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A Study of the Etiology and Prognosis of Breath-Holding Spells*

Stati Leona, MD. / Kalbiye Yalaz, MD.*****

Breath-holding attack's are a clinical entity which is frequently observed in early childhood. In response to stressful situations, the child begins to cry, then usually during the second or third expirium holds his breath, suddenly, becomes cyanotic, and loses consciousness. Tonic-clonic convulsive movements may follow the relaxation of the extremities. The attack may terminate at any phase and the child may go to sleep.

The earliest written description of this ailment was perheps given by Nicholas Culpeper (1916-1954) in the seventeenth Century.[1] Neumann, also tried to distinguish in 1905 breath-holding spells from tetanoid Laryngospasm.[2]

The pathophysiology of breath-holding spells is still uncertain. Epilepsy[3, 4, 5, 6], anemia[7, 8, 9, 10, 11], hypoxia[12], psychogenic factors such as a history of migrain headaches in the parents[13] have been proposed to account for the occurrence of this disorder.

The present study is restricted to an outline of the etiology, therapeutic needs and prognosis of the 55 children who were considered to having the criteria of breath-holding spells (BHS) which are mentioned above.

Subjects and Methods

The subjects for this prospective survey were derived from the Out-patient-Deparment of Hacettepe Childrens Hospital. 28 healthy children were used for the control group.

* From the Deparment of Pediatric Neurology, Hacettepe University, Faculty of Medicine, Ankara, Turkey.
** Resident in Pediatrics.
*** Associate Professor of Pediatrics and Pediatric Neurologist.

The age among the 55 breath-holders ranged from 3 months to 5 years and was fairly well matched with the control group.

All cases selected had a clear-out history of recurrent episodes, triggered by frustration, minor injury or fright. The spells were clearly observed during the examination of six patients (10.9 %). The above precipitating factors of the spells and histories of epilepsy, migraine and motor development were studied. Mental and motor development were recorded and physical and neurological examinations were carried out.

Laboratory Studies: X-rays of the skull and of the wrist, telecardiograms, tests measuring serum calcium, phosphorus, alkaline phosphatase, hemoglobin, blood smear for erythrocyte morphology) and electroencephalograms were performed.

The Chi Square test was used for the statistical analyses.

Each child was seen at least twice during a three month interval to follow the result of therapy.

Results

Among the 55 breath-holders, 34 were boys (61.8 %), and 21 were girls (28.2 %). Their age ranged between 3 months and 5 years with a mean-age of 2.8 years. The age distribution of the patients is shown in Figure 1. BHS stardes as early as the neonatal period in 6 patients. The latest starting age was 29 months.

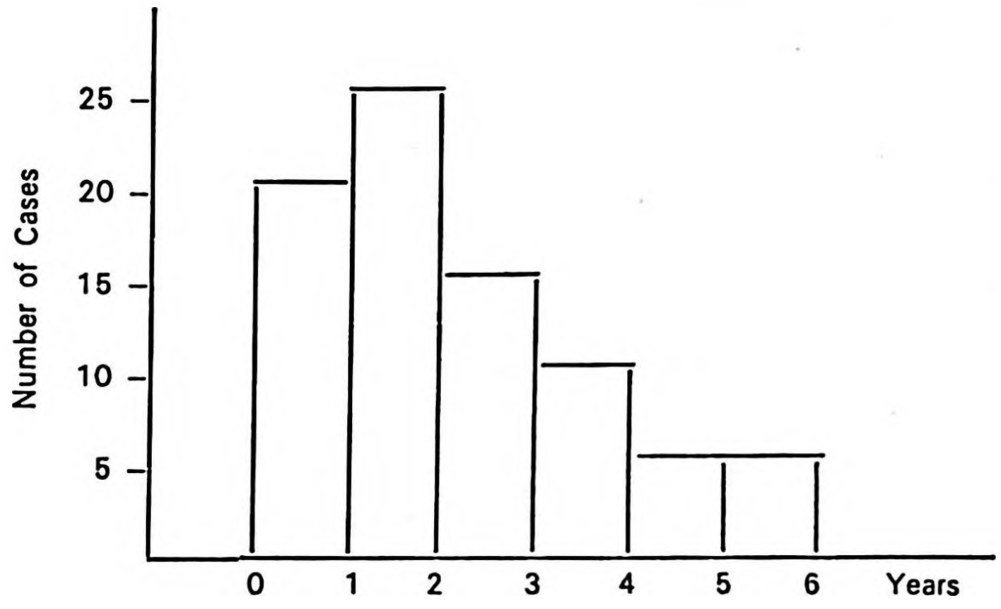

Figure 1
Age distribution of patients with Breath-Holding Spells

Crying was present previous to the attacks of apnea and loss of consciousness in all cases.

