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The prevalence of allodynia, osmophobia and red ear syndrome in the juvenile headache: preliminary data

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Abstract The aim of the study was to determine the frequency of clinical allodynia, osmophobia and red ear syndrome in a young population. Medical records of the children admitted for headache between 1 December 2004 and 31 March 2005 were consecutively studied. A questionnaire was used to find the prevalence of allodynia, osmophobia and red ear syndrome. We visited 96 children with headache. The range of age was 6–18 years. We classified migraine in 57%, other primary headaches in 25% and secondary headaches in about 18%. The presence of ipsilateral clinical allodynia was 14.5% in migraine, osmophobia in 20% of migraine and red ear syndrome in about 24% of migraine cases and they were absent in the other two headache groups. Our study shows that features like osmophobia, allodynia and red ear syndrome are not uncommon in migraine while they are absent in other types of headache.

Key words Allodynia • Osmophobia • Red ear • Migraine • Children

Introduction

Migraine is a complex disease with many possible clinical features associated with painful episodes and some of these characteristics are useful to establish well defined diagnostic criteria for migraine by the new IHS classifications [1]. Recently the attention of several researchers has been focusing on some less frequent features of migraine attacks such as allodynia, osmophobia and red ear syndrome, that can help to underline some pathophysiological mechanisms of migraine and to define the diagnosis and prognosis of migraine [2–6].

Cutaneous allodynia is a pain or discomfort resulting from a non-noxious stimulus to normal skin, due to mechanisms of central sensitisation [2] and pathophysiological and therapeutic implications have been suggested in migraine [3, 4]. It has been reported in 53.3% of an adult migraine population [2].

Osmophobia is intolerance to smells and its addition to criteria D in the appendix of the new IHS classification has been proposed [1]. Recently Kelman [5] found about 24.7% of adult migraineurs with osmophobia and has suggested that osmophobia is very specific, but very insensitive in diagnosing migraine.

Red ear syndrome [7] is characterised by uni/bilateral painful attacks to the ears, which become red and burning. Lance [7] suggested the presence of local axon reflexes to explain the two described idiopathic cases. Later, we described eight idiopathic red ear syndrome cases in close relation with migraine and suggested an association between the two conditions [6]. Other sporadic cases

[8, 9] have been described. In our migrainous patients red ear syndrome showed unilateral, bilateral or alternating localisation, in isolation or associated with migraine (before, during and after).

However, there are no data about the prevalence of these three clinical features in the paediatric population affected by headache; nevertheless there are interesting physiopathological and clinical implications for the study of headaches.

The aim of our study was to determine the frequency of clinical allodynia, osmophobia and red ear syndrome in a young population visited in our Center for Diagnosis and Treatment of Headache in Youth.

Methods

Medical records of the children admitted for headache between 1 December 2004 and 31 March 2005 were consecutively studied. The children, affected by headache, were classified according to the 2004 IHS criteria [1]. A semi-structured interview was always administered by the same two doctors. General and neurological examinations were carried out for all children. Other examinations (blood tests, neuro-physiological and neuro-radiological examinations and other specialist controls) were carried out when necessary. Nine items of a questionnaire aimed at finding out the prevalence and some characteristics of allodynia, osmophobia and red ear syndrome (presence and localisation of allodynia, discomfort when combing, brushing, touching; presence of osmophobia, sensitivity to perfume, food smells; presence of red ear syndrome, localisation, duration and relation to migraine attacks).

Results

We visited 96 children with migraine in our Headache Center during the study period. The range of age was 6–18 years and the average age was 10.7 years. There were 49 males and 47 females. There were 48 children below 11 years and 48 children over 11 years. According to IHS criteria we classified migraine in 57%, the other primary headaches (primary stabbing headache, episodic and chronic tension-type headache) in 25% and secondary headaches in about 18%. Ipsilateral clinical allodynia was present in 9 children with migraine (14.5%), osmophobia in 23 children with migraine (20%) and red ear syndrome in 16 migraine cases (24%). These three features did not occur in the other two headache groups. There was a slight prevalence of allodynia, osmophobia and red ear syndrome in the males.

