

Secondary nummular headache: a literature review

Cefaleia numular secundária: Revisão de literatura

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ABSTRACT

Introduction: Despite being recognized only as a primary headache, some studies in the literature suggest the existence of nummular headache secondary to structural lesions and other diseases. The aim of this literature review is to compile the cases of probable secondary nummular headache reported in the literature. **Methods:** The terms "nummular headache" and "coin-shaped headache" were searched in five databases (MEDLINE, Scopus, SciELO, LILACS and Cochrane Library). Only articles that described cases of patients diagnosed with nummular headache were included. The cases were considered as secondary when a cause for the headache was suggested by the authors. **Results:** After excluding reviews and non-related or duplicated studies, 50 articles were included. A total of 343 cases of nummular headache were, at least, mentioned by these studies, and 30 of them were considered as secondary. **Conclusion:** there are few reports of secondary nummular headache in the literature; still, they represent a considerable percentage of the reported cases of nummular headache. They support that structural lesions and other diseases should be considered in the differential diagnosis of primary nummular headache.

Keywords: Nummular headache, Headache

RESUMO

Introdução: Apesar de ser reconhecida apenas como uma cefaleia primária, alguns estudos na literatura sugerem a existência de cefaleia numular secundária a lesões estruturais e outras doenças. O objetivo dessa revisão de literatura é compilar os casos de provável cefaleia numular secundária reportados na literatura. **Métodos:** Os termos "nummular headache" e "coin-shaped headache" foram pesquisados em cinco bases (MEDLINE, Scopus, SciELO, LILACS e Cochrane Library). Apenas artigos que descrevem casos de pacientes

diagnosticados com cefaleia numular foram incluídos. Os casos foram considerados secundários quando uma causa para a cefaleia foi sugerida pelos autores. **Resultados:** Após excluir artigos de revisão e artigos não relacionados ou duplicados, 50 artigos foram incluídos. Um total de 343 casos de cefaleia numular foram, ao menos, citados por esses estudos, e 30 deles foram considerados secundários. **Conclusão:** Existem poucos relatos de cefaleia numular secundária na literatura; ainda assim, representam uma porcentagem considerável dos casos reportados de cefaleia numular. Eles apoiam a ideia de que lesões estruturais e outras doenças devem ser consideradas no diagnóstico diferencial de cefaleia numular primária.

Palavras-chave: Cefaleia numular, Cefaleia

INTRODUCTION

Nummular headache is described in the International Classification of Headache Disorders 3rd edition (beta version) (ICDH 3-β)⁽¹⁾ as a frequently chronic pain located in a small circumscribed area of the scalp, not caused by structural lesions. It can be continuous or intermittent, being sharply contoured, fixed in size and shape, round or elliptical and between 1 and 6 cm of diameter.⁽¹⁾

Despite being recognized only as a primary headache, some studies in the literature suggest the existence of nummular headache secondary to structural lesions and other diseases. In this context, the aim of this literature review is to compile the cases of probable secondary nummular headache reported in the literature.

METHODS

The terms "nummular headache" and "coin-shaped headache" were searched in five databases (MEDLINE, Scopus, SciELO, LILACS and Cochrane Library). The review was performed up to 2015. Only articles that described cases of patients diagnosed with nummular headache were included. The cases were considered as secondary when a cause for the headache was suggested by the authors.

RESULTS

The search in MEDLINE, Scopus, SciELO, LILACS and Cochrane Library yielded 68, 89, one, one and no articles, respectively. After excluding reviews and non-related or

duplicated studies, 50 articles were included. A total of 343 cases of nummular headache were, at least, mentioned by these studies, and 30 of them were considered as secondary.

Table 1 describes all the 30 cases and their suggested etiologies. Half of them were related to autoimmune diseases. Other possible causes associated with the onset of the headache were superficial masses (3 cases), tumors in the sellar region (3 cases), head trauma (3 cases), aneurysms (2 cases), insect bite (1 case), varicella-zoster infection (1 case), craniosynostosis (1 case) and a protruding bone lesion (1 case).

