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A double center study

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**Brief Communication** 





# Childhood primary stabbing headache: A double center study

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#### **Abstract**

**Background:** Primary stabbing headache (PSH) is an idiopathic headache disorder characterized by head pain occurring as a transient and localized single stab or a series of stabs. The present study aimed to examine the characteristics of childhood PSH and whether they fit the International Classification of Headache Disorders, 3rd edition (ICHD-3) criteria. We also investigated the association with migraine and episodic syndromes.

**Methods:** In this retrospective study, we included 60 patients seen at two headache clinics (Rome and Bari) between 2016 and 2022. A headache-focused history was obtained. All patients had normal neurological examination. PSH was defined according to ICHD-3 criteria.

Results: Twenty-three patients were male (38%) and median (range) age at disease onset was 8 (3–17) years. Stabs recurred with irregular frequency and their duration varied from a few seconds up to 30 minutes. Stabs were located in different head regions. Twenty-five patients (42%) underwent neuroimaging exams. Five children reported a limitation of daily activities and none had a chronic pattern. Forty-seven patients (78%) reported a family history of primary headache, especially migraine, and forty-three had episodic syndromes (i.e. infantile colic, benign paroxysmal vertigo, motion sickness, recurrent abdominal pain, cyclic vomiting). Twenty patients had an associated primary headache: 16 suffered from migraine and four suffered from tension type-headache. According to ICHD-3 criteria, thirty-one patients had a diagnosis of probable PSH as a result of a duration of stabs longer than a few seconds (>3 seconds).

Conclusions: Features of childhood PSH can vary widely. As seen in previous studies, several patients reported a stab duration longer than a few seconds and this might suggest that current ICHD-3 criteria may need adjustments to be suitable for children. High frequency of associated migraine and episodic syndromes could suggest a common pathophysiological mechanism between PSH and migraine. We can hypothesize that PSH and migraine attacks may be part of a spectrum of the same disease, although further evidence is needed. Larger studies with long-term follow-up are needed to improve understanding of this condition.

# **Keywords**

Children, migraine, primary headache, primary stabbing headache

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# Introduction

Primary headaches are common in childhood and adolescence, with migraine and tension-type headache being the most commonly reported (1). Other primary headaches, such as primary stabbing headache (PSH), appear to be rare and often remain under-recognized. According to criteria of the most recent International Classification of Headache Disorders (ICHD-3) (2),

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PSH is characterized by: (A) head pain occurring spontaneously as a single stab or series of stabs; (B) each stab lasts for up to a few seconds; (C) stabs recur with irregular frequency, from one to many per day; (D) no cranial autonomic symptoms; and (E) not better accounted for by another ICHD-3 diagnosis. If two out of three criteria among B, C or D are satisfied, the diagnosis will be probable PSH. This is very helpful considering that headache could last more than 2 minutes and up to 10-15 minutes in PSH (3-5). There are limited data available about pediatric PSH and its prevalence is not precisely known, ranging from 3.35 to 5.1% (6–7). However, a recent study reported a higher prevalence of 9.97% (8), suggesting that PSH is not uncommon in children and adolescents. There is no clear sex predominance and mean age of disease onset is 4.5–9 years (3,6,9). The pathophysiological mechanism is considered to be an excessive stimulation of neurons associated with central pain mechanisms (3,9). PSH can occur in association with other headache types, most commonly migraine, and this feature is stronger in adults than in children (4). Nonetheless, two pediatric PSH studies reported a high frequency (47 and 39%) of episodic syndromes typically related to migraine, such as infantile colic and cyclic vomiting (3,6). This finding could suggest a common pathophysiological mechanism between migraine and PSH. The present study aimed to examine clinical characteristics of childhood PSH and whether they fit the ICHD-3 criteria. We also investigated the association with migraine and episodic syndromes.

# Table 1. Clinical characteristics of included patients with PSH.

# **Methods**

A retrospective observational study was conducted in two Italian headache centers (Bambino Gesù Children's Hospital of Rome and San Paolo Hospital of Bari) from 2016 to 2022. All patients were evaluated by one of the investigators with a special interest in childhood headaches. We included typically developing patients under 18 years of age with a follow-up of minimum 6 months. ICHD-3 criteria were used and we included patients with PSH and probable PSH. Secondary causes were excluded based on detailed clinical evaluation, headache history, neurological examination and brain imaging. Demographics, clinical data and information regarding stabs (i.e. duration, frequency, intensity and location) were collected. Duration of stabs was categorized as stabs lasting up to a few seconds (3-120 seconds) and more than 2 minutes. Associated primary headaches and episodic syndromes were also investigated. Continuous and categorical variables were reported as median (range) and proportion (percentage), respectively. Consent for the use of medical data for research purposes was obtained. The study was approved by the Institutional Review Board of the two institutions.

