JOURNAL OF CASE REPORTS 2015;5(2):503-505

Recurrent Life Threatening Events in a Neonate

Rahul Kadam, VSV Prasad
Lotus Hospitals for Women and Children, Lakdikapul, Hyderabad- 50004, Telangana, India.

Abstract:

Introduction: Breath holding spells are a common disease entity in children and mostly occur between 6-18 months of age and terminate by 3-4 years. *Case Report:* We present a case of a neonate with cyanotic breath holding spells which presented as recurrent apparent life threatening events. Infant was evaluated for possible causes of recurrent life threatening events and diagnosed as cyanotic breath holding spells. *Conclusion:* We have reported this case to highlight that breath holding spells can present early in the life and needs to be considered in the differential diagnosis of any neonate presenting with cyanotic spells.

Key words: Breath Holding, Child, Cyanosis, Infant, Respiratory Disorders.

Introduction

Breath holding spells (BHS) are a common disease entity in children and mostly occur between 6-18 months of age and terminate by 3-4 years [1]. Onset below 6 months of age is seen in 13-18% of patients [1,2]. We present a case of a neonate with cyanotic breath holding spells which presented as recurrent apparent life threatening events.

Case Report

This preterm 29 weeks, 1100 grams birth weight baby, required resuscitation at the birth and developed respiratory distress soon after birth. Infant's clinical and X-ray findings were consistent with respiratory distress syndrome. Infant received one dose of surfactant and was ventilated for a day followed by CPAP support for 3 days. Infant's 2D Echo on day 3 revealed large patent ductus arteriosus (PDA) which was treated with two courses of ibuprofen followed by one course of paracetamol. Infant required packed red blood cell transfusion twice in view of anemia. Orogastric tube feeds were initiated on day 2 of life and were graded up as tolerated. Oral feeds were initiated at corrected gestational age (CGA) of 36 weeks (day 53 of life) and breast feeds were initiated on day 58 of life. Infant was transferred to room on day 62 of life (CGA 37 weeks). Infant's weight at transfer was 1.68 kg.

On day 66 of life (CGA 38 weeks) infant developed apparent life threatening event (ALTE): apnea and bradycardia which required chest compressions and bag and mask ventilation, infant was transferred back to NICU and was evaluated for possible causes. Infant's sepsis screen was

Corresponding Author: Dr. Rahul Kadam
Email: drrahul19@gmail.com
Received: September 25, 2015 | **Accepted:** October 28, 2015 | **Published Online:** November 25, 2015

Conflict of interest: None declared | **Source of funding:** Nil | **DOI:** http://dx.doi.org/10.17659/01.2015.0129

negative, metabolic parameters were normal, Neurosonogram was unremarkable. Infant's barium study revealed presence of grade III gastro-esophageal reflux disease (GERD). Infant was managed conservatively with medical management and anti-reflux measures. Breast feeds were reintroduced on stabilisation of the condition and infant was transferred back to the room on day 75 of life (CGA 39 weeks).

On day 80 of life, infant again had similar episode of apnoea, bradycardia and desaturation, was transferred to NICU where infant had multiple episodes of seizures. Seizures were controlled with phenobarbitone. Infant was evaluated for possible causes. Infant's sepsis screen was positive and CSF study was suggestive of meningitis. Infant's MRI brain was normal while EEG was abnormal with multifocal epileptic activity. Infant was treated with intravenous antibiotics for 3 weeks. Infant continued to have intermittent apnoeic episodes. In view of recurrent ALTE, repeat barium study was performed which revealed persistence of grade III GERD. Infant underwent fundoplication with gastrostomy tube in situ on day 92 of life. Infant was discharged home on day 107 (CGA 44 weeks) of life: was seizure free, on gastrostomy tube feeds, gaining weight adequately and weight at discharge was 2.435 kg.

Infant was readmitted on day 114 of life (CGA 45 weeks) with history of 3 episodes of excessive cry followed by cessation of breathing and bluish discoloration. Possibilities considered were persistent GERD, infantile epilepsy syndrome, sepsis, vascular ring and cyanotic breath holding spells. Infant was again evaluated for possible causes. Infant's sepsis screen was negative, metabolic parameters were normal. Infant's repeat EEG was normal. Milk scan revealed no evidence of GERD. 2D ECHO was normal. CT angiography revealed no evidence of vascular rings. Infant's haemoglobin level was low.

