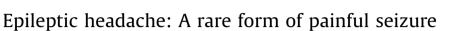
Contents lists available at ScienceDirect

Seizure

journal homepage: www.elsevier.com/locate/yseiz



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ARTICLE INFO

Article history: Received 4 July 2017 Received in revised form 9 October 2017 Accepted 12 October 2017 Available online xxx

Keywords: Epileptic headache Ictal epileptic headache Painful seizure Aura Hemicrania epileptica Migralepsy

ABSTRACT

Purpose: To describe the concept, features and mechanisms of epileptic headache (EH). *Methods:* Analysis of all published articles concerning EH and related subjects. *Results:* There are more than 30 published case studies of patients with headache as the only

manifestation of a seizure, a condition that has been variously called "EH", "ictal epileptic headache", "hemicrania epileptica", "cephalic pain seizure". It is necessary to differentiate EH from "migralepsy" and "ictal non-epileptic headache".

EH may be an isolated event or the initial phase of a seizure followed by other manifestations. An isolated EH is clinically relevant because it is often symptomatic of structural brain disease; this underlines the importance of a differential diagnosis as the head pain of EH has no specific diagnostic characteristics. The described cases indicate that the location of the foci may vary, thus suggesting the involvement of different parts of the pain network. EH is a "focal aware" seizure, but there are a few reports of cases in which it was associated with generalised epileptiform activity. A correct diagnosis of EH requires an ictal EEG recording showing epilepsy-compatible discharges that coincide with the onset and cessation of the headache. A rapid response to the acute administration of an antiepileptic drug may support the diagnosis.

Conclusions: EH is a particular type of pain seizure that has a complex pathophysiology and, when isolated, requires differential diagnostic consideration. We believe that, although it is not frequent, pain as an ictal symptom should be highlighted in the operational classification of seizure types.

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1. Introduction

There has recently been growing interest in reports of patients having headache as the only manifestation of an epileptic seizure, [1-11] and it has been suggested that the most appropriate term for this condition is "epileptic headache" (EH), [12] although called "ictal epileptic headache" by some authors. [10] However, there is still debate concerning the characteristics, clinical features and mechanism(s) of EH, and some published reports show that it is not always correctly identified. [13,14]

In this paper, the term EH will be used to define an episode of head pain caused by an epileptic discharge: i.e. a painful seizure. According to the recent ILAE operational classification, [15] EH is a "focal aware" seizure corresponding to the former definition of "aura" and, as such, can occur in isolation or precede other more clinically recognisable epileptic manifestations. The aims of this review are to analyse the clinical characteristics of the cases reported in the literature, suggest criteria for a correct diagnosis of EH, and discuss the mechanisms underlying it.

2. Methodology

A systematic search of the PubMed database was made in order to identify all of the articles containing the words "epileptic headache", "headache and epilepsy", "migraine and epilepsy", "ictal epileptic headache", "hemicrania epileptica", "migralepsy", "migraine-triggered seizures" and "ictal headache (for which PubMed also gives pre-, post and inter-ictal headache).

For practical reasons of brevity, some articles that merely discuss previously published data without introducing any new elements have not been cited.

3. Published cases

* Corresponding author. E-mail address: cianchet@unica.it (C. Cianchetti). It should be noted that the terms 'epileptic headache' [16], "epileptic cephalea" [17] or "céphalée epileptique" [18] were once

https://doi.org/10.1016/j.seizure.2017.10.010

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Characteristics of the 32 sufficiently documented cases reported in the literature.

