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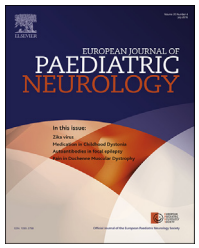

Original article

Benign paroxysmal migraine variants of infancy and childhood: Transitions and clinical features

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ABSTRACT

Introduction: Migraine variant disorders of childhood include benign paroxysmal torticollis of infancy (BPTI) and benign paroxysmal vertigo of childhood (BPVC). This study aimed to review our experience with BPTI and BPVC and determine the incidence of children transitioning between each of these disorders and to vestibular migraine (VM).
Methods: We retrospectively reviewed the medical records of patients seen at the Balance and Vestibular Program at Boston Children's Hospital between January 2012 and December 2016 who were diagnosed with BPTI, BPVC, and/or VM.
Results: Fourteen patients were diagnosed with BPTI, 39 with BPVC, and 100 with VM. Abnormal rotary chair testing was associated with progression from BPTI to BPVC ($n = 8$, $p = 0.045$). Eight (57.1%) patients with BPTI and 11 (28.2%) with BPVC had motor delay. Eleven (78.6%) patients with BPTI and 21 (53.8%) with BPVC had balance impairment. Six BPTI patients developed BPVC (42.9%), six BPVC patients developed VM (15.4%), and two patients progressed through all three disorders (2%). One BPTI patient progressed directly to VM.
Discussion: Most patients with BPTI will experience complete resolution in early childhood, but some will progress to BPVC, and similarly many patients with BPVC will progress to VM. Parents of children with these disorders should be made aware of this phenomenon, which we refer to as "the vestibular march." Children with BPTI and BPVC should also be screened for hearing loss, otitis media, and motor delay.

1. Introduction

Migraine disorders are collectively the most common cause of vertigo in children and adolescents, as reported in a number of epidemiologic studies.[1–6] Three migraine variant disorders that are frequently described in the pediatric population are benign paroxysmal torticollis of infancy (BPTI), benign paroxysmal vertigo of childhood (BPVC), and vestibular migraine (VM).

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BPTI is characterized by spontaneous, recurrent episodes of head tilting in infants and toddlers [Fig. 1].[7] The episodes typically last for a few days at a time and resolve altogether by approximately three years of age. Accompanying symptoms can include vomiting, irritability, vertigo, ataxia, and pallor. The diagnostic criteria for BPTI as outlined in the International Classification of Headache Disorders, 3rd edition (ICHD-3) are shown in Fig. 2.[8] Studies have found links between BPTI and migraine, including multiple reported cases of patients with BPTI that demonstrated mutations in the neuronal calcium channel gene CACNA1A, which has been associated with familial hemiplegic migraine and episodic ataxia.[9–11] Additional features of BPTI that suggest a migrainous origin include its periodicity, frequent association with other migrainous symptoms, and high incidence of family history of migraine.[7,12] A high incidence of gross and fine motor delay in children with BPTI has also been reported.[7]

BPVC is characterized by episodes of room spinning vertigo in young children that typically last for minutes at a time and resolve spontaneously without any post-ictal symptoms. It has been described as a migraine precursor or equivalent due to multiple features, including its periodicity, the presence of migrainous symptoms in some patients, the high incidence of adult migraine in patients with a history of BPVC, and a high rate of family history of migraine.[13–17] The diagnostic criteria for BPVC as outlined in the ICHD-3 are shown in Fig. 3.[8,15] The age of onset of BPVC ranges between two and 12 years old, with a mean age of onset of six years old.[18] A few studies have mentioned a relationship between BPVC and BPTI.[19,20]

VM was originally described by Lempert and Neuhauser in 2001.[21] Since that time it has become widely accepted as the most common cause of adult vertigo. The diagnostic criteria for VM as outlined in the ICHD-3 are shown in Fig. 4. Our experience with VM in pediatric patients has been previously reported.[22]

Progression from BPTI to BPVC[7,10,23] and from BPVC to migraine[14–17] has been reported in some studies, although such progression has not been further investigated or characterized. Additionally, reports on the clinical features of BPTI and BPVC are particularly limited in the medical literature, making it difficult to counsel families the on prognoses of these disorders. Here we discuss our experience with the largest reported series of patients with BPTI and with BPVC, respectively. We aim to further outline the clinical characteristics and natural history of these disorders, as well as evaluate the progression between BPTI, BPVC, and VM.

Fig. 1 – Photo of patient with benign paroxysmal torticollis of infancy during a torticollis episode.

2. Materials and methods

2.1. Patients

We retrospectively reviewed our internal database of all patients seen at the Balance and Vestibular Program at Boston Children's Hospital from January 2012 to December 2016 to identify all patients ≤21 years of age that were diagnosed with BPTI, BPVC, and/or VM. The electronic medical records of patients were reviewed to determine demographic features, clinical presentation, and progression of the disorders over time. This retrospective review was approved by the Institutional Review Board of Boston Children's Hospital.

Diagnosis of BPTI, BPVC and VM were confirmed to meet the respective diagnostic criteria for these disorders outlined in the ICHD-3 (Figs. 2–4). Patients with congenital torticollis, which is characterized by chronic non-periodic torticollis starting in infancy and gradually resolving spontaneously or with physical therapy, were not included.

2.2. Testing

All patients had undergone a complete otologic and neurologic examination by a Pediatric Otolaryngologist (JB). A majority of patients also underwent a variable combination of objective vestibular and balance tests. All testing was conducted in our clinical vestibular laboratory at the Balance and Vestibular Program at Boston Children's Hospital by a licensed audiologist (GWZ) and a trained assistant. Sinusoidal harmonic rotary chair testing was performed using Micromedical equipment (System 2000 and VisualEyes; Micromedical Technologies, Chatham, Illinois). Cervical vestibular evoked myogenic potential testing (cVEMP) was recorded with Biologic Navigator Pro Evoked Potential System (Natus Medical Inc, San Carlos, California). Additional vestibular testing was performed on older patients, but testing varied widely by age and clinical presentation. Only rotary chair and cVEMP results are reported in this study since these tests were completed in most cases. Rotary chair testing was performed using videonystagmography (VNG) goggles in older patients, while an observational camera technique was used in patients ≤ 3 years of age in the dark rotary chair enclosure with the child on a parent's lap.

When VNG goggles were used, rotary chair results were considered abnormal if gains for all frequencies tested were

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