



Paroxysmal vertigo with nystagmus in children



Tomoko Okumura^a, Takao Imai^{a,*}, Kayoko Higashi-Shingai^b, Yumi Ohta^a,
Tetsuo Morihana^a, Takashi Sato^a, Suzuyo Okazaki^a, Yoriko Iwamoto^a, Yukiko Hanada^a,
Yoshiyuki Ozono^a, Ryusuke Imai^a, Kazuya Ohata^a, Hidenori Inohara^a

^a Department of Otorhinolaryngology – Head and Neck Surgery, Osaka University Graduate School of Medicine, 2-2 Yamadaoka, Suita, Osaka 565-0871, Japan

^b Department of Otolaryngology, Suita Municipal Hospital, 2-13-20, Katayama-cho, Suita, Osaka 564-0082, Japan

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ABSTRACT

Introduction: A pathological nystagmus is an objective sign that a patient feels vertigo. However, there have been few opportunities to observe and record pathological nystagmus during a paroxysmal vertigo attack. Furthermore, it can be difficult to obtain cooperation in pediatric patients. We present two cases of paroxysmal vertigo in children in whom we successfully recorded and analyzed their pathological nystagmus during a vertigo attack.

Methods: Of a total sample of 4349 patients seen at our hospital for dizziness in the last decade, a retrospective analysis revealed that 68 were children (<15 years old; 1.6%). Of these 68 children, we successfully identified pathological nystagmus during paroxysmal vertigo in only two (2.9%).

Results: Case 1 was a 4-year-old girl. She felt vertigo the strongest when her left ear was down in the supine position. We observed and recorded her nystagmus during a vertigo attack with her mother's permission. Her positional nystagmus in the supine position was horizontal persistent apogeotropic nystagmus. Rightward nystagmus in the left-ear-down supine position was stronger than leftward nystagmus in the right-ear-down supine position. Therefore, the diagnosis was right lateral canal type of benign paroxysmal positional vertigo, of which the pathophysiology was cupulolithiasis. The other patient was an 11-year-old boy. He had a family history of migraines. His vertigo attacks occurred after onset of a severe migraine and lasted between 2 and 48 h. During an attack that we observed, he showed nystagmus, which was direction-fixed right torsional and rightward in darkness. His mother had noticed that his eyes moved abnormally and that his left eye did not shift to the left side when he looked leftward. He was old enough to clearly express his own symptoms. Other neurological examinations were normal. The diagnosis was vestibular migraine.

Conclusions: We analyzed a pathological nystagmus during paroxysmal vertigo in two children. We conclude that children can be diagnosed with a combination of careful history taking and accurate examinations of a pathological nystagmus.

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1. Introduction

Observation of a pathological nystagmus is an objective sign that patients feel vertigo. A pathological nystagmus is not always easily observed, even in adult patients with paroxysmal vertigo [1]. Recording and observing pediatric paroxysmal nystagmus presents further difficulties in terms of obtaining cooperation of the child. In examinations of adult patients, they are usually instructed to bring

their head to the position that they feel vertigo. However, the fear of vertigo makes it difficult for children to comply with these commands; they are reticent to do something that will make them feel vertigo. Even if pathological nystagmus can be induced in children, they commonly refuse to keep their eyes open during paroxysmal vertigo. As a result, pathological nystagmus cannot be observed during paroxysmal vertigo. Therefore, various examinations should be combined. Furthermore, because children do not directly consult physicians when they feel vertigo, they do not visit physicians unless their family considers the vertigo to be serious enough. For these reasons, pediatric nystagmus during paroxysmal vertigo has

* Corresponding author.

E-mail address: timai@ent.med.osaka-u.ac.jp (T. Imai).

not been well studied compared with those reported in adults [2]. Here, we analyzed nystagmus during paroxysmal vertigo in two children and diagnosed them with a combination of careful history taking and accurate recording of nystagmus.

2. Material and methods

We retrospectively reviewed case notes for 4349 patients with vertigo or dizziness who presented to the Department of Otorhinolaryngology-Head and Neck Surgery, Osaka University Hospital from 2001 to 2014. Of these, 68 were aged younger than 15 years (1.6%), and there were 9 (0.2%) younger than 5 years old. Of these 68 patients, only two had their pathological nystagmus recorded during paroxysmal vertigo. Case 1 was a 4-year-old girl and case 2 was an 11-year-old boy. This study was approved by the Ethics Committee of Osaka University Hospital (No. 10091) and registered at the University Hospital Medical Network (UMIN) (UMIN000020047). The study was performed in accordance with the second Declaration of Helsinki. Written informed consent was obtained from the patients' parents before examinations.

