ANTHROPOCEPHALOMETRIC ASPECTS OF FRONTOETHMOID ENCEPHALOCELE PATIENTS

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ABSTRACT

Frontoethmoid encephalocele (FEE) causes maxillofacial structural abnormalities and also covers some bony landmarks, especially in the glabella region. This study examined the faces of frontoethmoid encephalocele patients before undergoing corrective surgery with the aim of identifying and assessing their facial morphological differences. This was a cross-sectional study from January 1, 2016 to June 31, 2017. A simple anthropocephalometric measurement was performed on the patient's face who would undergo surgery. Periorbital and mid-face measurements showed differences between the patients and normal groups. The midface structure of the patients clearly changed. The age when the procedure was taken also had effect. Preoperative 3DCT is proposed to describe the facial anatomy of EFE patients. Further studies in this field are suggested to produce a comprehensive database of facial morphology changes in FEE populations, both pre- and post-operatively, with the intention of producing a redefinition of FEE cases based on anthropo-cephalometric principles.

***Keywords**: Frontoethmoidal encephalocele; anthropometry; cephalometry*

ABSTRAK

Ensefalokel frontoetmoid (EFE) menyebabkan kelainan struktur maksilofasial dan juga menutupi beberapa bony land-mark, terutama pada regio glabela. Studi ini meneliti wajah penderita EFE sebelum menjalani operasi korektif dengan tujuan untuk mengidentifikasi dan menilai perbedaan morfologi wajah mereka. Penelitian ini adalah penelitian cross sectional sejak 1 Januari 2016 hingga 31 Juni 2017. Pengukuran antroposefalometri sederhana dilakukan pada wajah penderita yang akan menjalani operasi. Pengukuran periorbital dan midface menunjukkan perbedaan antara kelompok kontrol dan EFE. Struktur midface pada penderita berubah dengan jelas. Usia saat dilakukan tindakan juga berpengaruh. 3DCT praoperasi diusulkan untuk menggambarkan anatomi wajah penderita EFE. Studi lebih lanjut di bidang ini disarankan untuk menghasilkan database perubahan morfologi wajah yang komprehensif pada populasi EFE, baik sebelum dan sesudah operasi, dengan tujuan untuk menghasilkan definisi ulang kasus EFE berdasarkan prinsip antroposefalometrik.

***Kata kunci**: Ensefalokel frontoetmoidal; antropometri; sefalometri*

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INTRODUCTION

Frontoetmoid encephalocele (FEE) is a herniation of brain tissue through bone defects in the midline of anterior cranial fossa base and outward of the facial bone, resulting in a bizzare lesion on the face (Hoving & Vermeij-Keers 1997). FEE is divided into three types: nasofrontal, nasoethmoidal and nasoorbital (Suwanwela & Suwanwela 1972). Monasterio and del-Campo divided the craniofacial deformity due to FEE into three. First, the increase in local volume and shift of the medial canthus and one or two medial orbital walls to the lateral. Second, eyebrow cranial shift and decrease the tip of the nose or both, which gives a long nose picture. Third, the separation of one or two orbites from the midline, resulting in true hypertelorism (Monasterio & del-Campo 1981).

In FEE, the pyriform aperture and nasal cartilage are deformed. The aperture is shorter and wider than usual and is pushed to the inferior. The presence of tooth malocclusion may be associated with deformity of vertical lamina of ethmoid bone that bound to the lamina cribrosa being pushed posteriorly, thus causing secondary maxillary hypoplasia. Telecanthus is generally mild, and, not as severe as that in facial cleft, it is generally classified as Tessier number II with normal Lateral Intercanthal Line (LIL) (Tessier 1976). The telecanthus degree in FEE patients depends on age and mass size. True hypertelorism is more common in adult patients due to continuous pushing of orbital walls during the early phases of growth.

Due to the existence of maxillofacial structural abnormalities and the closure of several bony landmarks, especially in the glabella region, pre- and post-operative anthropocephalometric procedures of FEE patients become the standard protocol that must be performed at neurosurgical care center. Until now no studies have been done that combines anthropometric and cephalometric aspects in FEE patients. The purpose of this study was to identify and assess the difference in facial morphology of preoperative FEE patients compared with normal anthropocephalometric data.