#### Results

Sixty patients were included (Table 1). Twenty-three were male (38%) and median (range) age at disease onset was 8 (3–17) years. Median (range) follow-up was 12 (8–60) months. Twenty-five patients (42%) underwent neuroimaging exams; specifically, nine

	PSH (n = 60)
Median age at disease onset, years (range)	8 (3–17)
Female, n (%)	37 (62%)
Median follow-up, months (range)	12 (8–60)
Stab duration <3 seconds, n (%)	29 (48%)
Stab duration >3 seconds and <120 seconds, n (%)	21 (35%)
Stab duration > 120 seconds, n (%)	10 (17%)
Stab location, (n)	Parietal (5), frontal (20), occipital (7), temporal (10), fronto- temporal (8), fronto-parietal (2), diffuse (5), vertex (3)
Pain intensity	Mild 19
	Moderate 28
	Severe 13
Family history of primary headaches, n (%)	47 (78%)
Associated migraine, n (%)	16 (27%)
Associated TTH, n (%)	4 (7%)
Episodic syndromes, (n)	Infantile colic (13), benign paroxysmal vertigo (7), motion sickness (25), recurrent abdominal pain (19), cyclic vomiting syndrome (1)
Associated symptoms, n (%)	17 (28%)

PSH, primary stabbing headache; TTH, tension type headache; associated symptoms: phonophobia, photophobia, nausea, vomiting and cranial autonomic symptoms.

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patients underwent brain computed tomography and 16 patients underwent brain magnetic resonance imaging and the results were all normal. Stabs recurred with irregular frequency and duration varied from a few seconds up to 30 minutes. Therefore, according to ICHD-3 criteria, thirty-one patients received a diagnosis of probable PSH as a result of a duration of stabs longer than a few seconds (>3 seconds). Twenty-one out of 31 patients reported stabs lasting up to 120 seconds and ten patients between 120 seconds and 30 minutes. Only one patient had a pain duration up to 30 minutes. Stabs were located in different head regions, including parietal (n = 5), frontal (n = 20), occipital (n = 7), temporal (n = 10), fronto-temporal (n = 8), fronto-parietal (n = 2), diffuse (n = 5) and vertex (n = 3). Therefore, stabs did not occur in a specific location and this finding is in line with previous studies (5,9). According to ICHD-3 criteria, intensity of headache was defined as mild, moderate or severe. Nineteen patients reported mild pain, 28 moderate and 13 reported severe pain. Despite 41 patients (68%) having a moderate-severe pain, only five reported a limitation in activities of daily living. They received prophylactic therapy with palmitoylethanolamide or tryptophan with benefit. None had a chronic daily pattern (i.e. disease duration over 3 months with almost daily stabs occurrence) (10,11). Response to analgesic therapy was not investigated. Triggers for PSH are not common, but some potential triggers have been described (5,9,12). In our cohort, frequency of stabs was worsened by stress in five patients.

Forty-seven children (78%) had a family history of primary headache, especially migraine (40 patients out of 47), whereas no family history of PSH was reported. In our cohort, 20 patients (33%) had an associated primary headache: 16 suffered from migraine and four suffered from tension type-headache. Associated symptoms as phonophobia, photophobia, nausea, vomiting and cranial autonomic symptoms (i.e. conjunctival injection and/or lacrimation, nasal congestion and/or rhinorrhea, eyelid oedema, forehead and facial sweating, forehead and facial flushing, sensation of fullness in the ear, miosis and/or ptosis) were investigated. Forty-three children (72%) had no associated symptoms. Two patients described autonomic symptoms (one had conjunctival injection and the other had forehead and facial flushing) in a small percentage of their attacks. They did not have a strictly unilateral pain and they did not report a sense of restlessness or agitation. These two patients received a diagnosis of probable PSH. Nausea was present in five patients, phonophobia in 10 and photophobia in seven. None reported vomiting. As regards episodic syndromes, 13 children had infantile colic, seven reported benign paroxysmal vertigo, 25 had motion sickness and 19 had recurrent abdominal pain. Only one patient had

a history of cyclic vomiting syndrome. Seventeen patients did not have any episodic syndrome.

