors suggest that an interdisciplinary team should include orthodontists, psychiatrists, social workers and cleft coordinators. Key factors that will assist with the overall treatment include: 1) financial support if necessary, 2) support for transport to a treatment center, 3) provision of education and information to patients and their parents; and, 4) health support.

Since these groups of patients have complex problems needing interdisciplinary craniofacial team management, a craniofacial center is needed as a referral center with super-tertiary care. Indeed, the establishment of "The Tawanchai Foundation for Cleft Lip-Palate and Craniofacial Deformities" has contributed to the future care of these groups of patients.

Further research in the epidemiology, prevention, improvement of care and outcome evaluation should also be conducted. Further innovative surgeries such as minimal invasive surgery, distraction osteogenesis and tissue engineering should be considered.

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การผ่าตัดศีรษะและใบหน้าในผู้ป่วยครานิโอซินอสโตสิส: ความท้าทายในด้าน การวินิจฉัย การรักษา และผลลัพธ์ในระยะยาว

ไชยวิทย์ ธนไพศาล, บวรศิลป์ เชาวนชื่น, ปารธนา เชาวนชื่น

ภูมิหลังและวัตถุประสงค์: การผ่าตัดศีรษะและใบหน้าในผู้ป่วยครานิโอซินอสโตสิสเป็นหัตถการที่มีความท้าทายมากอย่างหนึ่ง การคงสภาพทั้งด้านหน้าที่การทำงานและกายวิภาคเป็นสิ่งจำเป็นต่อการพัฒนาการตั้งแต่เด็กจนเป็นผู้ใหญ่ วัตถุประสงค์ของการศึกษาคั้งนี้ คือการนำเสนอประสบการณ์ของผู้นิพนธ์ในการผ่าตัดศีรษะและใบหน้าในผู้ป่วยครานิโอซินอสโตสิส ในโรงพยาบาลศรีนครินทร์ จังหวัดขอนแก่น ประเทศไทย โดยเน้นที่ความท้าทายในด้านการวินิจฉัย การรักษา และผลลัพธ์ ซึ่งอาจนำไปประยุกต์ใช้ได้ในประเทศที่กำลังพัฒนาอื่นๆ

วัสดุและวิธีการ: ผู้นิพนธ์นำเสนอความผิดปกติของกะโหลกศีรษะและความพิการอื่น การวินิจฉัยการตรวจพบทางรังสี การประเมินก่อนการผ่าตัด การผ่าตัดศีรษะและใบหน้าและการผ่าตัดกระดูกส่วนกลางของใบหน้า และผลการรักษาผู้ป่วยครานิโอซินอสโตสิส ที่มีการดูแลที่ประกอบด้วยศัลยแพทย์ระบบประสาท ศัลยแพทย์ ตกแต่ง รังสีแพทย์ จักษุแพทย์ และกุมารแพทย์ ได้กำหนดแนวทางการดูแลรักษาทางศีรษะและใบหน้าในผู้ป่วยครานิโอซินอสโตสิส ช่วงระยะเวลาการรักษาตั้งแต่ขวบปีแรกถึงวัยผู้ใหญ่ของศูนย์ตะวันฉาย

ผลการศึกษา: ผู้นิพนธ์รายงานผลการตรวจร่างกายและลักษณะทางรังสีวิทยาของผู้ป่วย 2 ราย ที่มีภาวะ sagittal synostosis และ plagiocephaly ผู้ป่วย 3 ราย ที่ได้รับการรักษาโดยการผ่าตัดศีรษะและใบหน้า เป็นกลุ่มอาการเอเปอร์ต 1 ราย และกลุ่มอาการโครซอง 2 ราย ผู้ป่วยทั้ง 3 ราย ขาดการติดตามการรักษา ตั้งแต่ช่วงระยะแรกหลังการผ่าตัดผลการรักษาในด้านรูปร่างและความพึงพอใจของผู้ป่วยและครอบครัว ของผู้ป่วยกลุ่มอาการโครซอง 2 ราย เป็นที่น่าพอใจ โดยผู้ป่วยกลุ่มอาการโครซอง 1 ราย กำลังศึกษาในโรงเรียน และมีผลการเรียนอยู่ในระดับดี ผู้ป่วยกลุ่มอาการเอเปอร์ตยังอาศัยอยู่กับครอบครัว ผู้ป่วยทุกรายได้รับการ แนะนำการผ่าตัดเสริมสร้างเพิ่มเติม ปัญหาด้านเศรษฐกิจ การไม่ได้รับข้อมูลที่เพียงพอ เป็นสาเหตุที่สำคัญ ของการขาดการติดตามการรักษา

สรุป: การตรวจร่างกายและประเมินทางรังสีวินิจฉัยอย่างเป็นระบบโดยทีมการดูแลผู้ป่วยศีรษะ และใบหน้า มีความจำเป็นสำหรับการวินิจฉัย การประเมิน การวางแผนการรักษา และการประเมินผลการรักษาในผู้ป่วยครานิโอซินอสโตสิสในประเทศไทยและประเทศกำลังพัฒนาอื่นๆ ความท้าทายของการรักษาของผู้ป่วยเหล่านี้ คือ การพัฒนาวิธีผ่าตัดศีรษะ และใบหน้าที่เป็นมาตรฐานการจัดการทีมการดูแล ด้านศีรษะและใบหน้า และการประสานการรักษาที่ดีการวางแผนการรักษา และการประเมินผู้ป่วยตั้งแต่ขวบปีแรกจนเป็นผู้ใหญ่ ระบบสาธารณสุขของประเทศที่ดี การจัดตั้งศูนย์การดูแลผู้ป่วยพิการศีรษะ และใบหน้า และมูลนิธิมีความจำเป็นสำหรับการดูแลที่เหมาะสมของผู้ป่วยเหล่านี้ในประเทศที่กำลังพัฒนา
