UNUSUAL PRESENTATION OF CYCLIC VOMITING SYNDROME: A CASE REPORT.

Manas Ranjan Sahoo[1], Majeti Srinivasa Rao[2], A. Vasundhara[3], P. Sudarsini[4], K. Umamaheswra Rao[5]

HOW TO CITE THIS ARTICLE:
Manas Ranjan Sahoo, Majeti Srinivasa Rao, A Vasundhara, P Sudarsini, K Umamaheswra Rao. "Unusual presentation of cyclic vomiting syndrome: a case report". Journal of Evolution of Medical and Dental Sciences 2013; Vol2, Issue 48, December 02; Page: 9280-9282.

ABSTRACT: We here with report a 10 month old infant with recurrent episodes of vomitings. All necessary investigations were done and are found to be normal. Hence a diagnosis of Cyclic vomiting syndrome (CVS) was made. It is unusual for CVS to present under the age of 1 year, hence we are reporting this case.
KEYWORDS: vomitings, dehydration, hypokalemia.

INTRODUCTION: Cyclic vomiting syndrome (CVS) is a functional disorder that is considered to be a manifestation of migraine diathesis. It is characterized by stereotypical episodes of severe nausea and vomiting lasting several hours or days, with return to baseline health between episodes. CVS is still an insufficiently known syndrome among physicians, and is therefore often misdiagnosed. Treatment focuses on the different phases of CVS, with inter episodic prophylaxis, abortive therapy in the prodromal phase of CVS, and supportive care during an acute vomiting episode.

Cyclic vomiting syndrome (CVS) is defined as fits of self-limiting vomiting, lasting minutes to hours to even days at a time, which are reoccurring[1,2] and affect mainly preschool children. It is second only to gastroesophageal reflux as the most common cause of recurrent vomiting,[2] affecting 1.9% of school-aged children. It has also been described as a migraine-equivalent disease; and in one third of the patients, the CVS will evolve to a migraine headaches. The symptoms are usually relieved by sleep, but most children will continue vomiting after they awake.[1] It is important, when caring for these children, to remember that during the symptomatic episode, the child may become dehydrated and dangerously ill.[1]

CASE REPORT: A 10 months old female child, product of non-consanguineous marriage, presented to our hospital with 6 episodes of recurrent severe vomiting, several of them being managed locally in other hospitals over a period of 3 months. Child was admitted in our hospital with sudden onset of non-bilious vomiting, frequency varying from 2-6 episodes per hour lasting for 2-3 days for 3 times. Each time she was severely dehydrated, eyes sunken, lethargic not able to drink. Reviewing history mother informed that she was absolutely fine in between these episodes which varies from 1-2 weeks.

On investigation hyponatremia (s.Na:125mEq/l), hypokalemia (s.K+: 2.5mEq/l), and metabolic alkalosis (pH:7.86) were found. Initially acute episode was treated with NS fluid bolus followed by correction of dehydration. Then hyponatremia was corrected slowly over 24 hrs. Hypokalemia was corrected by iv infusion containing KCl, followed by oral supplementation. Ondansetron 1.2mg iv 8th rly was administered to control vomiting for 3 days. After resuscitation USG abdomen was done which was normal. Barium meal was also normal. Serum ammonia, lactate were within normal limits.17 hydroxy progesterone, serum cortisol taken at 8AM were normal.

Tandem Mass Spectroscopy (screening test of metabolic disorder) for urea cycle defect, organic aciduria ,fatty acid oxidation disorder, amino acid disorder was also normal. Her elder brother also had similar history of recurrent severe vomiting starting around 6month of age, requiring multiple admissions, which subsided by 2 years of age. There was no family history of migraine .Child was discharged with oral cyproheptadine 1mg 12th hrly for prophylaxis..In our case we could not find any triggering factor for such episode. There was good response to cyproheptadine. Parents were counseled regarding good outcome of this disease in long term once acute episodes were managed successfully.

DISCUSSION: Cyclic vomiting syndrome was first described by Samuel Gee in 1882 (3) and named cyclic vomiting by Smith in 1937 (4). CVS is characterized by recurrent, sudden, stereotypical, disabling, discrete episodes of intense nausea and vomiting that can last a few hours to days interspersed with varying weeks of symptom-free intervals.

The estimated prevalence of CVS in children is in the range of 0.3%–2.2 % (5). This disorder is primarily recognized in children, primarily Caucasians (mean age of onset at five years), with increasing recognition in adults (mean age of onset at 35 years). There have been case reports of symptoms starting as early as the sixth day of life and as late as 73 years. In children, females appear to be more affected than males, compared to a male predominance in adults (6,7).

