Environmental Risk Factors for Anterior Encephaloceles: 15 Years’ Experience at a Tertiary Centre in India

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Abstract
Anterior encephalocele (AE) is a rare congenital malformation in the central nervous system with a reportedly high prevalence in Assam, Northeast India. As there exists substantial variations in the etiology of the disease, determination of risk factors and timely intervention could reduce associated morbidity. A retrospective chart review of patients with AE who were born at or referred to Assam Medical College & Hospital (AMCH), a tertiary health care centre between January 1999 and December 2013 was performed to assess the environmental risk factors associated with AE. 64 patients were treated at the pediatric surgery department during the period with a male to female ratio of 1:1.2 and mean age of 36 months. 62.5% patients had naso-ethmoidal followed by 31.3% naso-frontal and 6.2% naso-orbital encephalocele. 61 cases were of the tea garden community with low socio economic status. Parental exposure to pesticides was reported in 58 cases. Maternal age < 20 years and >39 years accounted for 40.6% of the cases. 73.5% of the mothers had parity ≥3 and 15 reported of past abortions. Maternal malnutrition was observed in 38% cases and parental consanguinity was noted in 12 cases. Thus the study suggest that low socioeconomic status, pesticides exposure, advanced maternal age, increasing parity, parental consanguinity and maternal malnutrition may contribute to the occurrence of AE.

Keywords: Anterior encephalocele; Sincipital encephalocele; Congenital malformations; Risk factors; Assam

Introduction
Anterior encephaloceles (AE) are rare forms of neural tube defects (NTD) which are characterized by the herniation of brain tissue through a congenital defect in the skull. Based on their location they are subdivided into sincipital and basal. Sincipital encephaloceles are further classified into nasofrontal, nasoethmoidal, and nasoorbital.

Although sporadic in the western world, the condition is frequent in few Southeast Asian countries like Myanmar and Malaysia [1]. In India however, this malformation is seen frequently among the tea garden communities of Assam [2,3]. As NTD show marked ethnic and geographic variations, the role of environmental factors causing these malformations could be significant. Accordingly, WHO has outlined maternal diabetes, alcohol abuse, antenatal X-irradiation, aminopterin and hallucinogen ingestion as known risk factors of NTD. Tea garden communities in Assam are socioeconomically backward and they are exposed to various chemicals used in tea gardens. So this study was conducted among patients attending Assam Medical College & Hospital (AMCH), a tertiary health care centre in Assam (Northeast India) to identify possible environmental risk factors of AE.

Materials and Method
A retrospective chart review of patients with AE who were born at or referred to AMCH between January 1999 and December 2013 was performed to evaluate various lifestyle and environmental risk factors which may contribute to the disease. This study was approved by the institutional ethical committee and consent was obtained from all participating parents. Parental BMI (kg/m²) was classified according to the criterions suggested by WHO for the Asian populations. In case of children, the BMI-for-age percentile was used to interpret ‘Weight Status Categories’.

Results
64 patients were treated at the Pediatric surgery department during the 15 years period. The number of female cases was marginally higher than the males at a ratio of 1:2:1 with a mean age of 36 months (1 month-14 years). 40 children had naso-ethmoidal, 20 children had naso-frontal and 4 had naso-orbital encephaloceles (Figure 1). Of the 64, 61 (95.3%) cases were of the tea garden community with either one
The use of weedicides and pesticides is widespread in tea gardens. Pesticides such as organophosphates and pyrethrinns are known to cause neurotoxicity by inhibiting acetylcholine activity [4]. In utero exposure to some pesticides can lead to psychomotor developmental delay in the child [5]. Studies have also associated parental exposure to pesticides with the risk of NTD [6-8]. In the present series, one or both the parents of 58 children reported exposure to various agricultural pesticides and weedicides. Male members were mostly involved in spraying of pesticides. It was also significant to note that families of all the 64 children lived in huts either in the midst or in the vicinity of tea gardens and it is likely that the water and food they consume could also be contaminated with pesticides.

Advanced maternal age was also a distinct risk factor in this study. According to Vaughan et al. children born to mothers aged ≤17 years and ≥40 years were at a greater risk of having a congenital anomaly [9]. A systematic review analyzing risk factors associated with stillbirth in high-income setting also concluded that maternal age >35 years is a major modifiable risk factor [10]. Similar observations were also reported during independent studies conducted by Suphapattiporn et al. and Mashuda et al. [11]. In the present series too, 22 children were born to mothers aged < 20 years and 4 to mothers aged ≥40 years. This is in corroboration with finding by Taksande et al. from Maharashtra [12] and Grover et al. from Shimla [13]. Previous studies have associated grand multiparity with the risk of congenital malformations [13,14,11] but in their systematic review Shah et al. did not observe any association between grand multiparity, great grand multiparity and pregnancy outcomes [15]. In this study however, 56% of the mothers had parity of 3 or more and 17% had parity more than 5, indicating a possible association. We believe that advanced maternal age along with multiparity could result in unfavorable intrauterine environment causing AE among the newly born.

Maternal nutrition is a modifiable risk factor associated with adverse birth outcomes, particularly in developing economies/low-income populations [16]. In the present series, 35% of the mothers had BMI <18.5, 53.1% were anemic and 65.6% mothers had serum albumin levels <3.5gm/dl. These figures clearly indicate that low socio economic status had a bearing on the mother's diet during pregnancy which might have affected the growing fetus. It is known that supplementing 4 mg folic acid per day for at least one month prior to conception to 3 months post conception reduces the risk of recurrence of NTD by 70% [17], but in the present series, mother of 30 children reportedly did not receive folate supplementation during pregnancy and 32 mothers consumed folic acid only after 3 months of pregnancy.

In this report, 61 cases were from the tea garden community of which 12 were born to consanguineous parents. As the association of birth defects with consanguinity is known [18], intrafamilial marriages within this community could explain for the high incidence of AE as it increases the prevalence of homozygotes for autosomal recessive genetic disorders. The study concludes that low socioeconomic status, pesticides exposure, advanced maternal age, increasing parity and maternal malnutrition may contribute to the occurrence of AE. Although there were no clustering of AE cases in this present study, 90% of the patients hailed from the 6 upper Assam districts, where density of the tea gardens are one of the highest in the world.

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Conflict of Interest
None declared

Contribution of Authors
The study was designed by Dr. Hemonta Kumar Dutta. Both the authors contributed substantially towards the collection, compilation and interpretation of data.

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References