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Nummular Headache in a Young Nigerian Man-Associated with History of Load Carrying on the Head

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Authors' contributions

This work was carried out in collaboration among all authors. Author MAF evaluated, managed the patient and wrote the protocol, the first and final draft of the manuscript. Author FJ carried out the psychiatric assessment of the patient and wrote out the report of the psychiatric assessment and author NI managed the literature review. All authors read and approved the final manuscript.

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Case Study

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ABSTRACT

Nummular Headache (NH) is a rare headache disorder characterized by sharply contoured pain fixed in size and shape within a round or elliptical region of the head.
We present Mr EAO, a 25 year old young man who presented with a history of recurrent well-circumscribed headache of about 7 years duration over a circular area on the vertex of the head measuring, 3.0 cm × 3.0 cm in size, and exhibited a clear-cut boundary from non-painful surrounding.
A brain CT scan showed normal brain and right maxillary sinus polyp with bilateral chronic maxillary sinusitis. An assessment of nummular headache to rule out trigeminal neuralgia was made.
Capsules Tramadol and Neurobion were added to his previous medications of amiptryptilline and Pregabalin and there was a good clinical response.
This patient fulfils most of the International Headache Society diagnostic criteria (ICHD-3 beta) for NH and is currently stable on his medications.

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1. INTRODUCTION

Nummular headache, previously called Coin-shaped headache, is one of several relatively uncommon headache syndromes that may occur either as a primary headache or as a headache symptomatic of potentially malignant processes [1]. It is a rare entity and only a limited number of patients have been described worldwide [1,2]. Dai *et al* reported a case of a 63 year old male suffering from focal head pain after a cold shower in the summer of 1994. The painful area was perfectly circular, with a diameter of 10 cm in the vertex. The pain was continuous, pulsatile, severe and did not change in size or shape. After several futile therapeutic attempts, the patient requested for a surgical removal of the tendinous part of the epicranius muscle in the symptomatic area but surprisingly, the pain re-appeared in another area that overlapped the previous symptomatic area. In their literature review, they further described other instances of NH such as bi-focal, multifocal and large diameter coin-shaped headache which remained confined to the same area and did not change in shape and size over time [2]. More than 200 cases of nummular headache have been reported since it was first defined in 2002, but the pathogenesis remains unclear [2]. An extensive search of the electronic media did not yield any information on nummular headache in Nigeria. We searched the PUBMED database for nummular headache with emphasis on studies on headache conducted in Nigeria using a combination of the following words; headache, prevalence, epidemiology, subtypes, treatment and prevention issues in Nigeria. Hard copies of articles not found in Pub Med were also searched for and included. There was no information on nummular headache in Nigeria.

This is probably the first documented case in the literature from Nigeria signifying that nummular headache does occur but remains mostly undiagnosed .NH is universal and cuts across geographical, racial and gender divides.

The diagnostic criteria consist of a pain of highly variable duration, but often chronic, in a small circumscribed area of the scalp and in the absence of any underlying structural lesion [3]. The pain is generally mild or moderate, commonly described as oppressive or stabbing, and lasting minutes, hours, or days, with a remitting or unremitting pattern, with lancinating exacerbations lasting for several seconds or gradually increasing from ten minutes to two hours may superimpose the baseline pain [4].

The temporal pattern is either chronic or remitting [4]. During and between symptomatic periods, the affected area may show variable combinations of hypoesthesia, dysesthesia, paraesthesia, tenderness, trophic changes [5] or allodynia, especially when the headache remits [6]. The diagnosis of NH remains infrequent because it is easily benign and not easily recognized by non –neurologists [7].