The patients' nystagmus in the left eye during paroxysmal vertigo were recorded with an infrared charge coupled device camera (720 × 480 dots, 30 Hz) (RealEyes; Micromedical Technologies, IL, USA). The nystagmus for each patient was three-dimensionally explored by axis-angle representations [3,4]. An eye position can be reached by rotating the eye from the reference position around a single axis. The eye position was represented by a vector around the axis, the length of which was proportional to the angle of rotation. The reference position was defined as the eye position when the subject was looking straight ahead with the head in an upright position. The term 'straight ahead' was defined as looking at a target that was located horizontally in front of the eye [5]. The method of analysis of the eye rotation vector and its accuracy have been described previously [6,7]. Videotape images were converted into digital images (720 × 480 pixels) and the space coordinates at the center of the pupil and an iris freckle were reconstructed in three dimensions. The space coordinates were defined as follows: the X axis was parallel to the naso-occipital axis (position forward), the Y axis was parallel to the inter-aural axis (positive left), and the Z axis was normal to the X–Y plane (positive upwards). The X, Y, and Z components mainly reflect roll, pitch, and yaw components, respectively. The direction of rotation was described from the subject's point of view. The axis angle α describing rotation of θ around the axis \mathbf{n} is provided by the following formula: $\mathbf{a} = \theta\mathbf{n}$, where \mathbf{n} is the unit vector of which the direction represents its axis [3,4].

3. Results

3.1. Case 1

A 4-year-old girl visited our hospital with her mother complaining of positional vertigo. She described her symptoms as vertigo that occurred when rolling over in bed and when climbing onto a chair. Her mother observed her abnormal eye movements at home. The patient explained that she felt vertigo strongest when her left ear was down in the supine position. There was no history of headache, nausea, or vomiting. Pure tone audiometry demonstrated mild, bilateral, low-tone, sensorineural hearing loss (Fig. 1A). There was no prior history of inner ear disease or neurological disease. A neurological examination showed positional nystagmus, but was otherwise unremarkable. Head computed tomography and brain magnetic resonance imaging were both unremarkable.

When she turned her head leftward in the supine position,

positional nystagmus with a rightward Z component (horizontal component) was observed (Fig. 1B). Her nystagmus was reversed when she turned her head rightward (Fig. 1C). Her positional nystagmus in the supine position was horizontal, persistent, apogeotropic nystagmus, and the rightward nystagmus in the left-ear-down in supine position was stronger than the leftward nystagmus in the right-ear-down supine position. Therefore, we diagnosed her with the right lateral canal type of benign paroxysmal positional vertigo (BPPV), of which the pathophysiology was cupulolithiasis [8].

As she explained that she felt strong vertigo when her head was turned to the left in the supine position, and her mother said that her eye oscillated horizontally in the supine position, we were able to estimate her provocative head position before the examination took place. We observed paroxysmal nystagmus efficiently with minimal maneuvers as simply as possible. During recording, we encouraged her to keep her eyes open. The patient's positional nystagmus disappeared at a follow-up visit 1 week later.

3.2. Case 2

An 11-year-old boy visited our hospital with his mother complaining of vertigo attacks and migraines, which lasted from 2 to 48 h. He had a history of episodic migraine headaches from the age of 4 years that were unilateral to the left hemisphere and throbbing in nature. His headaches started from the left temple and spread to the left half of his head. There was no history of aura. He was admitted to the pediatric unit of a peripheral hospital with his first episode of vertigo with nausea, vomiting, photophobia, and phonophobia in conjunction with migraine symptoms. There was no history of tinnitus or ear fullness. He had a history of motion sickness. He also had a family history of migraines with visual aura in his sister, mother, and grandmother.

He had the second vertigo attack when he was hospitalized at a peripheral hospital. During his second vertigo attack, which lasted 48 h, his mother noticed abnormal eye movement. His left eye failed to turn to the left when looking to the left side. She took a photograph of his eyes (Fig. 2A). Head computed tomography and brain magnetic resonance imaging were performed, but these were both unremarkable. The migraine associated with the second vertigo attack lasted for 7 days, and he reported that it was the worst migraine he had ever experienced.

The patient was referred to our department 7 days from the onset of the second vertigo attack because his doctor asked for further neurological examinations by an otolaryngologist. Pure tone audiometry demonstrated normal hearing ability (Fig. 2B). He had a third vertigo attack in the waiting room of our department and we examined him immediately. His direction-fixed nystagmus was successfully recorded with a CCD camera. In the light, he showed weak rightward horizontal nystagmus, and in the dark, he showed a clear right torsional (X component) and rightward (Z component) nystagmus (Fig. 2C). The Y component (vertical component) was indistinct. The nystagmus increased in the dark. Pure tone audiometry (Fig. 2A) and somatosensory testing were unremarkable.

In the 2-year follow-up period, he had two further attacks of vertigo with migraines lasting for 24 h. The abduction deficit of his left eye when looking to the left side was not observed any more during these attacks. He has had a total of five vertigo attacks. His vertigo attacks were triggered by anxiety. The patient met all of the diagnostic criteria of vestibular migraine (VM) in the *International Classification of Headache Disorders*, third edition beta version (ICHD-3 β) (Table 1) [9]. Therefore, we diagnosed him with VM.

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