# **Discussion**

Primary stabbing headache is an under-recognized primary headache disorder and only a few studies have investigated stabbing headache in children (3–7). In our cohort, median (range) age of disease onset was 8 (3–17) years and this finding was in line with previous studies (3,7), although a great variability is reported and also younger children can develop PSH (10). We found a clear female predominance, as observed in adults (11.13.14). Accompanying symptoms were reported by 17 patients (28%) and this association was previously described by Soriani et al. (6). Two patients had an autonomic symptom and were classified as probable PSH. Furthermore, 31 out of 60 patients reported stabs lasting more than a few seconds, leading to a diagnosis of probable PSH. This finding has already been reported by previous studies (3,5,7) and might suggest that current ICHD criteria should be further modified to be suitable for children. In particular, the criterion of stab duration is too restrictive and could lead to diagnostic difficulties (15). We confirmed that a family history of primary headaches is common in pediatric PSH, with migraine being the most common type (3,9). Unlike prior studies, a high percentage of our patients (33%) had an associated primary headache; in particular, 16 reported migraine, as observed in adults (10). It is also important to emphasize that migraine frequency was higher than generally reported in this age group (1,16). Moreover, 43 patients (72%) had almost one episodic syndrome (13 infantile colic, seven benign paroxysmal vertigo, 25 motion sickness, 19 recurrent abdominal pain and one cyclic vomiting). There is compelling evidence that infantile colic, benign paroxysmal vertigo, motion sickness, recurrent abdominal pain and cyclic vomiting syndrome are migraine equivalents within the migraine syndrome (17). Accordingly, the high frequency of associated migraine and episodic syndromes could suggest a link between PSH and migraine. Central sensitization secondary to peripheral mechanisms could have a role in PSH, as in migraine, and this is also supported by a previously described correlation between PSH and migraine (9-10). In pediatric age, a higher PSH prevalence in very young children (7) may also suggest that PSH represents an early phenotypic variant of migraine. In our cohort, clinical pattern of stabs was monophasic or intermittent (11) and prophylactic treatment was not required because of an irrelevant impact on daily activities. Finally, although this was not our primary field of investigation, we consider that routine brain imaging may not be needed for children with PSH and whose neurological exams are normal. A better understanding 4 Cephalalgia

of this condition will help clinicians to assess whether neuroimaging is needed or not. We are aware that all these features are based on a limited number of studies and will require further confirmation.

# **Conclusions**

To the best of our knowledge, this is the largest study including children with PSH with a follow-up of minimum six months. Childhood PSH has a heterogeneous presentation with stabs varying in duration, location and intensity. This feature may require a greater flexibility of ICHD criteria, in particular in relation to duration criterion. We consider that patients with stabs

lasting more than a few seconds should not receive a diagnosis of probable PSH. Targeted studies are needed to evaluate a revision of PSH criteria to better reflect current clinical characteristics. In our cohort, PSH was frequently associated with migraine and episodic syndromes and this might suggest a common pathophysiological mechanism. We can hypothesize that PSH and migraine attacks may be part of a spectrum of the same disease, although further evidence is needed. The present study has some limitations, such as the small number of patients and the retrospective design. Larger studies with long-term follow-up are needed to improve what we know about PSH in pediatric age.

# Clinical implications

- Childhood primary stabbing headache has a heterogeneous presentation
- Current ICHD-3 criteria may need further adjustments to be suitable for childhood PSH
- Primary stabbing headache and migraine attacks may be part of a spectrum of the same disease

#### **Author contributions**

GM, LP, FU, GS, ST, MCP, DDA and VS acquired the clinical data. GM and MV drafted the manuscript. GM, MV and VS critically revised the manuscript. All authors contributed to the article and approved the final version of the manuscript submitted for publication.

# Data availability

De-identified datasheets are available for other researchers upon reasonable request to the corresponding author.

# **Declaration of conflicting interests**

The authors declare that there are no potential conflicts of interest with respect to the research, authorship and/or publication of this article.

#### Ethical statement

Consent for the use of medical data for research purposes was obtained. The study was approved by the Institutional Review Board of the two institutions.

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