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Case Report

A case of apogeotropic horizontal canal benign paroxysmal positional vertigo (cupulolithiasis) due to head contusion in an adolescent

Abstract

A 17-year-old high school boy who complained of severe positional vertigo visited our hospital. He belonged to an American football team in high school, and his head often hit opponents during games. The vertigo was strongest when his left ear was down in the supine position. Physical examination and computed tomography revealed no abnormalities. However, positional testing revealed apogeotropic direction-changing horizontal nystagmus. Rightward nystagmus in the left-ear-down supine position was stronger than leftward nystagmus in the right-ear-down supine position. Additionally, in the supine position, a null point for horizontal nystagmus was identified, beyond which the nystagmus changed direction. This null point was evident when the head was turned 10 degrees to the side. The diagnosis was right lateral canal type of BPPV caused by cupulolithiasis. He was treated with the Gufoni maneuver and reported a decreased incidence of positional vertigo upon re-evaluation.

Introduction

Benign paroxysmal positional vertigo (BPPV) is the most common vestibular disorder. It is caused by dislodged otoconia floating in the semicircular canals (canalolithiasis) or, less frequently, by otoconia attached to the cupula (cupulolithiasis) [1,2]. Traumatic brain injury and head contusion have been reported as common causes of BPPV [3]. Other reported causes are a prolonged recumbent position and various disorders of the inner ear [4]. BPPV is also associated with high morbidity in the older adult population [4,5]. However, BPPV in children and adolescents has been studied less extensively than in the adult population.

Previous studies have shown that BPPV of the posterior canal (PC-BPPV) occurs four times more frequently than BPPV of the horizontal canal (HC-BPPV) because of anatomical and positional differences [6]. In 1985, however McClure [7], reported several cases of pure geotropic nystagmus (nystagmus beating toward the ground) during the Dix-Hallpike maneuver in patients with no evidence of central nervous system lesions, corresponding to canalolithiasis of the horizontal canal. In 1995, the apogeotropic type of BPPV was also reported, in which the nystagmus beats toward the ceiling during lateral head turning in the supine position, corresponding to cupulolithiasis of the horizontal canal [8]. Since then, the number of published cases of HC-BPPV has gradually increased.

We herein present an extremely rare case of apogeotropic HC-BPPV (cupulolithiasis) caused by head contusion in an adolescent in whom we successfully analyzed the pathological nystagmus during a vertigo attack.

Case Report

A 17-year-old high school boy presented with positional vertigo and nausea after he woke up. The vertigo was strongest when his left ear was down in the supine position. He belonged to an American football team in high school, and his head often hit opponents during the game. Physical examination and brain computed tomography revealed no evidence of neurologic disease or abnormal findings.

When we conducted positional testing, we observed apogeotropic direction-changing horizontal nystagmus with Frenzel goggles. Rightward nystagmus in the left-ear-down supine position was stronger than leftward nystagmus in the right-ear-down supine position. In the supine position, we identified a null point for lateral nystagmus, beyond which the nystagmus changed direction [9]. This null point was evident when the head was turned 10 degrees to the right side [9]. When we conducted a positional test, we detected no nystagmus. Therefore, the diagnosis was right lateral canal type of BPPV caused by cupulolithiasis. Due to the clarity of the direction and intensity of the nystagmus observed with Frenzel goggles, we were able to identify the affected side.

The patient was treated with the Gufoni maneuver for HC-BPPV cupulolithiasis [10]. Briefly, his head was quickly turned upward toward the ceiling after first being brought to the side-lying position on the affected side. This position was maintained for 5 minutes. We then found that his leftward nystagmus in the right-ear-down supine position disappeared during positional testing with Frenzel goggles; however, his rightward nystagmus in the left-ear-down supine position was still observed, although the rightward nystagmus was slightly attenuated compared with the initial observation. After another 10 minutes, we again observed the presence or absence of the nystagmus during positional testing with Frenzel goggles; however, the nystagmus was weak. The patient also reported that his vertigo became weak. Thus, he returned home.

Discussion

Onset of BPPV is rare in young people [11–14]. A previous study also showed that BPPV, head trauma, and other inner ear diseases were less common causes of vertigo in young people [15]. In contrast, head injuries and head contusion are known causes of BPPV in adults [13]. It is believed that in adults, especially older adults, otoconia are easily detached from the otolith organ due to head contusion, and the otoconia enter the semicircular canal, resulting in the development of BPPV triggered by head position conversion and postural change.

Many cases of BPPV are believed to arise from the posterior semicircular canal, which is the most gravity-dependent canal [16]; however, the proportion of patients with HC-BPPV may be underestimated because involvement at this site is more likely to remit spontaneously than is involvement in the posterior semicircular canal [7]. However, since canalolithiasis and cupulolithiasis associated with HC-BPPV were reported with no evidence of central nervous system lesions [7,8], the number of reported cases of HC-BPPV has gradually increased.

Based on previous reports, a possible explanation of the BPPV in the present case is as follows. The patient was performing American football in high school and frequently sustained strong head contusions. Otoconia then detached from the otolith organ, and HC-BPPV developed after postural transformation. This suggests that young patients can develop BPPV due to head injuries and head contusion.

In the case of HC-BPPV, we have to consider the possibilities of both canalolithiasis and cupulolithiasis. Furthermore, the apogeotropic form of HC-BPPV can be even a canalolithiasis with the debris in the anterior part of the canal [16]. In our case, when we conducted positional testing, we observed apogeotropic direction-changing horizontal nystagmus. In addition, we identified a null point for lateral nystagmus, beyond which the nystagmus changed direction in the supine position [9]. Therefore, we were able to diagnose cupulolithiasis.

A recent study showed that the cupulolith repositioning maneuver, called the Gufoni maneuver, is an effective method for treating horizontal canal cupulolithiasis [10]. Kim et al. [10] showed that the Gufoni maneuver was effective in the treatment of HC-BPPV cupulolithiasis with a success rate of

73.1%. Although attenuation of nystagmus was moderate, we thought that the Gufoni maneuver was effective even in our patient. Further studies are required to assess the effect of the Gufoni maneuver in the treatment of HC-BPPV cupulolithiasis.

Conclusion

We analyzed HC-BPPV (cupulolithiasis) in an adolescent patient. We conclude that children can be diagnosed with a combination of careful history-taking and accurate examination of pathological nystagmus.

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