2. CASE REPORT

Mr EAO, 25 year old single Nigerian man, a phone accessory seller, resident in Ado Ekiti, The capital city of Ekiti state in Southwest Nigeria who presented with history of recurrent headache of about 7 years duration. The pain was persistently localised to the vertex of the head over a well-circumscribed circular area measuring 3x3 cm, which also exhibited a clear cut boundary from non-painful surrounding areas. The pain peaked and subsided spontaneously. It was mild to moderate in intensity, lancinating or electric in nature and sometimes disabling and is often precipitated or exacerbated by cold weather, loud noise or ingestion of beer. The pain was occasionally severe enough to restrict daily activities and was said to be the reason why he dropped out of secondary school. The pain did not radiate to any other part of the head. It was not associated with periorbital pain, excessive lacrimation; the pain did not exhibit any periodicity and was not related to posture. There was a positive history of carrying heavy logs of wood on his head in the village for more than 10 years which he, however stopped a few years ago. No history of previous head injury, fever, neck stiffness, or swelling over the head. Positive history of poor sleep pattern was also present. The protracted nature of the headache has been a major concern and a source of worry to the patient. The patient has consulted so many health practitioners both orthodox and unorthodox in search of a permanent cure for this unpleasant sensation.

On examination, he was healthy looking though anxious, not in any obvious distress, not pale, afebrile, anicteric and nil pedal oedema.

CNS examination was unremarkable: he was conscious and alert, well oriented with preserved

cognitive functions. There were no signs of meningeal irritation or obvious cranial nerve palsy. Both pupils were large and showed sluggish reaction to light. No facioparesis and no focal or global motor/sensory deficits.

Cardiovascular examination showed a pulse rate of 108bpm, full volume and regular. BP: 151/81mmHg. Normal heart sounds S1 and S2 were heard. The chest was clinically clear.

On abdominal examination, the abdomen was flat, soft, moves with respiration. There was no palpable organomegaly or tenderness.

Investigations done were as follows:

Full blood count done showed PCV of 48.4%, white blood cell count of 8,400cells/cm3 and platelets of 179,000cells/cm^3. ESR was 1mm in the first one hour. Haemoglobin genotype is AA.

A brain CT scan showed normal brain and right maxillary sinus polyp with bilateral chronic maxillary sinusitis. An assessment of nummular headache to rule out trigeminal neuralgia was made.

Further assessment by a psychiatrist revealed that the patient had features of moderate depression through the administration of the Hamilton Depression Scale (a score of 15 was recorded). On the Hospital and Anxiety Scale (HADS), patient had borderline abnormal rating with a score of 10 but had no features of anxiety. Psychotherapy was given to the patient. He was also seen by an ophthalmologist who found features of early glaucoma. The Optical Coherent Tomography analysis of Optic Nerve Head and Retinal Nerve Fiber Layer showed significant thinning of the neuro-retinal rim, suggestive of glaucoma

He was earlier been maintained on Pregabalin and Amiptryptilline and capsules tramadol 50 mg twice daily and tabs Neurobion 1 tab daily were added for a few days.

Patient is currently clinically stable on his current routine medications and is being followed up in the neurology clinic of our hospital.

3. DISCUSSION

Nummular headache is a rare entity and only a limited number of patients have been described worldwide. This is possibly the first case to be documented in the literature from Nigeria because there was no previous case reported based on our PUBMED search and a review of hard copies of articles on headache in Nigeria which were not on PUBMED.

NH is not frequently described in the literature, the true incidence likely exceeds the number of reported cases as the condition is mostly unrecognized as many patients with NH may not seek medical help due to the mild/moderate pain intensity [7]. Also, the condition may have been either ignored or shown only to general practitioners without further follow-up by a neurologist due to its relatively benign nature [7].

In a study carried out in USA, a review of more than 250 published cases suggests a female predominance, with a gender ratio of approximately 1.8:1 [8]. The mean age of onset is 45.4 years and the mean duration of symptoms before presentation is 4.4 years (range, less than 1 month to 50 years).In this index case, the diagnosis of nummular headache was made after 7 years the onset of the symptom. In most cases there is no identified precipitant, although 12.8% report a remote history of head trauma. Almost half of NH patients (46.7%) carry concurrent or prior headache diagnoses including migraine, tension-type head-ache, medication overuse headache, and primary stabbing headache [8].

In another study carried out in Spain, A total of 11 females and 3 males were studied. The incidence was 6.4/100,000/year [9]. The mean age at the onset was 38 years (range: 13□72). Only three patients had another concurrent headache: migraine (n = 2), and trigeminal neuralgia (n = 1) which proved to have an independent course [9].