CVS is now considered to be a functional brain-gut disorder in which central signals initiate a peripheral gastrointestinal manifestation—vomiting. There appear to be a number of host susceptibility factors including a family member with migraine headaches (82% of CVS versus 14% of chronic vomiting patients), mitochondrial dysfunction, and autonomic dysregulation. There is a strong matrilineal inheritance of CVS from migraines, elevated lactic acid, and several heteroplasmies in the control region of the mtDNA supporting involvement of mtDNA (8). There is also heightened sympathetic cardiovascular tone in children with CVS compared to controls. These factors taken together suggest that in adequate cellular energy production at times of heightened needs (trigger factors mentioned below) leads to a metabolic crisis. This in turn leads to a deleterious effect on high energy requiring autonomic neurons resulting in an episodic autonomic crisis with vomiting.Similar to migraines, there appear to be common triggering factors including psychological stress (especially excitement) and infections potentially triggers vomiting.

Criteria for Cyclic vomiting syndrome (2008): At least 5 attacks in any interval, or a minimum of 3 attacks during a 6-mo period Episodic attacks of intense nausea and vomiting lasting 1 h–10 days and occurring at least 1 wk apart. Stereotypical pattern and symptoms in the individual patient. Vomiting during attacks occurs at least 4 times/h for at least 1 h. Return to baseline health between episodes Not attributed to another disorder.

As our case met most of the criteria, and as all other investigations were normal, the diagnosis of Cyclic vomiting syndrome, was made.

CONCLUSION: Here onset of presentation was at early age of 8 months, with family history of similar attack suggesting hereditary link could be there in this disease. This case depicts the severity of presentation and acute management as well as prophylactic treatment.

REFERENCES:

1. Stein M.T., Katz R.M., Jellinek M.S., Cassidy L., Olness K. Cyclic vomiting. Pediatrics.2001;107(4):940–944.
2. Haghighat M., Rafie S.M., Dehghani S.M., Fallahi G.H., Nejabat M. Cyclic vomiting syndrome in children: experience with 181 cases from southern Iran. World J Gastroenterol. 2007;13(12):1833–1836.
3. Gee S. On fitful or recurrent vomiting. St. Bart Hosp Rep,1882;18:1.
4. Smith PS. Cyclic vomiting and Migraine in children. Vir Med Month, 1934;1:591-593.
5. Abu-Arafeh I, Russell G. Cyclical vomiting syndrome in children: a population-based study. J Pediatr Gastroenterol Nutr,1995;21(4): 454-458.
6. Namin F, et al. Clinical, psychiatric and manometric profile of cyclic vomiting syndrome in adults and response to tricyclic therapy. Neurogastroenterol Motil, 2007;19(3):196- 202.
7. Li BU, Misiewicz L. Cyclic vomiting syndrome: a brain-gut disorder. Gastroenterol Clin North Am, 2003;32(3):997-1019.
8. Boles RG, Adams K, Li BU. Maternal inheritance in cyclic vomiting syndrome. Am J Med Genet A, 2005;133(1):71-77.

AUTHORS:

1. Manas Ranjan Sahoo
2. Majeti Srinivasa Rao
3. A. Vasundhara
4. P. Sudarsini
5. K. Umamaheswra Rao

PARTICULARS OF CONTRIBUTORS:

1. Assistant Professor, Department of Paediatrics, Alluri Sitarama Raju Academy of Medical Sciences.
2. Associate Professor, Department of Paediatrics, Alluri Sitarama Raju Academy of Medical Sciences.
3. Professor, Department of Paediatrics, Alluri Sitarama Raju Academy of Medical Sciences.
4. Professor & HOD, Department of Paediatrics, Alluri Sitarama Raju Academy of Medical Sciences.
5. Professor, Department of Paediatrics, Alluri Sitarama Raju Academy of Medical Sciences.

NAME ADRRESS EMAIL ID OF THE CORRESPONDING AUTHOR:

Dr. Majeti Srinivasa Rao,
Associate Professor of Paediatrics,
Alluri Sitarama Raju Academy of Medical Sciences,
Eluru – 534005, W.G. District,
A.P. State, India.
Email – majetisrinivas@gmail.com

Date of Submission: 12/11/2013.
Date of Peer Review: 13/11/2013.
Date of Acceptance: 19/11/2013.
Date of Publishing: 26/11/2013