The distribution of characteristics (Table 2) was similar to those of the two previous literature reviews.^(2,3) Nummular headache was more common in women (73.3%), being restricted to a circular area (73.3%) in the parietal region

Table 1 - Cases of nummular headache with suggested etiologies

Study	Age (sex)	Site (side)	Size ^a	Quality (intensity)	Suggested etiology	Effective treatment ^b
Dash, 2006	27 (M)	P (L)	2.5	Burning (mod)	Head trauma	None
Guillem, 2007	60 (F)	T (L)	-	- (mild)	Subtentorial meningioma	Surgery
Pareja, 2008	65 (F)	P (R)	4.5	Stabbing (sev)	Head trauma	-
	60 (M)	P (L)	4	Pressing (mild)	Insect bite	-
Alvaro, 2009	67 (M)	P (L)	5	Pressing (mild)	Protruding lesion	Nerve block
	72 (F)	V	4	- (mod)	Post-op. (pit. adenoma)	None
Guillem, 2009	52 (F)	PO (R)	2.5	-	Arachnoid cyst	None
	36 (F)	PT (R)	5	Pressing (mod)	Arachnoid cyst	Pregabalin
Moon, 2010	77 (M)	P (L)	10	-	Head trauma	-
Chen, 2012a	62 (F)	P (L)	4	Electric (mod)	VZ infection	Fanciclovir, gabapentin
Chen, 2012b	75 (F)	P (R)	-	-	Prim. SjS	-
	52 (F)	P (B)	-	-	Prim. SjS	-
	38 (F)	PT (R)	-	-	RA, APS	-
	48 (F)	P (R)	-	-	SS	-
	74 (F)	P (L)	-	-	Prim. SjS, RA, AS	-
	73 (F)	P (L)	-	-	APS	-
	64 (F)	P (L)	-	-	APS	-
	72 (F)	P (R)	-	-	SS	-
	66 (F)	P (R)	-	-	SjS	-
	51 (F)	P (B)	-	-	SS	-
	68 (F)	P (R)	-	-	SS, prim. SjS	-
61 (F)	P (L)	-	-	Prim. SjS	-	
57 (F)	P (R)	-	-	Prim. SjS	-	
47 (M)	P (R)	-	-	RA	-	
Chui, 2013	54 (F)	V	3	Electric (mod)	Pit. oncocytoma	Surgery
Irimia, 2013	33 (F)	O (R)	3	Boring (mod)	Alopecia areata	Botulinum toxin A
Yin, 2013	58 (F)	V	4.5	Electric (-)	Post-op. (pit. prolactinoma)	Gabapentin
Mesonero, 2014	41 (F)	P (L)	3	Pressing (-)	Craniosynostosis	No treatment
Ruiz, 2014	67 (M)	T (R)	4	Pressing (sev)	TA fusiform aneurysm (R)	Surgery
	60 (M)	O (R)	5	Boring (mod)	OA fusiform aneurysm (R)	Triptans

M - male; F - female; P - parietal; T - temporal; O - occipital; PT - Parieto-temporal; PO - Parieto-occipital; V - vertex; L - left; R - Right; mod - moderate; sev - severe; post-op. - postoperative; pit - pituitary; VZ - Varicella-zoster; Prim. SjS - primary Sjogren syndrome; RA - rheumatoid arthritis; APS - antiphospholipid syndrome; SS - sicca syndrome; AS - ankylosing spondylitis; TA - temporal artery; OA - occipital artery. a - in cm, representing the wider diameter; b - treatment with the best results.

(66.7%). It presented more frequently as a mild to moderate (83.3%) pressing (41.7%) pain in a continuous pattern (85.7%), with exacerbations (58.3%), sensory disturbances (46.7%) and tenderness to palpation (56.3%).

The pain was, diminished in half of the cases in which anticonvulsants, as gabapentin, were used, while antidepressants, as amitriptyline, were not useful in any patient. The headache disappeared only in two patients treated with gabapentin^(4,5) and in the three who underwent surgical procedure.⁽⁶⁻⁸⁾

Table 2 - Characteristics of possible secondary cases described in the literature.