MATERIALS AND METHODS

This was a cross sectional study. Data collected were primary data obtained from anthropocephalometric examination in FEE patients who would undergo encephalocele surgical excision in dr. Soetomo Hospital between January 1, 2016 to June 31, 2017. This study involved a group of FEE patients undergoing simple anthropometric examination, cephalometry by examination of head AP plain x-ray and head 3D CT. The study was conducted at pediatric wards and surgical wards of Dr. Soetomo Hospital. The population in this study was FEE patients who underwent hospitalization at Dr. Soetomo Hospital. Samples in this study were 81 FEE patients hospitalized at Dr. Soetomo Hospital who met the inclusion and exclusion criteria. The inclusion criteria in this study were male or female FEE patients with no other congenital abnormalities in maxillofacial region, willing to participate in the study, and signed informed consent, while the exclusion criteria were the presence of sealed encephalocele pockets or failing to undergo anthropocephalometric examination.

Anthropometric method

The subjects of the study are seated on a chair with a FHP (Frankfurt Horizontal Plane) line parallel to the floor. The reference point is made with wetted ink pencil. At the time of measurement, subjects are attempted not to move their heads, then measurements are made using sliding calipers, angle finders and term calipers. Craniofacial proportions are determined by direct anthropometric measurements include the Intercanthal Width (en-en), Binocular Width (ex-ex), nose bridge, eye angle, nasal tip (NT), GIS, GCL, and GCM. The maximal projection point of the nose is used as a nasal tip reference because this point is relatively undisturbed by FEE mass (Tessier 1976). Distance between GIS and nose tip, distance between GIS-GCL, and GCL-GCM were measured. Medial and lateral canthus and nose tip are marked, so are GIS, GCM, GCL and centrophasial proportions. The overall measurement is compared to normal population. From here we determine the magnitude of the shift that will be done on nasoorbital component so as to achieve the harmony of the face.

Cephalometric method

The first step is to examine the patient in the cephalostat according to standard measurements for cephalometric analysis. The subject head is positioned at an angle of 90 degrees to the X-ray, with a distance of 1.5 meters from the tube, and the film is placed 15 inches from the patient's head. After taking the head X-ray, the film needs to be traced with multiple angles using a pencil or a small pen (0.5 mm). Tracing should be at a 1: 1 ratio on radiography. When searching, all lights are turned off and the area around the radiograph should be dark so the reading will be clear. Clear and accurate marking of anatomical structures is important because the angle and the reference line depend on the accuracy. Cephalometry includes measurements of IOD, EOD, CW, LLC, and IOD/CW ratios. The procedure for making head X-rays that cannot meet the standard of cephalometric techniques can be replaced by relative measurements of head 3D CTscan (Mafee et al 1986).

Management and data analysis

Data obtained underwent the process of coding, entry, cleaning, and editing. Furthermore, data were analyzed descriptively comprising basic characteristics of research subjects, the results of anthropometric and cephalometric measurements. Numerical data were expressed in mean ± standard deviation when data were normally distributed, or median (lowest to highest) when data were not normally distributed. Data were displayed in Tables. Furthermore, the data were analyzed using SPSS version 24.0.0.

Ethical clearance

Approval from the Ethics Committee of Dr. Soetomo Hospital was proposed before the implementation of the study. Patients' consent declaration is disclosed in the form of informed consent signed by the patients' parents. Patients and their families are not imposed and are not burdened with any fees associated with this study. Identity data and examination results are withheld from unauthorized parties.

RESULTS

The number of studied patients were 81, consisting of 43 (53.1%) males and 38 (46.9%) females. The oldest age was 20 years, the youngest was 1 month, the average patients' age was 3 years 9 months and most of them were less than 1 year. Interval age group can be seen in Figure 1.

