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Expert Opinion

Trochleodynia and Migraine

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The small region of the superior oblique muscle pulley (trochlea) may be the source of a distinctive pain (trochleodynia) originated in the superior oblique muscle-tendon-trochlea complex.[1] Trochleodynia is mostly felt in the inner-upper angle of the symptomatic orbit and may extend to the ipsilateral forehead. Trochleodynia is commonly produced by trochleitis (primary or symptomatic)[2,3] or primary trochlear headache.[4] Furthermore, trochleodynia may be a trigger, common to different headaches. Concurrency with migraine may bring about a syndrome of trochlear migraine[1,3,4] with important therapeutic implications. Trochleodynia has also been associated with tension-type headache[4] and paroxysmal hemicrania[5] and may, in the future, be an integral part of the pathogenic mechanisms of other concurrent headaches.

CASE 1

This 54-year-old man was seen in headache consultation with a 5-month history of a right superior medial orbital pressure type pain with an intensity of 5-6/10 which had been constant since onset. The pain was not worse with eye movement, coughing, sneezing, or bending over. He had no visual complaints, tearing or redness of the eye, right-sided ptosis, or nares congestion or drainage.

There was a many-year history of bilateral migraine without aura occurring about once a month and migraine aura without headache occurring 1-2 times per year.

His primary care physician placed him on an antibiotic for a presumed sinus infection without improvement. An ENT physician obtained a CT scan of the sinuses with normal findings. An Ophthalmologist found a normal exam except for 1 mm of left eyelid ptosis with normal pupils. The first neurologist found a normal exam and obtained a normal MRI of the brain with and without contrast, and a Westergren erythrocyte sedimentation rate with normal findings. He was tried on loratidine, mometasone furoate monohydrate nasal spray, montelukast sodium, and gabapentin without benefit.

There was a past medical history of well-controlled hypertension on medication. Neurological examination was normal. There was tenderness of the right superior medial orbital area in the area of the right trochlea. He was then seen by a neuro-ophthalmologist with no additional findings.

CASE 2

A 36-year-old female patient had a family history of migraine in her father.

Since she was 10 years old she has been suffering from attacks of left hemicranial headaches that were

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diagnosed as migraine without aura. The pain was severe in intensity and pulsatile in character. The attacks were regularly accompanied by nausea, vomiting, and photo-phonophobia. Each episode lasted for about 12-24 hours. The usual frequency was of 1-2 episodes monthly but in the last 2 years progressively reached 2-4 attacks weekly. By the time she experienced such frequent attacks she also noticed almost continuous pain in the inner part of the left orbit, which persisted in between migrainous episodes. The situation was disabling both socially and professionally. The attacks had been treated with paracetamol, metamizol, ibuprofen, and naproxen that provided partial relief. Sumatriptan and almotriptan were effective in aborting migraine attacks but did not influence the course of the orbital pain. Preventive treatments with Nadolol, Flunarizine, and Amitriptiline were of no avail. Upon examination the left trochlear region was found extremely tender. Routine blood analysis was normal. A contrast MRI of the head and orbits was normal. Both ENT and Opthalmologic consultations were normal.

QUESTIONS: WHAT IS THE DIAGNOSIS? WHAT TREATMENT WOULD YOU RECOMMEND?

Both patients are migraineurs and are also suffering from a chronic, continuous pain, in the upper-inner angle of the orbit. Upon examination, there was tenderness on the superior medial area of the symptomatic orbit, ie, on the area of the trochlea. The clinical picture is consistent with pain stemming from the trochlear region. Trochlear pain (trochleodynia) is characterized by pain in the upper-inner angle of the orbit, occasionally spreading to the ipsilateral forehead, that typically increases upon palpation and supraduction[1-4] and is accompanied by tenderness in the trochlear region. The structures involved in the genesis of trochleodynia are the superior oblique muscle, its tendon, and the throclea itself. In a few patients the underlying process of trochleodynia is an inflammatory – usually primary – disorder of the trochlear/peritrochlear area – ie, trochleitis.[2,3] However, most patients have a recently recognized disorder named primary trochlear headache,[4] which can be conceived as a primary, non-inflammatory trochleodynia.