Osmophobia, clinical allodynia and red ear syndrome did not appear to be related to age. Osmophobia and clinical allodynia seemed connected to the severe intensity of pain (100%) and the presence of an important neuro-vegetative symptomatology (at least three symptoms of criteria D: vomiting/nausea/phono/photophobia) (respectively 85% and 60%), while red ear syndrome appeared to be less associated to severe pain (62.5%) and to neuro-vegetative symptomatology (50%).

Red ear syndrome was bilateral in 80% and unilateral in 20%; the duration was about 30–60 min and in 10% of the subjects red ears preceded the onset of a painful attack.

Discussion

Our preliminary study shows that features like osmophobia, allodynia and red ear syndrome are not uncommon in the juvenile headache. These features, however, seem to be highly specific to migraine because they are absent in other types of headaches, but they are not sensitive for the diagnosis of migraine. These results are similar to those reported in the adult headache population for allodynia and osmophobia [2, 5]. Yet, the prevalence of allodynia and osmophobia appears lower than that reported in adult migraine and possible explanations are the longer duration of illness in the adult population as well as the major difficulty in asking children about these clinical features.

The absence of a link between the prevalence of these features and age (before or after 11 years) in our population can suggest that their presence in migraine does not derive only from factors of brain maturation. Allodynia and osmophobia were often present together and they were associated to the severe intensity of pain (100% cases), and to an important neuro-vegetative syndrome (when at least three symptoms of criteria D were present) (respectively 60% and 85%). Thus, allodynia obviously seems due to a central sensitisation mechanism made easier by the frequency and intensity of pain [3].

The mechanism of osmophobia in migraine is not well known, however we know that trigeminal activation can control the olfactory mediated sensation [10] and this interaction may be at the nasal level or at the thalamus or cortex level [10]. There are contradictory results that show the presence of sensorial hyperacuity to odours in migraineurs [11], but also that migraineurs can be more anosmic or hyposmic than normal subjects [12]. The less common prevalence of osmophobia in migraine, compared to phono- or photophobia, can suggest that only severe painful attacks can activate the olfactory system, while the activation threshold is lower for the visual and auditory systems.

Considering also the different responses to triptans in migraine, with or without allodynia [4], we may suppose

that allodynia and osmophobia help distinguish two migraineur subgroups with different disabilities, different therapeutic responses and perhaps different follow-ups; therefore, we think that it is useful to ask patients with migraine about these two features. Allodynia and osmophobia could be predictive factors for a negative evolution of migraine.

Red ear syndrome was present also in "moderate migraine" (not severe pain and no important associated vegetative symptoms), not necessarily associated to other specific features of migraine or to allodynia and osmophobia. Lance [7] suggested a sympathetic hypofunction in the secondary red ear and an antidromic axonal reflex with the release of vasodilator peptide syndrome in idiopathic cases. Trigemino-vascular activation has been called to explain red ear syndrome associated to migraine [6, 8]. The innervation of facial skin includes vasoconstrictor (under prevalent control of the sympathetic system) and vasodilator fibres (under control of the parasympathetic system) [7, 13, 14]. In migraine attacks, we often observed a facial pallor probably due to sympathetic hyperactivation or parasympathetic hypofunction. When red ears appear during migraine episodes, we can also observe the contemporary presence of facial vasoconstriction and ear vasodilatation, as if they were activated at the same time by separate functional neuronal modules. Also, the observation of red ear syndrome beginning before the migraine pain, like an "aura equivalent", induces us to think of red ear syndrome as an isolated neuronal system which can be activated during the migraine. The presence of red ear syndrome before or during migraine attacks (or like equivalent) can support the recent "Modular Theory" of migraine [15]. To explain atypical migraine that showed atypical symptoms or the coexistence of features of migraine and other features of another primary headache, Young et al. [15] propose "that groups of neurons, called modules, become activated to produce each symptom of a primary headache disorder and that each module is linked to other modules that together produce an individual's headache". Recently, Silberstein [16] appealed to this theory to explain the symptoms. In our opinion, red ear syndrome seems to be a good example to support this theory because in the same patient it can be associated to or dissociated from the migraine attack; it can be observed either before or during the migraine attacks and can be present together with the activation of the opposite functional system. The clinical meaning of red ear syndrome is not known in migraine and other studies are needed.

In conclusion, allodynia, osmophobia and red ear syndrome are not infrequent in migraine, also in affected children, and can probably help to distinguish different migraineur subgroups.

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