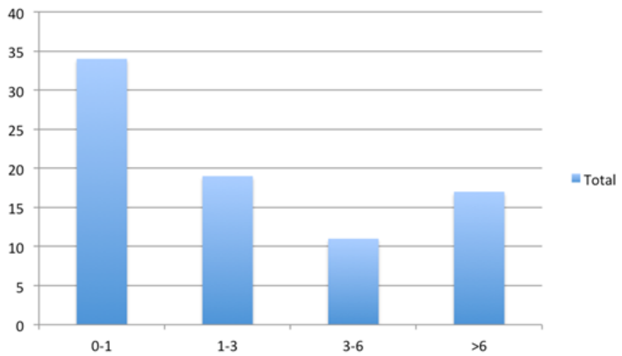

Figure 1. Age group distribution

All patients had a lump near nasal root at birth. This mass size remained constant in 2 patients, grew rapidly in 4 patients and grew slowly in 75 patients (92.6%). FEE wall pulsation was found in 43.6% of cases, positive transillumination in 45.5% of cases, and congenital stigmata in the skin of the cover was found in 7.3% of cases. The local status of FEE by size indicated that the smallest size was 1 cm^3 (nasoorbital case), the largest is 1800 cm^3 (nasofrontal case), averaging 71 cm3 and the most was 2-36 cm3. The rate of growth, type, consistency, and location can be seen in Table 1. Eye distortion was found in 19 patients (23.46%). Nasal compression by encephalocele resulting in airway obstruction was present in 2 cases. Hydrocephalus was present in 7 patients, 4 of whom with rapid encephalocele growth.

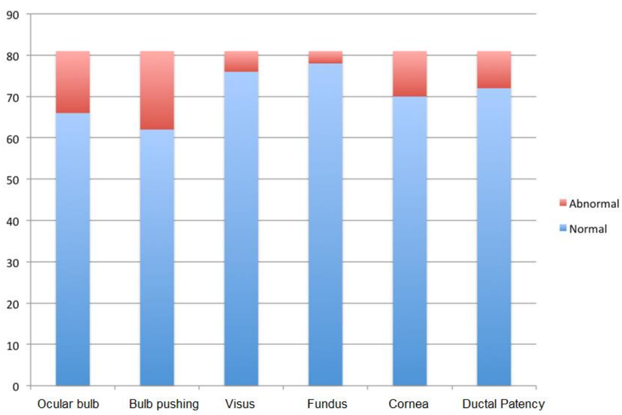

Fig. 2. Eye and adnexal conditions.

Neurological examination revealed that there were 7 patients with nystagmus, no patients with seizures, 15 patients with microophtalmia, 1 patient with microcornea, 5 patients with preoperative vision disorder, 3 with opacification of eye media and 9 with nasolacrimal duct obstruction. The circumstances of the eyes and their adnexa from each group, the ocular bulb, ocular bulb pushing, visus, funduscopy, corneal abnormalities, and nasolacrimal ductal abnormalities, can be seen in Figure 2. Based on anthropometric and cephalometric techniques, we obtained the results as in Table 2.

Table 1. FEE local status

Local status		Total	%
Growth speed	Constant	2	2.47
	Slow	75	92.59
	Rapid	4	4.94
Types	NO	12	14.81
	NF	21	25.93
	NE	48	59.26
Consistency	Cystic	22	27.16
	Dense	4	4.94
	Mixed	55	67.90
Location	Left	14	17.28
	Right	28	34.57
	Middle	21	25.93
	Bilateral	18	22.22

Table 2. Anthropocephalometric results of the patients

	Values	Mean
IW	15-50 mm	35 mm
BW	61-124 mm	96,5 mm
CW	95-195 mm	143 mm
IW/BW	7.1-25.9%	16,6%
LLC	15-83%	52%
IOD	11-37 mm	24.7 mm
EOD	65-125 mm	85.7 mm
ID	30-250 mm^2	73 mm^2
ED	20-300 mm^2	122 mm^2

Note: IW: intercanthal width, BW: biocular width, CW: cranium width, IOD: interorbital distance, EOD: external orbital distance, ID: Internal defect, ED: external defect

Statistical test was performed by Lambda test to determine the correlation between age and with IOD and correlation netween age and IOD/CW ratio. Apparently, there was correlation between JlO distribution and age. Fig. 3 shows the IOD-age correlation in FEE patients with normal populations. From the statistical test using Lambda test it was found that there was correlation between age and IOD/CW. Fig. 4 shows the age correlation with the IOD/CW ratio.

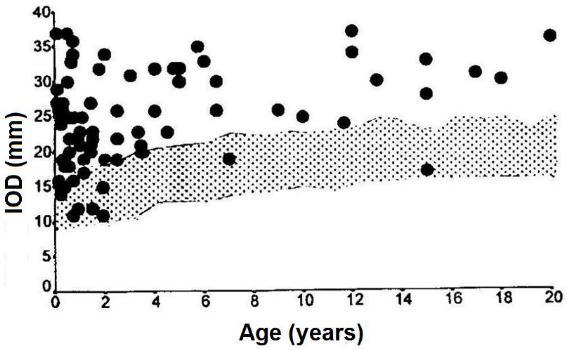

Fig. 3. Correlation between IOD and age. Black dots are IOD values of the patients, while shaded area is the normal values.