During hospital stay infant had four similar episodes with spontaneous resolution. Infant's history and clinical features were consistent with cyanotic breath holding spells. Infant's gastrostomy tube was removed, was started oral feeds followed by breast feeds. Infant was accepting feeds well with adequate weight gain. Parents were reassured and counselled regarding benign nature of the illness. Infant was discharged home on iron supplementation and piracetam.

Infant is being followed in out-patient department at regular intervals. The frequency of the breath holding episodes reduced over time. Infant's repeat EEG at 3 months is normal and neurologic examination is normal for age. Infant's growth and development is appropriate for age.

Discussion

Breath holding spells are generally benign, but may cause intense parental concern. In 1967, Lombroso and Lerman [1] in a study of 5000 patients identified 225 patients with severe breath holding spells amounting to a prevalence of approximately 4.6%. Breath holding spells commonly occur between the ages of 6 months and 2 years. The spells usually start before 18 months of age and stop by six years. These spells are a reflex reaction to an unpleasant stimulus and not a deliberate behavior on the child's part. In epileptic seizures, the child may turn blue, but it will be during or after the seizure and not before. A child may lose control of their bladder and bowels during an epileptic seizure, but this is rare with breath holding spells. The age of onset of BHS in most of the studies has been in the first 12 months of life [2].

The pathophysiological mechanisms of BHS remain controversial. The presence of an underlying dysfunctional autonomic nervous system with cerebral anoxia or cerebral ischemia resulting from

vagus mediated cardiac arrest may be responsible for the condition [3-6]. The maturation delay in myelination of the brainstem could be the cause of breath-holding spells in children [7].

What provoked our patient to have BHS at such an early age remains unknown. Although BHS can occur in neonates with a family history [4], the same was absent in our patient. The hemoglobin and hematocrit in the patient were in the lower range. Subclinical anemia could have precipitated the condition in this neonate with a dysfunctional autonomic nervous system. This is emphasized by the response to the iron therapy shown by the infant. It has been shown that iron therapy can cause significant reduction in the frequency of spells [8,9]. In most patients with BHS, spontaneous recovery occurs by 36- 42 months of age [2,3].

Conclusion

BHS can present early in the life and needs to be considered in the differential diagnosis of any neonate presenting with cyanotic spells. Iron therapy may be considered in the termination of BHS in neonates.

References

1. Lombroso CT, Lerman P. Breath holding spells (cyanotic and pallid infantile syncope). Pediatrics. 1967;39:563-581.
2. DiMario FJ Jr. Prospective study of children with cyanotic and pallid breath holding spells. Pediatrics. 2001;107:265-269.
3. Bhat M, Ali W, Mohidin K, Sultana M. Prospective study of breath holding spells and role of iron. J Pediatr Neurol. 2007;5:27-32.
4. Breukels MA, Plötz FB, van Nieuwenhuizen O, van Diemen-Steenvoorde JA. Neuropediatrics. 2002;33(1):41-42
5. Orii KE, Kato Z, Osamu F, Funato M, Kubodera U, Inoue R, *et al*. Changes of autonomic nervous system function in patients with breath-holding spells treated with iron. J Child Neurol. 2002;17:337-340.
6. Kolkiran A, Tutar E, Atalay S, Deda G, Cin S. Autonomic nervous system functions in children with breath-holding spells and effects of iron deficiency. Acta Paediatr. 2005;94:1227-1231.
7. Vurucu S, Karaoglu A, Paksu SM, Oz O, Yaman H, Gulgun M, *et al*. Breath-holding spells may be associated with maturational delay in myelination of brain stem. J Clin Neurophysiol. 2014;31(1):99-101.
8. Mocan H, Yildiran A, Orhan F, Erduran E. Breath holding spells in 91 children and response to treatment with iron. Arch Dis Child. 1999;81(3):261-262.
9. Daoud AS, Batieha A, al-Sheyyab M, Abuekteish F, Hijazi S. Effectiveness of iron therapy on breath-holding spells. J Pediatr. 1997;130(4):547-550.