## Benign Paroxysmal Vertigo in Childhood: A Long-term Follow-up

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Benign paroxysmal vertigo in children is characterized by sudden attacks of vertigo lasting seconds or minutes. During the attack, the child has nystagmus and is unable to stand without support. Initially, the attacks are frequent, later slowly disappearing. Nineteen children who were diagnosed in 1975-1981 participated in a follow-up study. Sixteen of them were examined with audiometry and electronystagmography. Age at onset was from 5 months to 8 years, and the symptoms disappeared after 3 months to 8 years. The follow-up was performed 13 to 20 years after diagnosis. Twenty-one percent developed migraine which is somewhat more than in a normal population of this age. Thirty-nine percent had a family history of migraine which is a figure considerably lower than in a migraine population. None still had vertigo or a balance disorder. Our conclusion is that benign paroxysmal vertigo has a favorable outcome, and it is not a general precursor of migraine.

Key words: benign paroxysmal vertigo, migraine, positioning nystagmus, child, follow-up

Abbreviations: BPV benign paroxysmal vertigo, BPT benign paroxysmal torticollis

(Headache 1999;39:33-37)

In 1964, Basser¹ described a distinct clinical entity, designating it benign paroxysmal vertigo (BPV) of childhood. The cardinal symptom is attacks of vertigo of sudden onset without warning or provocation lasting from seconds to a few minutes. Additional symptoms are pallor, sweating, vomiting, and often nystagmus. The child is mostly unable to move, sit, or stand without support. Consciousness is not impaired. The child recovers completely after an attack. The age of onset is within the first 4 years of life, but it may occur later. The sex distribution is equal. The vertigo attacks tend to present more frequently during the first months after onset, from several times a day to some times a week. The attacks then gradually become more widely separated and finally cease

spontaneously after months, but often after a couple of years.

Over the years, some reports have appeared in literature giving a better understanding of the disorder. Benign paroxysmal vertigo has been described thoroughly by Koenigsberger et al,<sup>2</sup> Dunn and Snyder,<sup>3</sup> Koehler,<sup>4</sup> Mira et al,<sup>5</sup> Lanzi et al,<sup>6</sup> and Finkelhor and Harker.<sup>7</sup> A possible relation to migraine has been mentioned in several reports, initially by Fenichel.<sup>8</sup> However, the relationship of BPV to migraine, as well as the long-term prognosis, need to be clarified.

We reported a group of 15 children in 1982.<sup>10</sup> These children and a few others have now been investigated over a long time period. The aims of the present paper are to present a long-term follow-up, and especially to discuss the relationship of BPV to migraine and other forms of headache and to present changes in vestibular function over time.

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Accepted for publication July 29, 1998.

### **SUBJECTS AND METHODS**

The study included 19 consecutive children, 12 girls and 7 boys, who were examined in the departments of pediatrics and otolaryngology in the years 1975-1981. They were referred for fluctuating vertigo and fulfilled

Basser's description of BPV. All had normal mental and motor development.

A follow-up evaluation was performed in 1994 in the same departments. The age of the subjects at follow-up was from 18 to 24 years (mean 19.8, median 19.5 years). The history was carefully obtained (from one or both parents), especially focusing on the family history, history of migraine or other headache problems, and vertigo. Physical and neurological examinations were performed, and electroencephalography was carried out.

Using a standard technique, pure tone audiometry was performed using the ascending method according to ISO 6189. This test mainly reflects the peripheral function of the auditory system.

Electronystagmography to detect the presence of spontaneous, positional, or gaze nystagmus was performed. Binaural caloric tests were also performed using water at 30°C and 44°C. The result was considered significantly abnormal if there was an asymmetry of more than 20% in the caloric test, or spontaneous nystagmus with a slow phase velocity of 2° per second or more, or positional nystagmus with slow phase velocity of such magnitude in at least one lateral position. During electronystagmography, head shake tests were also performed.

#### RESULTS

Sixteen of the 19 subjects in the follow-up study went through audiometry, electronystagmography, and clinical examination. Three of them could not participate for practical reasons, but they were interviewed by telephone. At the time of follow-up, all participants were healthy except for what is mentioned later, and there was no alcohol or drug abuse.

