Pediatric Neurology 71 (2017) 29–34

Contents lists available at ScienceDirect

Pediatric Neurology

journal homepage: www.elsevier.com/locate/pnu

Original Article

Growth and Psychological Development in Postoperative Patients With Anterior Encephaloceles

Hemonta Kumar Dutta MS, MCh [a,*], Chow Wachana Khangkeo MS [a], Kaberi Baruah MSc [b], Debasish Borbora MSc [c]

[a] *Department of Pediatric Surgery, Assam Medical College & Hospital, Dibrugarh, Assam, India*
[b] *Department of Psychiatry, Assam Medical College & Hospital, Dibrugarh, Assam, India*
[c] *Department of Biotechnology, Gauhati University, Guwahati, Assam, India*

ABSTRACT

PURPOSE: Anterior encephaloceles are rare malformations that are frequently associated with other brain anomalies. This study evaluates the growth and psychological development of children following encephalocele repair. **MATERIALS & METHODS:** Growth and psychological assessment was done in 24 children with only encephalocele (group I); nine children with encephalocele and hydrocephalus (group II); seven children with encephalocele, hydrocephalus, and secondary malformations (group III); and 40 apparently healthy control subjects. Psychological assessment was done by evaluating intelligence and temperament. **RESULTS:** Single-stage repair was performed in 38 children, and two underwent multistage repair. Major postoperative complications were noted in three individuals. The follow-up period ranged from 12 to 168 months, and during this time the growth velocity declined significantly among group II and group III patients when compared with control subjects. After adjusting the body mass index for age, our data revealed that group III participants had a significantly ($P = 0.02$) lower body mass index than the control group. Group III also had poor indices for intelligence quotient ($P \leq 0.01$) and temperament ($P \leq 0.01$). Female patients had lower temperament indices when compared with unaffected females with regard to approach withdrawal ($P \leq 0.01$), mood ($P = 0.026$), and intensity ($P = 0.03$). Overall, increased disease severity adversely affected the psychological indices. **CONCLUSION:** Individuals with anterior encephalocele without associated intracranial defects had excellent postoperative outcomes in terms of growth and psychological developments. Hydrocephalus and agenesis of corpus callosum had the least impact on psychological development. However, the presence of secondary brain defects led to developmental delays. Gender differences in temperament may suggest a need for distinct treatment regimens to assess psychosocial well-being for males and females.

Keywords: anterior encephalocele, hydrocephalus, holoprosencephaly, schizencephaly, agenesis of corpus callosum, physical, psychological growth, surgical repair

Pediatr Neurol 2017; 71: 29-34

Introduction

Anterior encephaloceles are midline frontonasal herniations of brain or meninges through a skull defect.[1] Von Meyer and Whatmore classified anterior encephaloceles into sincipital and basal and further into nasofrontal, nasoethmoidal, and naso-orbital encephaloceles. In India, anterior encephaloceles are common among the tea garden workers in Assam.[2] The ideal treatment for these malformations is transcranial repair, followed by repair of the external sac.[3-5] The heterogeneity of physical and psychological outcomes associated with neural tube defects stems from multiple influences ranging from genes to environment that interact in a complex way.[6] Because of this heterogeneity, a stratified analysis of the various subtypes to assess cognitive morbidity is essential. Children with these defects demonstrate an array of neuropsychologic deficits when compared with typically developing children, but the outcomes are variable. Because anterior encephalocele is rare,

Article History:
Received September 26, 2016; Accepted in final form January 29, 2017
* Communications should be addressed to: Dr. Dutta, Department of Pediatric Surgery; Assam Medical College & Hospital; Dibrugarh, Assam 786002, India.
E-mail address: hemontdut@gmail.com

0887-8994/$ – see front matter © 2017 Elsevier Inc. All rights reserved.
http://dx.doi.org/10.1016/j.pediatrneurol.2017.01.029

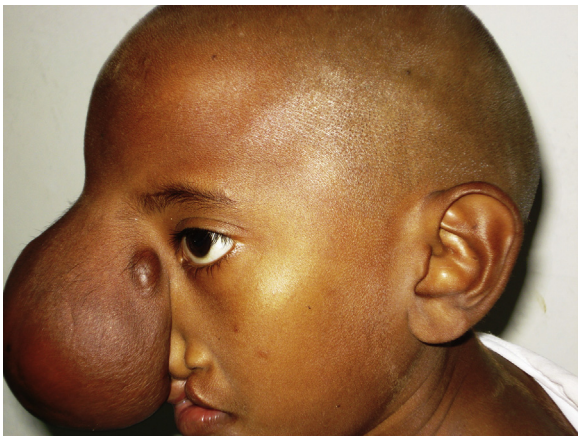

FIGURE 1.
Characteristic pre-surgical facial appearance of a child with an anterior encephalocele. (The color version of this figure is available in the online edition.)

there are no reports evaluating the intelligence and cognitive indices in patients with or without associated secondary intracranial defects. The present study was carried out to assess the growth and psychological development of post-operative patients with anterior encephaloceles.

