Vincenzo Raieli Gian Luca Eliseo Girolama La Franca Michela La Vecchia Domenico Puma Mario Eliseo

Cerebral polyopia in migraine: a clinical case

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V. Raieli (☒) • G.L. Eliseo • G. La Franca M. La Vecchia • D. Puma • M. Eliseo Division of Child Neuropsychiatry, G. Ingrassia Hospital, AUSL 6, Via Gaetano La Loggia 3, I-90129 Palermo, Italy E-mail: uslpanpi@tin.it

Tel.: +39-0917-033283 Fax: +39-0917-033253 Abstract We describe an 8-year-old, with a 1-year history of migraine without aura who experienced an episode of unusual visual aura, characterised by a fourfold view of her mother's image. These images were clearly visible only on their right half, and lasted about 30 min, followed by her typical migraine. A peripheral mechanism seems unlikely in this case, so we propose a cerebral mechanism, possibly by means of

spreading depression involving the occipital lobe with secondary spread to neighbouring areas.

Key words Polyopia • Aura • Migraine • Spreading depression

Introduction

A great variety of sensory phenomena have been reported as manifestations of migraine aura, mainly including visual disturbances caused by the coexistence of positive and negative visual symptoms [1]. The most common visual phenomena are photopsias, scotomata and fortification spectra [1, 2]. In contrast, optic illusions in the number of the same images (polyopia) have been rarely described during an episode of migraine with aura [3]. Herein, we report the case of a patient who experienced the appearance of polyopia associated with a negative visual disturbance at the beginning of a migraine attack.

Case report

An 8-year-old girl came to our observation in April 1997 because of a 1-year history of migraine without aura. The headache was bilateral, bitemporal, throbbing, of moderate-severe intensity, associated with nausea and photo-phonophobia, and lasted from 30 min to 3 h. It occurred 2–3 times

per month on average. Moreover, the child reported that about 3 months previously, she experienced an unusual migraine attack which was preceded by a visual aura of gradual onset. The aura was described as a multiplication of the right half of the objects and people in her visual field. The patient said that she also saw her "mother more times, four times, and that she was missing half of her body". The residual left half of the images was not well visible.

Figure 1 reports the drawing made by the child to describe the episode. The disturbance lasted 15-30 min and disappeared gradually. The patient was scared and needed her mother's reassurance. While aura was present, she experienced the gradual onset of headache with features similar to the previous episodes of migraine without aura.

For this episode, the patient was admitted to another paediatric hospital for testing; basal head computed tomography (CT) and detailed ophthalmological examinations were normal. The patient was discharged without therapy and only the hemivisual defect and migraine headache were pointed out.

When we observed the patient, we advised admission for diagnostic study for the girl and her 11-year-old brother who complained of many episodes of migraine without and with

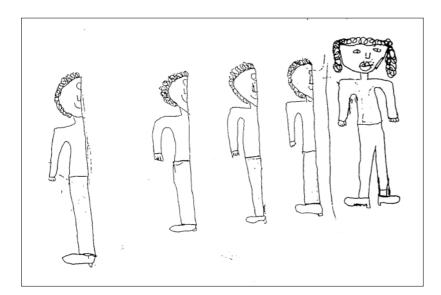


Fig. 1 Drawings made by the child to describe her visual aura

typical visual aura. We found a strong familiarity for migraine with and without aura. During hospitalization, the general medical and neurological examinations were completely normal.

Sleeping and waking electroencephalography (EEG), visual evoked potential (VEP)-pattern reversal, Doppler, neuropsychological tests (WISC-R), psychodiagnostic tests (Children Apperception Test (CAT) and family drawing), psychological examination, and ophthalmologic examination were performed to exclude eye diseases (e.g. Charles Bonnet syndrome), epilepsy, psychiatric disorders such as psychoneurosis and psychosis, and neuropsychological disorders of the visual perception, due to selective troubles or cognitive impairment. All were normal.

The child has been discharged from our division with the diagnosis of "migraine without aura, a single episode of aura migraine". She has been followed in our centre for one year without any new occurrences of visual episodes.