In trochleitis there is a typical induration and swelling of the inflamed trochlea which can be easily assessed by palpation. Eye movement restriction is not a typical feature of idiopathic trochleitis. This remark is important to distinguish idiopathic trochleitis from other syndromes of the superior oblique muscle-tendon-trochlea complex that typically present with restriction and diplopia. Brown's syndrome consists in a restrictive inability to elevate the eye into adduction specifically caused by an abnormality of the superior oblique muscle-tendon-trochlea complex. Congenital Brown's syndrome is a restrictive ophthalmopathy with shortening and fibrosis of the superior oblique muscle tendon that causes strabismus but no pain. Acquired Brown's syndrome[6-9] may be idiopathic, but it is generally produced by inflammatory processes such as rheumatoid arthritis, hyperthyroidism, lupus, psoriasis, or enteropathic artropathy (ulcerative colitis and Chron's disease). Exceptionally, it can be caused by sinusitis, trauma, or metastasis. Acquired Brown's syndrome of inflammatory nature may produce local pain and tenderness in the superior nasal orbit. Therefore, in cases of trochleitis with restriction and diplopia appearing a symptomatic form should be suspected, the disorder being better classified within the acquired Brown's syndrome.

Trochleitis may be documented by orbital echography or contrast enhanced CT or MRI of the brain, centered in the orbits. Routine blood work, ESR, [3] standard biochemical determinations, thyroid function with antitiroglobulin and antimicrosomal antibodies, ANAs, Rheumatoid factor, and urine analyses [4] are necessary to rule out a secondary form. Only exceptionally it is necessary to perform a biopsy. Etiology of both primary trochleitis and primary trochlear headache are largely unknown. However, several proposals have been intended. Idiopathic Trochleitis could represent a highly restricted form of orbital pseudotumor,[2] or an inflammatory myopaty/enthesopaty of the superior oblique muscle/tendon.[1] On the other hand, Primary Trochlear Headache could be due to a mechanic enthesopathy, a superior oblique muscle myofascial disorder,[10] or an

intraorbital neuropathy of the nearby nerves liable to persistent microtrauma by movements of the superior oblique muscle.

The two presented patients neither had an abnormal swelled and indured trochlea nor MRI showed abnormal findings in the trochlear region. In addition, analytical scrutiny was normal. Therefore, the plausible diagnosis is Primary Trochlear Headache. The diagnosis of Primary Trochlear Headache requires both the presence of trochleodynia and ruling out trochleitis and any structural abnormality in the trochlear region by both neuroimaging and analytical exams.

Follow-up: Both patients received an injection of local anesthetic and steroid by an oculoplastic surgeon with relief of their symptoms. Patient 2, also noted a great improvement of her migraine headache. In fact, no further migraine attacks have been reported, the follow-up period being 4 years.

Typically, trochleodynia is quickly relieved by injection of anesthetics on the sore trochlea, thus providing a confirmatory feature on the origin of the pain. In order to achieve a sustained relief, the peritrhoclear injection usually includes a mixture of anesthetic and longer acting steroid. This procedure proved to be simple, effective, and safe. In most patients one single injection is enough to provide long-lasting relief. Rarely, the procedure has to be repeated. On the contrary, systemic administration of NEAIDs or corticosteroids is of generally no avail.

There is an additional interesting point arising from the outcome after the therapeutic intervention: In patient 2 the successful treatment of trochleodynia was also effective to control migraine attacks. Patient 2 reported that her typical migraine attacks were hemicranial always recurring in the same side of the trochleodynia, co-localization and dual response to therapy suggesting both disorders may be pathogenically linked. On the contrary, in patient 1 the clinical course of migraine and trochleodynia seemed to be independent, both temporary and spatially.

Concurrence and co-localization of throcleodynia and migraine suggest a possible relationship. Moreover, successful treatment of either trochleitis[1,3] or primary trochlear headache[4] may improve concurrent migraine so indicating a pathogenic bond between both disorders. In such cases migraine was frequent or chronic, and strictly unilateral in the same side as trochleodynia. The term "trochlear migraine"[1] has been proposed to name the coexistence of strictly unilateral migraine and ipsilateral trochleodynia, with the improvement of migraine being dependent on the resolution of the trochlear complaint, thus indicating that the trochlear painful process could have contributed to the perpetuation – or worsening – of migraine.[1,3,4,11]

Painful inputs from both migraine and trochleodynia processes follow the pathway of the first branch of the trigeminal nerve. Concurrence of such afferences may overload the neurons of the caudalis trigeminal nuclei, resulting in temporal and spatial summation of neuron signals giving rise to a tendency to a progressive increase of headache, thus perpetuating or exacerbating migraine. Successful treatment of trochleodynia may considerably decrease the nociceptive contingent to the caudalis trigeminal nuclei, thus relieving such neurons from an excessive input and decreasing their overfiring. The liberation of those neurons from such trochlear peripheral sensitizers may be effective in controlling headache.

It is worth mentioning that trochleodynia may be masked by the concurrent migraine, so it requires direct questioning and appropriate examination. Accordingly, trochlear assessment should be incorporated in the evaluation of migraine, in particular in refractory cases or when migraine attains a frequent, even chronic course. When presenting as isolated pain, trochleodynia is easily recognized.

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