Materials and Methods

Ethical statement

This study was approved by our institutional ethical committee. Written consent was obtained from the parents or guardians of the participating children.

Study population and design

This case-control study involved 40 children with an anterior encephalocele (Fig 1) who underwent reconstructive surgery at the pediatric surgery department at Assam Medical College and Hospital, Dibrugarh, between June 2002 and December 2014 and consented to participate in this study between January 2013 and December 2015. The control group consisted of 40 age- and sex-matched apparently healthy children from the same ethnicity. Computed tomography/magnetic resonance imaging was done to identify associated intracranial anomalies, which may have a bearing on the future growth and development of the child. Patients were grouped into the following categories (Table 1): 24 children with only encephalocele (group I), nine children with encephalocele and hydrocephalus (group II), and seven children with encephalocele, hydrocephalus, and secondary malformations (group III). To collect relevant information, we administered a precoded close-ended questionnaire.

Growth velocity[7] was measured by recording the height and weight of the patients and their parents during the follow-up period, which ranged from 12 to 168 months. The adjusted midparental height (target height) of the patients at age 18 years was calculated (in centimeters) using the formula: (father's height + mother's height + 13)/2 for boys and (father's height + mother's height − 13)/2 for girls, and these values were plotted in the Indian Academy of Pediatrics growth chart used for urban Indian children.[8] Parental body mass index (BMI) was calculated by dividing the weight in kilograms by the height in meters squared (kg/m^2) and classified according to the World Health Organization BMI cutoff points. For children, the BMI-for-age percentile was used to interpret "weight status categories." We used the Centers for Disease Control and Prevention online Child and Teen BMI calculator.[9]

Psychological and cognitive assessment was done by a clinical psychologist by interviewing the mothers. The study relied on the Vineland Social Maturity Scale (Indian adaptation) for assessing intelligence. In brief, the scale evaluated eight social areas, namely, self-help general, self-help eating, self-help dressing, self-direction, occupation, communication, locomotion, and socialization to estimate the social age and social quotient of the patients and shows high correlation with the individual's intelligence quotient (IQ).[10,11] The IQ was then classified according to the Wechsler Intelligence Scale for Children.[12] To assess temperament, a Temperament Measurement Schedule devised for Indian children was used.[13,14] The schedule measures nine temperament variables (approach withdrawal, adaptability, threshold of responsiveness, mood, persistence, activity, intensity, distractibility, and rhythmicity), each consisting of five items, and rates them on a five-point scale. Mean scores for each of these variables were computed by dividing the total score by five. These nine temperamental variables were then reduced to five independent temperament dimensions (sociability, emotionality, energy, distractibility, rhythmicity).

Statistical analysis

Data were analyzed using IBM SPSS Statistics 20 (SPSS Inc, Chicago, USA) and Epi Info 7 (Centers for Disease Control and Prevention). Mann-Whitney *U* test was used to compare the mean temperament values. Chi-square test was done wherever applicable. One-way analysis of variance (ANOVA) was conducted to compare the effects of associated intracranial defects on the patient's cognitive abilities. Posthoc analyses were carried out using the Tukey multiple comparisons test.

Results

Demographic data

The age of the children ranged from four to 16 years (x = 8.65 years in cases; x = 8.66 years in control subjects). The male to female ratio was 0.90 (19/21), and the majority of them belonged to the tea garden community (n = 36, 90%). Thirty-seven children in the test group and 38 children in the control group were from lower socioeconomic strata (household income of 5000 or fewer Rs. per month).

TABLE 1.
Clinical Profile of Anterior Encephalocele Cases (n = 40)

Clinical Condition			N (%)
Encephalocele cases without associated intracranial defects (group I)			24 (60%)
Cases with associated intracranial defects (n = 16)	Only hydrocephalus (group II)		9 (22.5%)
	Group III	Hydrocephalus with corpus callosum agenesis	2 (5%)
		Hydrocephalus with dchizencephaly and arachnoid cyst	2 (5%)
		Hydrocephalus with holoprosencephaly with corpus callosum agenesis	2 (5%)
		Hydrocephalus with large posterior fossa cyst with corpus callosum agenesis	1 (2.5%)