

Vertigem posicional paroxística benigna na infância: relato de dois casos incomuns

Relato de Caso

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Benign positional paroxysmal vertigo in childhood: report of two unusual cases

Gustavo Polacow Korn¹, Roberta Ribeiro de Almeida², Roberto Augusto Carvalho Campos³, Maurício Malavasi Ganança⁴, Fernando Freitas Ganança⁵.

1) MSc candidate in Otorhinolaryngology by the Department of Otorhinolaryngology and Head and Neck Surgery, Universidade Federal de São Paulo/ Escola Paulista de Medicina.

2) Visiting Professor of the Department of Otorhinolaryngology and Head and Neck Surgery, Universidade Federal de São Paulo/ Escola Paulista de Medicina.

3) Head of Pediatric Neurotology unit of the Department of Otorhinolaryngology and Head and Neck Surgery, Universidade Federal de São Paulo/ Escola Paulista de Medicina.

4) Head Professor of the Department of Otorhinolaryngology and Head and Neck Surgery, Universidade Federal de São Paulo/ Escola Paulista de Medicina.

5) Affiliate Professor of the Neurotology division of the Department of Otorhinolaryngology and Head and Neck Surgery, Universidade Federal de São Paulo/ Escola Paulista de Medicina.

Instituição: Universidade Federal de São Paulo/ Escola Paulista de Medicina.

Endereço para correspondência: Av. Brigadeiro Faria Lima, 1811 Cj 907-908 – Jardim Paulistano – São Paulo.

01452-001 / Phone number: (11) 30317933/38121157

E-mail: gustavokorn@uol.com.br

RESUMO

A Vertigem Posicional Paroxística Benigna (VPPB), caracterizada por episódios breves de tontura rotatória desencadeada pela movimentação da cabeça, é muito comum no adulto, porém pouco freqüente na criança. Apresentamos dois casos: uma criança de sete anos, com quadro de VPPB bilateral, que, após manobra de reposicionamento de partículas e exercícios de reabilitação vestibular ficou livre de sintomas e outra de quatro anos com VPPB unilateral associada a otite média secretora, que apresentou melhora após várias manobras e foi orientada para tratamento cirúrgico (adenomigdalectomia e colocação de tubo de ventilação bilateralmente). A fisiopatologia é discutida ao longo do estudo.

Descritores: tontura, vertigem, infância.

ABSTRACT

Benign Postural Paroxysmal Vertigo (BPPV), characterized by rotational dizziness spells triggered by head movements, is a rather common condition in adulthood, but infrequent in children. We report two cases: a seven-years-old girl with the diagnosis of bilateral BPPV, who became symptom-free following canalith repositioning maneuver and vestibular rehabilitation exercises; and a four-years-old boy with unilateral BPPV associated with otitis media with effusion that became symptom-free after several canalith repositioning maneuvers, who finally was advised to undergo surgical treatment (adenotonsillectomy with ventilation tubes replacement) due to the persistent otitis media with effusion. The pathophysiology is discussed along the article.

Keywords: dizziness, vertigo, children.

INTRODUCTION

Benign positional paroxysmal vertigo (BPPV), the most common form of peripheral vertigo, consists of a series of brief vertigo episodes, nausea and/or positioning nystagmus following head rotation towards a determined position. Following the episodes, a vague fluctuation-like dizziness sensation may persist for hours or days. BPPV by its clinical and recurrent features may affect the patient's daily activities¹.

It may follow head trauma, labyrinthitis, vertebrobasilar insufficiency, post-otologic surgery, endolymphatic hydrops, vestibular neuritis and middle ear diseases, but is idiopathic in the majority of cases¹⁻².

Regarding its pathophysiology, there are two theories: cupulolithiasis (in which otoconia are attached to the cupula) and canalolithiasis (in which free-floating material is within the endolymph of the semicircular canal)³.