In another study carried out in United Kingdom, Sixteen patients were included in this series (6 men, 10 women) (10). The mean age at onset was 50 years (range, 19–79 years). No paediatric cases were identified. Mean duration of headache defined as the time until resolution or up until time of interview was 7.9 years including cases that were 14, 16, 21, and 40 years in duration [10]. Post-traumatic nummular headache was identified in one patient and was noted to be temporal, beginning immediately after trauma [10].

Even though it is considered as a primary headache syndrome, NH has also been described in patients with intracranial lesions [7]. Although NH is mostly regarded as a primary

disorder, various focal headaches with a nummular pattern have been attributed to local lesions of the scalp (fusiform aneurysm of a branch of the superficial temporal artery), the skull (fibrous dysplasia), Paget's disease, or adjacent intracranial structures (meningioma, arachnoid cysts) [8].

The diagnostic criteria of NH defined according to the guidelines of the International Classification of Headache Disorders (ICHD), 3rd edition (Beta version) include the following [3].

A. Continuous or intermittent head pain fulfilling criterion B
B. Felt exclusively in an area of the scalp, with all of the following four characteristics
1. Sharply contoured
2. Fixed in size and shape
3. Round or elliptical
4. 1-6 cm in diameter
C. Not better accounted for by another ICHD -3 diagnosis

The pain is generally mild or moderate, commonly described as oppressive or stabbing, and lasting minutes, hours, or days, with a remitting or unremitting pattern, with lancinating exacerbations lasting for several seconds or gradually increasing from ten minutes to two hours may superimpose the baseline pain [4].

The pain remains confined to the same symptomatic area, which does not change in shape or size with time [9]. It is exclusively felt in a rounded or elliptical area typically 2-6 cm in diameter [4]. Although any region of the head may be affected, the parietal area is the common localization of nummular headache. The temporal pattern is either chronic or remitting. During and between symptomatic periods, the affected area may show variable combinations of hypoesthesia, dysesthesia, paraesthesia, tenderness, trophic changes [5] or allodynia, especially when the headache remits. Also, a doctor may be able to reproduce the tenderness when pressing on the area during a physical examination.

The likely cause of a nummular headache is a localized irritation or neuralgia of one of the branches of the trigeminal nerve [6]. This is probably true for our patient who had a long history of carrying heavy loads on the head for a long time prior to the onset of his symptoms. In NH, the pressure pain threshold is decreased in the symptomatic area, while tenderness is not increased compared either to the non-symptomatic area in the same patient or to that of healthy subjects. These two findings support the fact that NH might be a non-generalised disorder with sensitization restricted to the painful area [2,6,11].