Case No.	Authors		Headache characteristics-location/ duration	EH accompanying symptoms	Manifestations occasionally following EH	Ictal EEG	Interictal EEG	Other seizures	MRI	Etiology
	Laplante et al. [20] case 1	17/F	vertex/1 min	dyspnoea, headedness, sometimes could not talk	automatisms, confusion, agitation	scalp: "bitemporal"/deep: r.hippocampus & amygdala	n.r.	drop attacks	n.r. (CT normal)	temporal focal malformation?
2	Laplante et al. [20] case 2	28/ M	temporal/30–60 s	n.r.	chewing, head-version, agitation, rarely generalised seizure	scalp: normal or 3–4 Hz or rapids/deep: spikes r. hippocampus	(normal)	no	n.r.	early encephalopathy
}	Isler et al. [21] case 1	22/ F	MA visual & auditory/ sec min	?	?	stereo-EEG: r. temporal, limbic discharges	n.r.	"complex partial"	n.r.	n.r.
ļ	Isler et al. [21] case 2	29/ M	MA (aura:strange feeling, speech arrest)/ sec-min	?	?	stereo-EEG: l. medial fronto-basal, supplementary motor (?)	n.r.	"complex partial"	n.r.	ischemic lesion
5	Isler et al. [21] case 3	52/ M	MA visual/sec-min.	?	?	Stereo-EEG: r.temporal, amygdala	n.r.	"partial complex"	n.r.	astrocytoma
5	Isler et al. [21] case 4	17/ M	left side/min	confusion, speech arrest, nausea	?	theta fronto-temporal	n.r.	n.r	widening l.frontal horn & sylvian	n.r.
7	Isler et al. [21] case 5	39/ M	bifrontal/hours?	scintillating vision, nausea	?	s&w generalised	sharp-waves bifrontal	GTC	n.r.	n.r.
8	Lindner et al. [23]	41/ M	r.MA/days?	nausea, photophobia; l. hemianopsia, l.hemi- hypo-dysesthesia	none	r.occipito-parietal spikes	no	no	hyperintense meningeal signal over r.visual cortex	transient perfusion changes
	Beauvais et al. [22] case 1	16/ F	bitemporal/n.r.	speech arrest, fear of falling	focal motor	spikes l.fronto-central	n.r.	focal motor	n.r.	n.r.
0	Beauvais et al. [22] case 2	20/ M	r.frontal/minutes	n.r.	focal motor	spikes l.parietal	spikes l. parietal	focal motor	n.r.	cortical dysplasia
1	Ghofrani et al. [1]	9/M	bilateral/more than 24 h	agitation, irritability, cry, moaning	no	continuous GSW	normal	no	brain atrophy	(histiocytosis)
2	Parisi et al. [2]	14/ F	MO r.backside/3 days	phono-photophobia, nausea, vomit	l.arm tonic, head version, then GTC	theta & sharp waves r. occipital	IPS r.sharp- waves parieto- temporal	reported as "GTC" (focal then GTC?)	normal	occipital epilepsy Gastaut-type
3	Perucca et al. [3]	56/ F	1)r.MA/ <1 min 2)l.MO/h (status)	1)l.hemivisual spots; 2)phono-photophobia, nausea	1) head deviation; 2)no	r.temporo-occipital rhythmic 11–12 Hz spikes, then slow-waves	n.r.	visual/ automatisms/ GTC	bilateral occipital cortex swelling, hyperintensity	perinatal hypoxic ischemic encephalopathy
4	Belcastro et al. [4]	20/ F	MO r.hemicranial/3 days	phono-photophobia, nausea, vomit	no	r.occipital rhythmic 11– 12 Hz spikes	n.r.	no	r.parieto-occipital scar/ictal DWI: r.occipital diffusion	post-trauma
5	Dainese et al. [5] case 1	11/ M	left side & vertex/ seconds	n.r.	visual hallucinations, oculoversion, loss contact	scalp EEG normal/stereo- EEG spikes r.occipito- parietal	normal	no	r.occipital (lingual) dysplasia	malformation
6	Dainese et al. [5] case 2	47/ F	r.frontal/few seconds	n.r.	tonic posture r.arm	spikes l.central	n.r.	no	l.inferior parietal tuber	tuberous sclerosi
7	Fusco et al. [6]	18/ M	MO/min 1–2 days	phono-photophobia; sometimes nausea, vomit	no	seizure A: spikes r.fronto- central; seizure B: r. occipital	rare focal discharges	"focal"	partial hemispherectomy; ictal SPECT seizure B: r.occipital hyperperfusion	Rasmussen's encephalitis
8	Fanella et al. [7]	37/ F	tension-type headache/ 1 h	no	no	subcontinuous GSW and GPSW	GSW and GPSW	absences & tonic-clonic	normal	idiopathic generalised epilepsy
9	Cianchetti et al. [8]	9/F	frontal/1-3 min	hypersensibility to noises	no	r.temporal spikes & slow- waves, then diffuse ^a	normal	no	dysplasia r. temporal cortex	dysplasia type IIa
20	Saitowitz et al.[9] case n.2	13/ M	Frontal sharp pain/min	unable to talk, urine incontinence	confusion 20–30 s	l. frontal spike and slow waves	normal	no	normal	not known
21	case il.2	4/F	MO r hemicranial/3 days		n.r.		l.spikes and sw			

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