The cases could be divided into two early recognizable groups on the bases of the severity of spells. In the first group, apneic spells and cyanosis following a short period of crying terminated with a brief period of loss of consciousness. Following this, the chlidren returned to normal states of health. This group consisted of 49 children (89.1%) in whom the evolution of the attacks fit the described pattern.

In the second group, consisting of six children (10.9 %), the prolonged and more severe attacks ended with tonic-clonic generalized convulsions.

The frequency of the spells varied from one child to another and even for the same individual case. It is rather difficult to estimate the exact duration of the spells, but it can be said thay they lasted no more than one minute.

Precipitating factors for each child can be listed according to the order of their frequency as follows: Pain such as blow to the head, injection, falls etc.; 11 cases, anger; 7 cases, withdrawal of a toy or of an object; 7, frustration; 2, fear.

Family histories of 38 children (26 %) revealed five (9.4 %) with mothers and three with fathers positive for BHS. In one case both parents had histories of BHS, one child had an epileptic aunt and another one had two cousins with BHS. 11 children (20 %) had parents with a history of migraine headaches.

Prenatal and birth histories were normal in all but three children; physical and neurological examination were normal except in two children with microcephaly. Four children demonstrated high blood pressure during attack, following the attacks it returned to normal.

In our study, of the two cases with microcephaly, one had severe mental-motor retardation (1.8 %),

Hemoglobin levels of the chlidren were noteworthy: Below 8 gr % in seven (13.1 %) patients, between 8 and 9.9 gr % in eight (14.4 %), a total of 15 cases (27.9 %) with below 10 gr of Hb/100 ml. Erythrocyte morphology in these 15 cases was suggestive of iron-deficiency anemia (Table I).

TABLE I

RATIO OF IRON-DEFIENCY IN PATIENTS WITH BREATH-HOLDING SPELLS

Hb. Level (g/100 ml)	Laxdal and Gomez (1969) 120 BHS Patients %	Hacettepe Study 230 Control Patients %	55 BHS Patients %	28 Control Patients %
8.0	2.5	0.9	13.1	3.7
8.0–9.9	10.8	12.6	14.1	25
10	86.7	86.7	72.8	71.3
Total	100	100	100	100

Rickets was observed in nine out of 25 cases (36 %) whose wrist X-rays were taken. Serum calcium levels were normal except in three cases. The phosphorus level was found to be normal in 39 cases. Alkaline phoshatase showed normal levels in 28 cases (75.6 %); it was found to be elevated in eight (22.2 %).

The EEG studies of 48 of the 55 cases are summarized in Table II. 41 cases (85.5 %) were recorded as normal. Paroxyomal activity was observed in seven (14.5 %)

TABLE II

COMPARISION OF THE EEG ABNORMALITIES OF CHILDREN WITH BREATH-HOLDING SPELLS WITH THOSE OF NORMAL CHILDREN

		Paroximal Activity		EEG Abnormality Diffuse and Focal Abnormality Non-paroxismal))			
	Total No. of Cases	No. of Cases	%	No. of Cases	%	No. of Cases	Total %
BHS Patients	48	6	12.5	1	2	7	13.5
Control	28	2	7	2	7	4	14

Five cases with pulmonary infections, two with heart murmur, one with pyodermy and three with whooping cough were recorded as incidental findings.

Prognosis and Therapy: 42 patients were followed up at regular intervals and BHS disappeared completely in 22 of them. 15 cases were treated for iron-deficiency anemia and six cases responded promptly to therapy. 28 cases with a normal Hb level and erythrocyt morphology were treated with anticonvulsives. (Phenobarbital 4 mg/kg/day to 10 mg/kg/day.) BHS disappeared in 14 cases (50 %) after a short period of time. A combination of phenobarbital and diphenylhydantoin (5 mg/kg/day) was given to six cases which did not respond to the earlier therapy.

Discussion

The pathophysiology of BHS is uncertain.

According to Livingston, the attacks usually start during the first year of age and were very unusual before the age of 6 months.[4] On the other hand, 19 children (34.5 %), in our study had their first attack before the age of 5 months and BHS starded as early as in the neonatal period (10.9 %). Laxdal[14] indicates a percentage of 5 % for the same age group.

In Lombrossa's studies, the familial incidence for BHS among the patient group was twice as high as the control group. Laxdal's findings of the familial incidence for BHS was 30 % and Levy's 3.8 %. We found familial incidence to be 26 % in the BHS and 5 % in the control group. ($X^2 = 11$ $p < .001$) this difference is statistically significant.