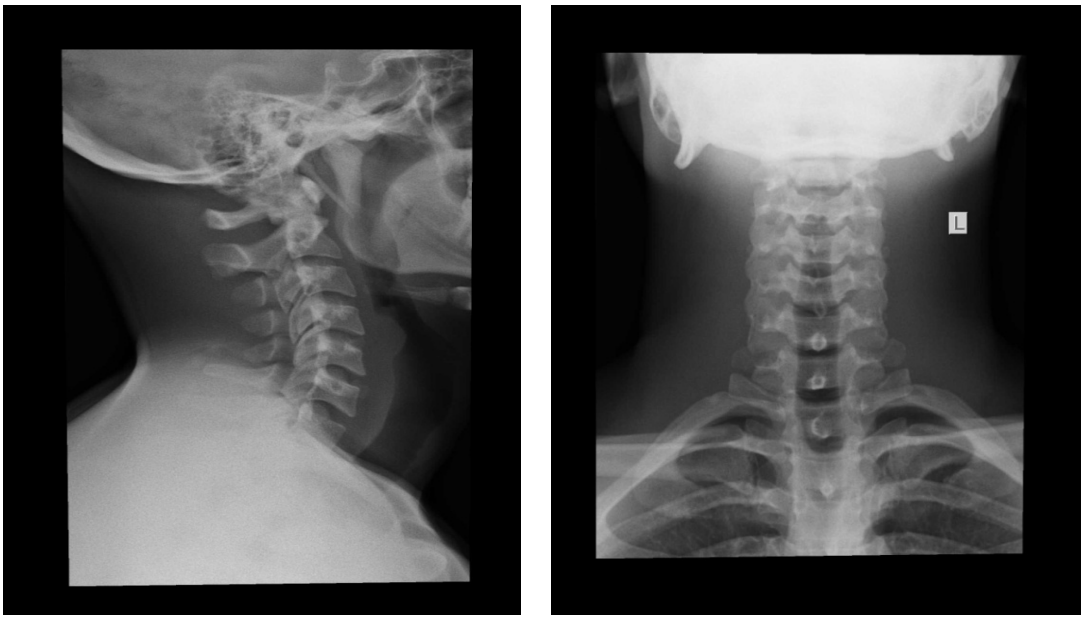

Fig. 1. cervical spine xray of the patient showing normal findings

Fig. 2. The brain CT scan of the patient showing normal findings in the brain

Since the initial paper describing NH, many case series and reports have been published that have speculated on the possible aetiology and pathophysiology of NH [11]. There has been evidence presented favouring both peripheral and central aetiologies, as well as descriptions of primary and secondary NH [11]. Currently, it appears the majority of evidence supports a peripheral cause [11].

In favour of the peripheral aetiology, theory is the fact that NH is localized to a precise area of the scalp with local tenderness and sensory disturbances such as paraesthesia and allodynia. Moreover, the pressure pain threshold is reduced in the affected area [12] and botulinum toxin type A gives effective relief of pain. Finally, trophic changes in the affected area of the skin have been described some patients [13].

Some literature advice scalp examination for rash to rule out herpes zoster [6]. Serious causes of headache that mimic nummular headache include multiple myeloma and Paget's disease [6].

In favour of a central aetiology, patients have been described with bifocal and multifocal NHs

[14] as have patients in whom the painful area extends over the midline of the scalp, which would not be expected in cases with peripheral aetiology [7]. A few interesting case descriptions such as large diameter NHs with bitrigeminal hyperalgesia and a case of NH in which pain recurred even after focal scalp resection of the affected area have also favoured a central causation of NH [15].

Although, some authors have considered the possibility of psychogenic factors in the aetiology of NH. There was no significant relation between the Hamilton Depression Rating Scale and the occurrence of NH in a study carried out amongst a southern Indian population [7]. This is consistent with the previous studies which have also not found any psychological deficit in patients with NH. Moreover, it is usually considered that strictly unilateral head pain has an organic cause. However, the possibility that episodes of pre-existing NH may be triggered by stress or psychological factors is likely and requires evaluation by further studies [7]. In this index case, patient had a score of 15 on the Hamilton depression scale which suggests mild depression and an abnormal borderline rating with a score of 10 on the Hospital Anxiety and Depression Scale(HADS) which suggest the psychological impact of this illness on the patient may be as a result of several futile attempts to get a permanent cure for this unpleasant sensation in the past. This rather supports a cause-effect relationship.

Treatment is seldom necessary and in most cases simple reassurance is sufficient [4]; although the symptoms are debilitating in some patients necessitating pharmacological treatment.

In a report from Japan, they describe a patient with NH who was successfully treated with Neurotropin (NTP), an analgesic drug currently used in Japan and China for the treatment of chronic pain conditions [16]. Although it is unclear whether NH represents a focal, nociceptive-type pain stemming from epicranial tissues or neuralgia of a terminal branch of a pericranial sensitive nerve the anti-nociceptive action of NTP seemed beneficial against NH in this patient [16]. Gabapentin seems to be the most effective oral medication; subcutaneous injection of the area with botulinum toxin type A also seems to be effective, and should be considered as an alternative to Gabapentin [17]. Because it is rare, there is no outlined treatment [18]. Gabapentin and Pregabalin are both new generation antiepileptic drugs which are also used to treat pain. Patient was placed on Pregabalin in this case because Gabapentin was not readily available and there was a good response to pain treatment.

4. CONCLUSION

In conclusion, our case report supports the peripheral origin of NH but we cannot definitely exclude the central mechanism. It is possible that peripheral nociception may be the initiating factor for the maintenance of the process of central sensitization. NH may be as a result of a complex pathogenesis involving an interplay of both peripheral and central mechanisms. Further clinical observership and basic research will help us to unravel this enigma in the future.

CONSENT

A well-informed consent was obtained from the patient well before writing this manuscript which has been kept in the custody of the authors.

ETHICAL APPROVAL

As per international standard or university standard ethical approval has been collected and preserved by the authors.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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