In the vast majority of BPPV cases a posterior variant canal is found, but anterior and lateral canals may also be involved^{1-2,4}. Besides its bilateral involvement, simultaneous different canal commitment is possible¹.

Positioning nystagmus features will identify the affected canal. Nystagmus and vertigo occur generally after a few seconds latency, are limited in their duration, and fatigue after repeated provocations⁵. In the absence of one of those features, a central positional vertigo must be excluded⁶. The diagnostic maneuvers most used are the Dix-Hallpike, Brandt-Daroff, and horizontal canal tests¹.

Two theories have been proposed for vertigo without nystagmus in BPPV patients: (1) the nystagmus may be fatigued following repeated testing or habituation through provocative head positioning before referral to the clinic care; (2) the neural signal is strong enough to provoke vertigo, but not to reach the necessary threshold to stimulate the vestibulo-ocular pathway⁷.

The treatment of BPPV is based, mainly, on therapeutic maneuvers. The most used are the Epley⁸, Semont⁹ maneuvers, and Brandt-Daroff habituation exercises¹. Those treatments are very effective, with a high symptoms resolution rate. The Epley maneuver (canalith repositioning) is 98% successful⁸. The Semont maneuver (liberatory maneuver) succeeds 84% of the time when following the first and 93% when following the second procedure⁹. Once these maneuvers are done, the patient is asked to restrain himself from lying down in the supine position and from stirring his neck for the next 48 hours and to stay with the assistance of a soft cervical collar. For the five following days, the patient should not lie down on the affected or treated side. As observed, its efficacy is not totally accepted². In some cases, more than one maneuver may be necessary¹⁰, and recurrence may happen¹⁰⁻¹¹. Furthermore, spontaneous resolutions have also been reported¹¹.

Known as the most frequent vestibulopathy in adulthood¹², it is scarcely found in children^{5,13}. Although there are some reports of vertigo in children, very few have pertinent BPPV description^{14,15}.

We herein report two cases of BPPV in children, enhancing their clinical features and treatment.

Case report

Case 1

C.A.S., a seven-year-old girl, was admitted to our department of otolaryngology with recurrent vertigo for 2 months. The vertigo lasted for a few seconds (always less than 30 seconds), followed by an instability period, which could last for hours. The episodes were triggered by bilateral head rotation and when she quickly assumed the supine position. The episodes were followed by pallor, perspiration, nausea, and vomiting, impairing her daily activities. She denied feeling aural fullness, hearing-loss, fluctuating hearing or headache.

Her mother's pregnancy was uneventful, as was her neonatal and childhood period until the beginning of those symptoms. When asked, her mother denied history of any physiological problems. No family history of headache or previous head trauma were reported.

In the physical examination, the otoscopy, test with the tuning fork of 512Hz, Romberg and Romberg-Barré tests, gait, cerebellar test and cranial pair evaluation were normal. The Unterberger-Fukuda test was positive with a shift to the left. Following the Dix-Hallpike maneuver, the patient experienced vertigo without nystagmus, with three seconds of latency, and ten seconds duration with the head pending to both sides, followed by perspiration and pallor. She mentioned that the vertigo increased progressively, reaching a peak, and then decreased. The symptom was prominent on the left side.

We performed Epley's repositioning maneuver to the left, prescribed a soft cervical collar for 48 hours, instructed her to avoid lying down on the left side in the next 5 days, and

scheduled her to return in one week.

Upon her return, she told us that she had followed the suggested recommendations properly, and had experienced a great clinical improvement. With the Dix-Hallpike maneuver the initial findings persisted, only with the head pending to the right. A new Epley's maneuver to the right was repeated. The same initial recommendations were advised and another return was scheduled.

On the following return, she was asymptomatic. With the provocational maneuver, less intense findings were seen than one week ago. A second Epley's maneuver to the right was repeated, followed by the same recommendations.