The age at onset of BPV varied from 5 months to 8 years (mean 3.4, median 3 years) (Table 1). The time of follow-up was from 13 to 20 years (mean 15.7, median 15 years). The symptoms appeared during time periods from 3 months up to 8 years (mean 2, median 1 year). The age at remission was between 2 and 16 years (mean 5.5, median 4.3 years).

From the family histories, it was found that in one family there were cases of BPV and also of migraine. In six other families, there was a history of migraine in either parents or siblings. Migraine was defined according to the IHS criteria.<sup>11</sup> One child was adopted and nothing was

known about the family history. Of the 18 subjects with an available history, 7 (39%) (95% confidence interval: 17 to 64), had a family history of migraine and 1 a family history of BPV.

At the time of follow-up, 4 (21%) of the 19 subjects (95% confidence interval: 6 to 46), 25% of the women and 14% of the men, had migraine according to the above-mentioned criteria. One man (No. 4) had migraine with aura irregularly; two women (Nos. 3 and 12) reported migraine without aura with a frequency of two attacks per year for the previous several years. One woman (No. 9) had more frequent attacks of migraine with aura and also had migraine without aura, and was on propranolol as a prophylactic medication. Two of the women with migraine (Nos. 3 and 9) also had attacks of vertigo which sometimes developed into migraine. Five of the 19 individuals had episodic tension-type headache. It occurred more often on stressful occasions, but in no case was it very severe or chronic.

One woman (No. 12) had had recurrent attacks of torticollis since the age of 6 months, diagnosed as benign paroxysmal torticollis (BPT). In the beginning of the disease, the attacks appeared every month or every second month, but the frequency gradually diminished. From the age of 3 1/2 years, she had attacks of vertigo combined with photophobia. At around the age of 10, attacks of quite severe headache with photophobia began. She has to lie down and usually sleeps. The headache lasts around 12 hours, and the attacks occur about twice a year. There is no family history of migraine.

Two subjects (Nos. 14 and 16) had abnormal hearing; No. 14 had profound congenital hearing loss and No. 16 had a bilateral hearing loss—in the left ear 18 dB, in the right ear 23 dB. All others had normal pure tone audiometry results.

#### **COMMENTS**

Benign paroxysmal vertigo in childhood is a relatively rare and rather unknown disorder. However, due to its typical symptomatology it is easily recognized when considered. The two important differential diagnoses which must be ruled out are posterior fossa tumor and epilepsy. Physical and neurological examination, EEG, audiometry, ENG, and caloric tests are normal.

This follow-up study was done to investigate the out-

34 January, 1999

Table 1.—Clinical Characteristics of Children With Benign Paroxysmal Vertigo (BPV)

Subject	Sex	Age at Onset, y	Age at Recovery, y	Age at Follow-up, y	Family History	Migraine	Other Headache	Vertigo
1	F	0.4	2	16	0	0	+	0
2	M	1.1	2.1	16	0	0	0	0
3	F	2	3.5	16	BPV/migraine	+	+	+
4	M	2	2.5	18	0	+	0	0
5	F	2.2	3.2	22	Adopted	0	0	0
6	F	2.5	5.5	18	Ō	0	0	0
7	F	2.8	3.8	23	0	0	+	0
8	F	3	4	17	0	0	0	0
9	F	3	6	21	Migraine	+	+	+
10	F	3	3.5	16	0	0	0	0
11	M	3	4	18	Migraine	0	+	0
12	F	3.5	4.5	20	0	+	0	0
13	F	4	6	20	0	0	0	0
14	F	4	12	19	0	0	0	0
15	M	4	4.3	18	Migraine	0	0	0
16	M	4	6	19	Migraine	0	0	0
17	M	5	5.5	21	Migraine	0	0	0
18	F	8	9	24	Migraine	0	0	0
19	M	8	16	22	0	0	0	0

come later in life for these children. In the first long-term follow-up by Lanzi et al,<sup>9</sup> it was found that the majority of cases (6 of 7) evolved into disorders related to migraine. Also in earlier studies,<sup>4-6,8</sup> the relationship to migraine or BPV as a migraine equivalent or migraine precursor has been pointed out.