Characteristics	
Gender, % (M/F, 30 cases)	26.7/73.3
Age when reported (30 cases), y	57.9 (29-77)
Duration of NH (22 cases), y	3 (0-22)
Prior or concurrent headache (24 cases), %	29.2
Side of the headache (30 cases)	
Left, %	40
Right, %	43.3
Midline, %	10
Bilateral, %	6.7
Localization (30 cases)	
Parietal, %	66.7
Vertex, %	10
Temporal, %	6.7
Occipital, %	6.7
Parieto-temporal, %	6.7
Parieto-occipital, %	3.3
Morphology (15 cases)	
Mean diameter (range), cm	4.5 (2.5-10)
Circular, %	73.3
Oval, %	26.7
Pain quality (12 cases)	
Pressing, %	41.7
Electric, %	25
Boring, %	16.7
Stabbing, %	8.3
Burning, %	8.3
Pain intensity^a (12 cases)	
Mild to moderate, %	83.3
Severe, %	16.7
Exacerbations (12 cases), %	58.3
Sensory disturbances (15 cases), %	46.7
Tenderness (16 cases), %	56.3
Trophic changes (14 cases), %	21.4
Temporal pattern^b (14 cases)	
Chronic-continuous, %	85.7
Episodic, %	14.3
Treatment responsiveness^c (13 cases)	
Analgesics (8 cases), %	12.5
Anticonvulsants (8 cases), %	50
Antidepressants (5 cases), %	0
Nerve block (4 cases), %	25
Botulinum toxin (1 case), %	100
Surgery (3 cases), %	100
Triptans (2 cases), %	50

M - male; F - female; NH - nummular headache. a - based on the visual analogue scale; b - based on presence or absence of remissions; c - considered when resulted in any improvement of the pain.

DISCUSSION

The current study reviewed the cases of nummular headache in the literature and described the information from studies that associated this headache to other conditions. While case reports and observational studies have been increasing in number, only few articles have suggested a cause for the reported cases,⁽⁴⁻¹⁶⁾ and a secondary entity for nummular headache is still not recognized.

The most common characteristics of these cases were similar to those found by Dai et al.⁽²⁾ and Schwartz et al.,⁽³⁾ as well as to those described in the ICDH 3- β ⁽¹⁾ for primary nummular headache. In fact, according to the ICDH 3- β ⁽¹⁾ diagnosis of nummular headache is made after exclusion of structural causes. Furthermore, a correlation between the headache and other diseases does not mean that the former is caused by the latter, and it is also possible that some of the cases were not related to the patient's history nor their concurrent conditions. However, some of the reports are highly suggestive of a causation relationship.

The three most compelling examples are described by Guillem et al.,⁽⁶⁾ Chui et al.⁽⁷⁾ and López-Ruiz et al.⁽⁸⁾ The patients were diagnosed with, respectively, a subtentorial meningioma, a pituitary oncocyoma and a fusiform aneurysm in the right temporal artery, after presenting nummular headache as the only or major symptom. The headache characteristics were different between the cases, but its location was related to the site of the pathology in all of them. The pain disappeared rapidly after the surgery and did not recur in the three patients.

Head trauma was suggested as the cause of nummular headache in only three cases reported in the literature,^(11,15,16) while it was not considered as a probable cause in other cases with history of trauma.⁽¹⁷⁾ The relationship between trauma and nummular headache is very difficult to establish; however, of the two cases with information available, one⁽¹⁵⁾ had a headache starting immediately after the event, and both^(11,15) had the location of pain restricted to the site of the trauma.

The difficulty in distinguishing primary from a possible secondary nummular headache has implications regarding its clinical assessment. As the headache is commonly mild and the possibility of an underlying cause is not often recognized, secondary conditions may be underdiagnosed, at least until the presentation of other symptoms. Despite of their rarity in the literature, secondary nummular headaches should be considered, and imaging evaluation can be a helpful tool for an early diagnosis.

The described cases also provide information to the debate regarding the pathogenesis of nummular headache. As the patient with the pituitary oncocytoma,⁽⁷⁾ two patients developed the headache in the vertex after surgery in the sellar region.^(5,9) The location in the midline and the possible referred aspect of the pain support a central mechanism for nummular headache.^(5,9) Meanwhile, the cases of subtentorial meningioma⁽⁶⁾ and arachnoid cysts,⁽¹²⁾ for example, suggest a peripheral mechanism by traction of dural nerve branches due to expansion of such lesions.^(6,12)

It is important to emphasize that the results of the present review should be analyzed carefully, as the cases and studies are scarce, and some details are lacking in many reports. These factors influenced the frequencies found for the headache features, especially the rate of suggested causes. One example is the high amount of autoimmune diseases, only described by two articles.^(10,13) Nevertheless, the information is useful in order to provide more attention to the existence of secondary nummular headaches.

In conclusion, there are few reports of secondary nummular headache in the literature; still, they represent a considerable percentage of the reported cases of nummular headache. They support that structural lesions and other diseases should be considered in the differential diagnosis of primary nummular headache. Only by recognizing it as an actual entity and by reporting new cases is that a better understanding about its clinical characteristics can be promoted, improving the diagnostic evaluation and the use of complementary exams.

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