In the fourth week, she was asymptomatic. The Dix-Hallpike maneuver did not result in either nystagmus or vertigo. Eletronystagmography was normal. A minimum distress with the positioning provocative maneuver with the head pending to the left was seen. Audiometric evaluation was normal.

We proposed Brandt-Daroff exercises for two months. She remained asymptomatic after one-year follow-up.

Case 2

W.A.F., a four-year-old boy, was admitted to our department of otolaryngology for vertigo episodes lasting for hours with resolution following treatment for otitis media with effusion (OME) one year ago. In the first episode, he was hospitalized for three days with an unconfirmed diagnosis of cerebellitis. At that time, neurological evaluation, spinal fluid analysis and cranial computed tomography were normal. Fifteen days before his first visit to our clinic, the patient experienced a brief vertigo episode, initially when getting up from bed, which became persistent throughout the following days. The episodes were associated with nausea, vomiting and pallor. In addition, he complained of intense motion sickness for the past two weeks. He also experienced bilateral hearing-loss. There was no tinnitus, convulsive spells, and conscience loss or head trauma. Besides a neonatal history of jaundice requiring phototherapy for two days, there was no history of any physiological problems. A familiar history of headache was obtained.

In the physical examination, the otoscopy revealed bilateral eardrum retraction, with radiated vascularization increase. Anterior rhinoscopy showed pallor of the inferior turbinate and oropharyngoscopy disclosed a pharyngeal tonsillae grade III. The Romberg and Romberg-Barré tests, Unterberger-Fukuda test, gait, cerebellar tests, cranial pair evaluation, and Dix-Hallpike maneuver were normal. Right ear audiometric evaluation showed a conductive hearing loss of 15dB and a tympanometric curve type B.

With diagnosis of vestibular disorder associated to OME, we proposed pharmacological treatment with oral amoxicilin and prednisone. A cavum radiography was ordered and a return was scheduled in two weeks.

Upon the first return, the patient reported vertigo at the moment his head was moved to perform the radiological

study. Otoscopy revealed right OME. With the Dix-Hallpike maneuver, there was vertigo with nystagmus (upside and counterclockwise, duration of 5 seconds, and three seconds of latency), with the head pending to the right. With the diagnosis of BPPV we proceeded to the Epley's maneuver. The patient was asked to return in one week. Upon his first return we became aware that the patient had refused to go through the recommended maneuver, due to intense fright of the vertigo feeling. He was then oriented to make the Brandt-Daroff exercises with his mother's support at his residence (two daily series of ten maneuvers for each side). One week later (fourth visit), he returned with the same findings. The Semont maneuver was then applied. On the fifth visit, he still experienced positioning nystagmus, however with subjective decrease in the vertigo intensity. A new Semont maneuver was repeated. On the sixth visit, he reported a great vertigo improvement. Neither nystagmus nor vertigo was detected.

One month later he was asymptomatic. During the follow-up period, persistent bilateral OME was observed, more predominant in the right ear, in spite of conservative management. An adenotonsilectomy with bilateral tympanostomy tube insertion was then proposed.

DISCUSSION

Case 1

What amazed us most in this case was that the girl at her age was able to detail her complaints precisely, thus helping enormously with the diagnosis and treatment.

Vertigo of limited duration (few seconds), when the head moved to a pending position on both sides while in the horizontal decubitus, was very typical, and with the provocative maneuver she experienced characteristic vertigo (with latency, lasting seconds, crescendo-decrescendo), but in the absence of nystagmus

Bilateral BPPV was suspect in this case by vertigo features resulting from the positioning maneuver to both sides. After left side treatment, symptoms persisted on the right side including positive findings with the respective positioning maneuver. Once the right side was treated, she became asymptomatic. This behavior suggests bilateral BPPV even in the absence of nystagmus.

Bilateral BPPV is rare, and we could not find any report on children in the literature. It was evidenced symptoms' improvement using the therapeutic maneuvers in patients with BPPV without nystagmus⁷. The left side was firstly treated in order to give attention to the most symptomatic side¹.