According to the studies made by Levy et al.,[13] migraine headaches of the mother has been considered to be related to BHS. A history of migrain headaches of the mother was obtained in 20 % the children with BHS and 8 % in the control grouh. The difference between these two groups was statistically significant ($X^2 = 7$ $p < .01$).

Correlation between anemia and the BHS was extensively studied. The incidence of iron deficiency anemia was found to be 23.5 % in Holowach,[9] 10.6 % in Maulsy-Kelloway,[10] 66.7 % in Paul's[2] studies. The Hb level was below 10 gm in 28 % of our cases with BHS. But the same figure was 28.7 % for the control group and the difference was insignificant (p. 70). Probably anemia is not responsible for the BHS but inceases the severety of the attacks.

Rickets was diagnosed through the X-ray of the wrist in 9 of 25 children (36 %). The serum Calcium levels were normal in all cases but their BHS ceased after the therapy with vitamin D and Calcium

preparations. Similar X-ray findings were observed (20.8 %) among the control groups. The difference between the patients and the control group was statistically insignificant ($X^2 = 0.03$ $p < 90$).

It is important to differentiate BHS from epilepsy. The EEG recordings were extensively studied. In BHS there were only two abnormal occurrences of EEG among the group of 198 patients in the Livinston study.

In the present study, EEG was obtained in 48 children with BHS and paroxysmal activity in 6 (12.5 %) and latent libateral epileptiform activity in 1 (2 %) cases were recorded.There was no statistically significant difference between the patients and the control group ($p < 90$).

Prognosis of the BHS was favorable. Only one patient died from food aspiration during the spell, which was demonstrated by a post-mortem examination and recorded by Paulson.[15]

32 patients attended the clinic regularly. The spells had ceased or disappeared completely in 27. But the duration of the follow-up was not long enough to draw a definite conclusion for the prognosis.

We may conclude from this study that there is more then one factor responsible for BHS and probably these children had abnormal cerebral mechanism at birth.

BHS has to be treated according to their etiology because each attack produces anoxia of the brain. Iron-preparation for anemia and anticonvulsive medication are the drugs of choice.

Summary

55 patients with BHS were studied for their family history, past history. Laboratory studies were done for iron. FFe definiency anemia, rickets, epilepsy, The results were compared with 28 children whom we considered normal. The therapeutic approach was also summarized.

REFERENCES

1. Gauk, E. W., Kidd, 1., and Prichard, J. S.: Mechanism of seizures associated with breath-holding spells, N. Eng. J. Med. 268: 1436, 1963.
2. Hinman, A., and Dickey, L. B.: Breath-holding spells; A review of literature and 11 additional cases, A.M.A.J. Dis. Child. 91: 23, 1956.
3. Gastaut, H. and Fisher-Williams, N.: Electroencephalographic study of syncope; its differetiation from epilepsy, Lancet 2: 1018, 1957.
4. Livingston, S.: Convulsive disorders in infant and children (breath-holding spells), Advances Pediat. 10: 119, 1958.

5. Low, N. L., Gibbs, E. I. and Gibbs, F. A.: Electroencephalographic findings in breath-holding spells, Pediatrics 15: 595, 1955.
6. Strauss, H.: Electroencephalographic observations in the mechanism of breath-holding spells, Electroencephalography, Montreal 9: 164, 1957.
7. Holowach, J. and Thurston, D. L.: Breath-holding spells and anemia, N. Eng. J. Med. 268: 21, 1963.
8. Chanda, R. K.: Iron deficiency anemia associated with Breath-holding spells and obesity. Lancet 1: 1106, 1964.
9. Chanda, R. K.: Association of b eath-holding attacks with anemia and their treatment, Indian J. Pediat. 31: 143, 1964.
10. Maulsby, R., Kellowey, P.: Transient hypoxic episodes in children in Kellowey, P., and Peterson, (eds) Neurological and Electroencephalographic Correlative Studies in Infancy, New York, Crune and Stratton, Inc., 1964, p. 349.
11. Paul, R., Manchanda, S. S.: Breath-holding attacks the role of Phychological factors and iron therapy, Indian J. Pediat. 36: 271, 1969.
12. Gauk, E., Kidd, L., Prichard, J. S.: Aglottic breath-holding spells, N. Eng. J. Med. 275: 1361, 1966.
13. Levy, D. M. and Patrick, H. T.: Relation of infantile convulsions, head-banging and breath-holding to fainting and headaches (migraine) in parents, Arc. Neurol. Psychiat. 19: 865, 1928.
14. Laxdal, T., Gomez, R. M., Reiher, J.: Cyanotic and pallid syncopal attacks in children (breath-holding spells), Develop. Med. Child. Neurol. 11: 755, 1969.
15. Paulson, G.: Breath-holding spells: a fatal case, Develop. Med. Child. Neurol. 5: 246, 1963.