Discussion

Polyopia is an uncommon visual phenomenon, defined as multiple copies of a percept or visual preservation in space, which can arise for peripheral or central nervous system disturbances [3, 4]. Cerebral polyopia is an extremely rare symptom of central nervous system disease. It can be due to a positive phenomenon representing hyperactivity of neuronal cells induced by seizure activity [3] or more often a negative manifestation of cerebral damage associated with a visual field defect [3, 4]. Often, polyopia has been associated with palinopsia [3, 5] – perseveration or the recurrent appearance of a visual image after the stimulus has disappeared – and for both a similar pathophysiology has been

hypothesised [3–5]. Areas involved in cerebral polyopia are the occipital lobe and neighbouring area [5]. Bender [6] postulated a defect of visual integration and fixation due to occipital lobe disease, and Gottlieb [4] suggested an "incomplete visual processing due to poor visuospatial localisation in a hemianopic field". However, the mechanism of cerebral polyopia remains unknown.

In our case, the ophthalmologic examination and VEP excluded peripheral causes while the normal results of the psychological examination and psychodiagnostic and neuropsychological tests excluded psychiatric disorders. The absence of epileptiform activity, the length of the phenomena, the transient dysfunction, the normal neurological examination and CT scan, and the following onset of migraine-like headache excluded epilepsy and stroke as possible causes.

The personal history of migraine without aura, the family history of migraine, the development of aura and the kind of subsequent headache are all features which support the interpretation of this phenomenon as an unusual manifestation of migraine aura.

Polyopia is rarely reported as a manifestation of migraine aura [3]. It has not been found in two large population-based surveys [2, 7].

In our patient, the polyopia was associated with a hemivisual defect of the multiple images. To the best of our knowledge this phenomenon has not been previously described.

A possible pathophysiological mechanism might be found in the concept of "spreading depression" [8], the cortical phenomenon more pointed out to explain the migraine aura [9]. We infer that a spreading depression, starting from the occipital lobe, causes a release of neighbouring visual association areas from visual inhibitory inputs [5], followed by the appearance of polyopia like that observed rarely in patients with occipital ischaemic lesions and hemianoptic

visual defect or with visual pathway lesions and eye diseases [4, 5].

However, it remains unclear why only the right half of the multiple images was visible. In Gottlieb's opinion [4], polyopia can result from incomplete visual processing due to poor visuospatial localisation in the hemianoptic visual field related to the peripheral retina that is responsible for localisation of visual stimuli. In contrast, identification of objects more linked to the fovea – usually less involved in occipital lesions by reason of macular sparing – is normal in these patients. In our case, we believe that spreading depres-

sion in the left occipital lobe did not spare the macula and, as a consequence, identification resulted hemidefective. This hemidefective perception should undergo a wrong spatial localisation (polyopia), owing to the changed activity of the neighbouring cortical areas released from inhibitory inputs of the primary visual cortex, by means of the spreading depression.

However, these are all hypotheses and the described episode remains of difficult comprehension. In spite of the incomplete understanding of the described phenomenon, it is evident how fascinating the study of migraine aura can be.

References

- Nappi G, Manzoni GC (1996)
 Migraine. In: Nappi G, Manzoni GC, (eds) Primary headaches. Smith-Gordon, London, pp 34–45
- Russell MB, Olesen J (1996) A nosographic analysis of the migraine aura in a general population. Brain 119:355–361
- Bender MB, Rudolph SH, Stacy CB (1986) The neurology of the visual and oculomotor systems. In: Baker AB, Joynt RJ (eds) Clinical neurology. Harper-Row, Philadelphia, pp 43–45
- Gottlieb D (1992) The unidirectionality of cerebral polyopia. J Clin Neuroophthalmol 12:257–262
- Vaphiades MS, Celesia GG, Brigell MG (1996) Positive spontaneous visual phenomena limited to the hemianopic field in lesion of central visual pathways. Neurology 47:408–417
- Bender MB (1945) Polyopia and monocular diplopia of cerebral origin. Arch Neurol 14:323–338
- Queiroz LP, Rapoport AM, Randall EW, Sheftell FD, Siegel SE Raskin SM (1997) Characteristics of migraine visual aura. Headache 3:137–141
- Leao AAP, Morrison RSJ (1945)
 Propagation of spreading cortical depression. J Neurophysiol 8:33–45
- Lauritzen M (1994) Pathophysiology of the migraine aura. The spreading depression theory. Brain 117:199–210