The repositioning maneuver associated with the patient's adherence made her symptom-free and granted her a normal life.

As is well known, BPPV diagnosis is made just on clinical grounds. This means that patients should undergo functional evaluation in order to discard other vestibulopathies that may progress with secondary BPPV, as we did.

Case 2

In the beginning, recurrent vertigo spells while the otitis media was active, together with a family history of migraine led us to the diagnosis of vestibular syndrome associated to OME, probably benign paroxysmal vertigo of childhood (BPV).

It was thought in BPV, once it is a paroxysmal, recurrent, non-epileptic event of vertigo that occurs in neurologically intact children¹⁷. Association between migraine and BPV¹⁵ and a family history of migraine was been described¹⁵. The most common causes of vertigo in children with normal eardrums are BPV and migraine¹⁸.

An association between migraine and BPV was made, stating that it as a migraine-like event, a vasospasm related phenomenon with subsequent ischemic damage of the utricular macula, and leading to the development of BPPV in children¹³.

On the second visit, the patient returned with a characteristic BPPV history, evidenced by a positioning maneuver, and right OME. In this case, it is important to analyze the relationship between BPPV and OME, because perpetuation of OME can account for failure of BPPV improvement after several therapeutic maneuvers. Some studies showed vertigo relief after tympanostomy with tube insertion¹⁵⁻¹⁶. Therefore adenotonsilectomy with tympanostomy with tube insertion for OME control would eventually abolish vertigo recurrent spells since it may be the casual factor of the dislodged otoconia.

Vertigo may be secondary to OME¹⁴⁻¹⁶. Three theories were proposed to support the relation between OME and dizziness: change of the normal pressure of the middle ear¹⁴; toxins leading to a serous labyrinthitis¹⁵; and ion transfer from the middle ear to the inner through the round window leading to an alteration in the endolymph composition¹⁶.

Although dizziness in children is believed to be uncommon, it may probably be more frequent than expected. In a study with 55 children with dizziness, between 5 and 15 years old, BPPV was found in only two (3,6%)¹⁸. In another study with 34 subjects, aging between 4 and 18, BPPV was found in 59% of the cases, showing high prevalence of BPPV in childhood¹⁹.

BPPV is believed to be rare in children because objective vestibular testing is difficult or impossible to perform in this age group⁵. We disagree with this opinion, because positioning and repositioning maneuvers are safe and easily managed⁸, can be performed in the doctor's office, and can be repeated whenever necessary, even in children. In case 2, the boy that refused to perform the maneuver up to the third visit, due to fear; after proper direction at his home, he accepted and was cooperative in accomplishing the repositioning maneuver. Although the maneuver should be avoided in patients with severe cervical arthrosis, vertebrobasilar insufficiency, those are rare entities in childhood; when a child does not tolerate the maneuver, sedation may be considered.

There is a lack of experience of otolaryngologists and neurologists with respect to BPPV diagnosis and treatment¹². We believe that the lack of investigation of BPPV may also

account for the low prevalence of this entity in childhood.

As for its pathophysiology there are beliefs that release of otoconia attributed to a transitory ischemia of the macula may migrate to the semicircular canals¹³. BPPV is not common in children because the otoconia is tightly bound to the macular membrane and difficult to dislodge¹³. Cupula deposit is less frequent in children than in adults, suggesting an aging phenomenon in vestibular labyrinth⁵.

It is important to mention that, differently from adults, children, due to lack of habit in expressing themselves, can have trouble with defining their own symptoms, or even be incapable of it, making diagnostic elaboration more difficult.

BPPV is a clinical entity whose diagnosis is made on clinical grounds. It is rarely bilateral, and scarcely documented in childhood. The repositioning maneuvers associated with the patient's adherence to the general directions led to the resolution of the symptoms in both described cases. In cases where BPPV is associated to other entities, both should be managed simultaneously.

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