In the present study, the proportion of girls who developed migraine was 25% and that of boys, 14%. The prevalence of migraine found in a study of an American population was roughly 10% for 20-year-olds; 15% for women and 5% for men. 12 Thus, there was an increased prevalence of migraine in our BPV population, though the figures were not statistically significantly raised. Two with a family history and two without developed migraine. However, since migraine is associated with age, with a peak at 35 to 40 years, it is not yet known how this BPV population will develop as adults. Five (26%) of the adolescents had tension-type headache of moderate character. This is equivalent to figures from a normal population. 13

In large studies of migraine, the frequency of a positive family history is 70% to 80%.<sup>14</sup> In the present study, 39% of the participants with BPV had a family history of migraine—a figure which is different from migraine pop-

ulations. A meta-analysis was done on six published BPV studies<sup>2-4,6-8</sup> in which the prevalence of family history of migraine was stated. Our earlier study<sup>10</sup> was excluded because several of those patients are included in the present study. Therefore, 86 children were included and 31 of them had a family history of migraine—a prevalence of 36% (95% confidence interval: 26 to 47). It can be concluded that patients with BPV have a frequency of family history of migraine that is clearly lower than in patients with migraine. Regarding the family history, it is tempting to suggest that BPV consists of two entities. One type would then be a migraine equivalent with a family history of migraine and the other a more pure form without any relation to migraine.

A relationship between BPV and BPT has been mentioned<sup>3,10,15</sup>; both conditions have been presumed to be precursors of migraine. One woman in this study had BPT as a very young child, then developed BPV and now has migraine. This is the first case presented with this triad as far as we know.

As a whole, the group of subjects had normal audiometry indicating that no cochlear sequelae were elicited by the BPV. The caloric results were normal with few excep-

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Table 2.—Results of Auditory and Vestibulo-oculomotor Testing 13 to 20 Years After Benign Paroxysmal Vertigo Diagnosis

Subject	Age, y	Sex	PTA*	Calorics, % LP	Lateral Position	Lying Down, Head Turned	Sponta- neous Nystagmus	Head Shake	Pursuit
1	17	F	Normal		+	+	Normal	Normal	Normal
2	17	M	Normal	Normal	+	+	Normal	+	Normal
3	17	F	Normal	Normal	+	Normal	Normal	Normal	Normal
4	19	M	Normal	Normal	+	+	Normal	Normal	Normal
5	23	F	_	_	_	_	_	_	_
6	19	F	_	_	_	_	_	-	_
7	24	F	Normal	Normal	Normal	+	Normal	Normal	Normal
8	16	F	Normal	Normal	+	+	+	+	Normal
9	22	F	Normal	Normal	+	+	Normal	+	Normal
10	17	F	Normal	Normal	+	+	+	+	Normal
11	19	M	Normal	Normal	+	+	Normal	+	Normal
12	21	F	Normal	Normal	+	+	Normal	Normal	Normal
13	21	F	Normal	Normal	+	+	+	+	+
14	20	F	102/97	Normal	+	+	+	+	Normal
15	19	M	Normal	Normal	+	+	Normal	Normal	+
1 <b>6</b>	20	M	23/18	23	Normal	Normal	Normal	Normal	Normal
17	22	M	-	_	<u> </u>	_	_	_	_
18	25	F	Normal	Normal	Normal	Normal	Normal	Normal	Normal
19	23	M	Normal	32	+	_	_		-

<sup>+</sup> Findings of unimportant pathology.

Eye movements were recorded as spontaneous nystagmus and positional nystagmus in lateral positions and lying down with head turned. Additionally, nystagmus after head shake and ocular smooth pursuit were recorded.

tions, also indicating that the peripheral function of the inner ear was not disturbed by the disease. Some unimportant pathology of the vestibulo-oculomotor system was found (Table 2).

In conclusion, BPV has a favorable outcome. It did not turn out to be a general precursor of migraine or any other type of headache. Migraine can cause BPV, but it is not the only cause.

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<sup>\*</sup> PTA indicates pure tone audiometry.

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