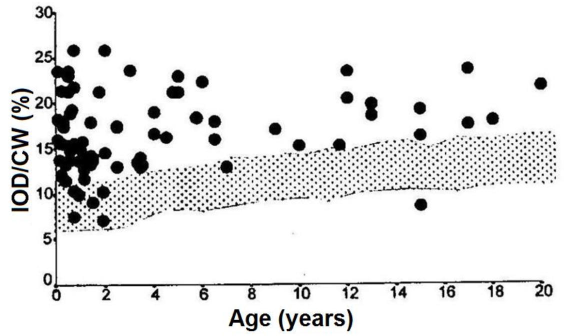

Fig. 4. Correlation between between IOD/CW ratios (%) and age. Black dots are IOD/CW (%) of the patients, while shaded area is the normal values.

DISCUSSION

Facial anthropometric measurements are different in each race or ethnicity. Therefore, as a reference in the handling of craniofacial abnormalities in a particular race, first there should be a database of normal values measurements (Arumsari et al 2004). Data obtained in this study were compared with those obtained by Arumsari et al (2004) who examined anthropocephalometric profile of Mongoloid race, subrace Deutero-Malay. From anthropometric data of FEE patients, it is known that the intercanthal distance is widened, but biocular distance is normal (Arumsari et al 2004. Tessier 1976), medial intercanthal line position changes from normal (Arumsari et al 2004, Tessier 1976), IOD widens without EOD change (Arumsari et al 2004, Mafee et al 1986, Tessier 1976), and IOD widens with age, indicating that the later the correction, the wider the IOD than that in children his age.

Until this study was made, there has never been a report that measures anthropometry and cephalometry of patients who would undergo corrective surgery. Photographic results in 2 dimensions and 3 dimensions are objective methods for analyzing the results of cephalometry, although it requires appropriate equipment and techniques for photographic polarization. Minimal changes in distance between the photograph and the patient can cause changes in the final measurement calculated in each cephalometric photo. Therefore, using value ratios can reduce errors due to distance changes between the photographer and the patient. From the quantification method of the results suggested in the literature, quantification of measure-ments through 2D photography is preferred, as it is an objective, reliable, and reproducible method at a cost not as high as that in 3D photographs. If there is difficulty performing standard radiographs to measure cephalometry, the skull base CT and orbital CT may also be used for cephalometric measurement (Mafee et al 1986). For surgical vision, head 3DCT is particularly useful for measuring cephalometry and observing deformation of the skull base to facial bone.

Anthropocephalometric problem of FEE is due to the complex problems of internal defect (ID) and ends in craniofacial bone known as the external defect (ED) and forms a bone duct, and there is brain tissue and pulsating brain fluid, causing deformity of the craniofacial structure, eyes and their adnexa, bone and cartilage of the middle area of the face (Hoving & Vermeij-Keers 1997).

ID in FEE patients is the lowest point of the anterior cranial fossa. Lamina cribrosa is always lower than normal, so the value of LLC ratio is always higher than normal and this lamina does not rise relative to age. This is in accordance with the research results of Costaras & Pruzansky (1982). This may be due to the presence of anterior growth of the maxilofacial portion of the face.

The anterior cranial fossa shift and the nasal cartilage at the midline, the rise of the superciliary arcus and the decrease of the nasal tip result in a long nose profile of nasophenoid type of FEE. IOD widens throughout FEE, while EOD remains normal. This means hypertelorism in FEE patients is interorbital hypertelorism, not true hypertelorism, so that surgical reconstruction no longer requires orbital translocation (Costaras & Pruzansky 1982) but requires only medial orbital wall shifts to medial direction (Holmes et al 2001).

CONCLUSION

Obtaining surgical outcome that reaches the target of bone defect closure, avoiding leakage of brain fluids as well as obtaining the aesthetics of the face according to anthropocephalometric standard, in patients with FEE undergoing correction, is a major challenge for neurosurgeons. In order to obtain good results on FEE patients care, a neurosurgery should have access to the database of anthropometric craniofacial measurements of the appropriate racial/national population. Therefore, normative data from craniofacial anthropometrics is of great importance in determining the degree of facial abnormalities compared with the normal population. In the case of FEE, based on anthropometric and cephalometric examinations, the majority of patients should be corrected by simple orbital medial shift techniques, without the need for radical correction of facial bone.

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