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CASE REPORT

Rhinolith–A Rare Cause of Sunct Syndrome: A Case Report

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ABSTRACT

Short-lasting unilateral neuralgiform headache attacks with conjunctival injection and tearing (SUNCT) is a rare cause of unilateral headaches affecting predominantly males in usually the fifth decade. The pain is usually moderate to severe in intensity affecting the ocular and the periocular area. The mean duration of paroxysms is 1 minute, with a range of 5 to 250 seconds. A 25-year-old male complained of progressive unilateral left nasal obstruction for 8 months along with ipsilateral daytime headache predominantly in the periocular and temporal region, conjunctival injection, tearing and minimal eyelid edema. The patient was admitted and kept under observation for the reported symptoms. Nasal endoscopy and probing revealed a greenish/ dirty grey and gritty mass filling posterior part of the left nasal cavity at the time of a headache, there was right sided ipsilateral congestion in the conjunctiva, lacrimation minimal and periorbital edema neurological examination of the patient was normal. The patient was posted for endoscopic rhinolith removal followed by infundibulotomy and maxillary sinusotomy. At 3 week follow-up, the patient was relieved of all symptoms and required no medication. The patient was asymptomatic at 6 week and 6 months follow up. Thus a diagnosis of secondary SUNCT cause rhinolith was confirmed which resolved with rhinolith removal.

Key words: Headache, Rhinolith, Sunct syndrome

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INTRODUCTION

A headache is a common symptom encountered by the neurologists, otolaryngologists, and physicians in their daily practice, the cause of which, commonly eludes the physician. The term "headache" refers to a pain with a predominantly neurocranial location, distribution of which does not correspond to the territory of distribution of single nerve trunks.[1,2] Thus 'neuralgias' are a separate entity which refers pain in the distribution of a nerve or nerves affected.[3] In 2013, the International Headache Society (IHS) developed diagnostic criteria for headaches laying to rest the diagnostic anxiety of physicians and otolaryngologists and divided these into primary headaches, secondary headaches and neuropathies and facial pain.[4]

The SUNCT is a rare form of trigeminal autonomic cephalalgia (TAC) mostly described as a primary headache syndrome. An acute headache is usually accompanied by ipsilateral conjunctival injection, lacrimation, rhinorrhoea and nasal congestion. Short-lasting unilateral neuralgiform headache with autonomic symptoms (SUNA) has been defined by the International Headache Society (ICHD-3) as similar to SUNCT with less prominent or absent conjunctival injection and lacrimation but manifesting other autonomic symptoms like rhinorrhoea, miosis, ptosis or sweating.[5]

Rare posterior fossa lesions, including ipsilateral cerebellopontine angle arteriovenous malformations, brainstem cavernous hemangioma and base of skull bony abnormalities, prolactinomas and vascular malformations (meningioma causing SUNCT) have been described to be associated with SUNCT although majority cases remain of unknown etiology and pathophysiology.[6]

CASE REPORT

A 25-year-old male complained of progressive unilateral left nasal obstruction for 8 months along with ipsilateral daytime headache predominantly in the periocular and temporal region, conjunctival injection, tearing and minimal eyelid edema. A headache remained refractory to treatment from several physicians. Attacks of headache were spontaneous, excruciating, short lasting and on an average 15 to 20 per day with a mean duration of one minute and without significant periods of remission. There was no history of nocturnal pain, loss of sleep, ptosis, nausea, vomiting, phonophobia or photophobia. Anterior rhinoscopy revealed foul smelling purulent

discharge in left nasal cavity. Nasal endoscopy and probing revealed a greenish/dirty gray and gritty mass filling posterior part of left nasal cavity. At the time of a headache, there was right sided ipsilateral congestion in the conjunctiva, lacrimation, minimal and periorbital edema. Neurological examination of the patient was normal. Plain CT scan paranasal sinuses revealed a radio-opaque shadow in the left nasal cavity with osteomeatal complex blocked on the same side. The patient was posted for endoscopic rhinolith removal followed by infundibulotomy and maxillary sinusotomy. At 3 weeks follow-up, the patient was relieved of all symptoms and required no medication. The patient was asymptomatic at 6 weeks and 6 months follow-up. Thus a diagnosis of secondary SUNCT cause rhinolith was confirmed which resolved with rhinolith removal.

DISCUSSION

Rhinoliths are rare foreign bodies of the nose, which may be encountered accidentally during the course of a routine examination.[7] Their formation is caused by in situ calcification of intranasal endogenous or exogenous foreign material.[8] Rhinolith creates pressure on the lateral wall of the nose known to cause sluders neuralgia though it is not validated by the International Headache Society (HIS), trigeminal autonomic cephalagia (TACs) are a group of primary headache disorders characterized by unilateral pain in the trigeminal nerve distribution, associated with ipsilateral cranial autonomic features. Despite their similarities, these disorders differ in their clinical manifestations and response to therapy. TAC includes a cluster headache, paroxysmal hemicranias, and SUNCT each of which is characterized by the duration, frequency, circadian rhythm and response to therapy. The above patient presented with unilateral headache with cranial autonomic signs consistent with TAC. As the distribution, frequency and duration of a headache fulfill the criterion of SUNCT laid down by the HIS part -3, a diagnosis of SUNCT was made. After the surgical removal of the rhinolith, patient-reported improvement, and the headache disappeared after about a month. The rhinolith probably due to pressure on the trigeminal nerve innervated area on the lateral wall of the nose was responsible for headache, and a parasympathetic reflex activation led to the cranial autonomic signs.

Till date, there has been no specific treatment for SUNCT, but recently Lamotrigine has emerged as the first line therapy and recommended as relatively successful therapy.[8-10] Gabapentin and topiramate are advised as second-line drugs in patients not responsive to lamotrigine.

Trigeminal autonomic cephalagia (TACs) are comprised in section 1(3) of the International classification of headaches disorders third edition-beta (ICHD-3ß) and include: a cluster headache, paroxysmal hemicrania, short-lasting unilateral neuralgiform headache attacks, hemicrania continua, probable trigeminal autonomic cephalalgia.[3]

The SUNCT is part of a group of idiopathic primary headaches called TAC caused by activation of trigeminovascular nociceptive pathways along with reflex autonomic activation. The autonomic signs are explained by the trigeminal-parasympathetic reflex (TPR) secondary to the initiation of a parasympathetic reflex through trigeminal nerve activation. Usually SUNCT is idiopathic but secondary causes of SUNCT may arise from rarely neoplastic, vascular, traumatic and infectious causes. To the best of our knowledge, no report of rhinolith as a cause of SUNCT has been published in the world literature.[10]

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