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Case Report

A Rare Cause of Facial Edema and Pain: Temporalis Muscle Hypertrophy

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Abstract

Temporalis Muscle Hypertrophy (TMH) is a rare entity of masticatory muscle hypertrophy. It is a disease of important differential diagnosis between peripheral nervous system dysfunctions and neuromuscular diseases. TMH is most commonly bilateral and usually associated with other types of masticatory muscles hypertrophy, such as masseter or pterygoid hypertrophy. However, isolated unilateral TMH is extremely rare. After collecting data from the electronic medical record and allowing the patient permission to report the anamnesis, we describe the clinical case of an unusual form of pain and facial edema. In this case report, we present an adult patient with unilateral temporalis muscle hypertrophy.

Keywords: Inflammatory Cells; Focal Myositis; Neuromuscular Diseases; Unilateral Temporalis Muscle Hypertrophy.

Introduction

Temporalis Muscle Hypertrophy (TMH) is a rare entity of masticatory muscle hypertrophy. TMH is most commonly bilateral and usually associated with other types of masticatory muscles hypertrophy. However, isolated unilateral TMH is extremely rare [1]. In this case report, we present an adult patient with a rare cause of facial edema: unilateral temporalis muscle hypertrophy.

Material and Methods

The data were collected from the patient and from an electronic medical record with the patient's permission. The patient signed the term clarification and agreement with medical research. The authors guarantee all ethical principles of research. Data accesses, data retention, confidentiality of patient's personal information, image integrity and clinical trial transparency have been preserved.

Case Report

A 57-year-old man was referred to the emergency medical service due to recurrent edema in the right temporal and periorbital region. The pain increased with increasing edema. There were no alleviation factors for the symptoms. The condition was associated with pain of mild intensity, without headache characteristics. The patient reports that the episodes occurred at large intervals, usually once every two months. And it took about 10 to 15 days to decrease the edema.

An important characteristic was the visual and psychological impact involved, since the people who saw him were scared with such edema in the facial region. The patient had already been treated for suspicion of allergic conditions and giant cell arteritis, excluding such diagnoses. He consulted several rheumatologists and neurologists before hospital admission, and did not succeed in diagnosis.

On evaluation in the neurology service, the patient presented unilateral edema on the right side. On physical examination, the patient reported pain of moderate intensity at the level of the ophthalmic and maxillary branches of the right trigeminal nerve. There was no visual impairment, autonomic signs, dysfunction in other cranial pairs or other associated symptoms. The patient followed up with investigation in the radiology department of the hospital, where the MRI examination was performed as shown in (Figure 1).

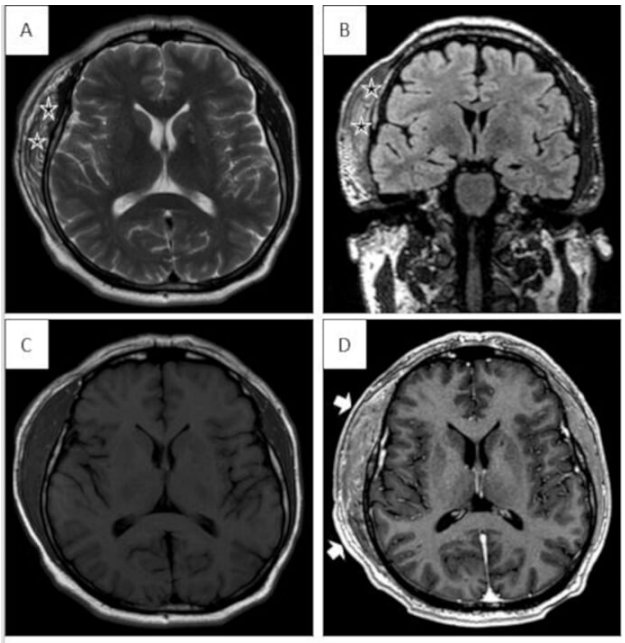

Figure 1: Axial (A) T2-weighted, (B) coronal T2/FLAIR-weighted MRI and (C) T1-weighted. Native followed (D) T1-weighted contrast-enhanced MRI show expansion and massive high signal intensity (black stars) see on (A) and (B), as well significant contrast enhancement (white arrows) involve the muscular compartment of the temporal muscle on (D) compared with (C).

The image shows the signal change with hyper intensity on T2 sequence in the temporalis muscle region on the right side. The suggested clinical condition was temporal muscle myositis.

The patient performed biopsy of the right temporal muscle, confirming the diagnosis of focal TMH according to (Figure 2).

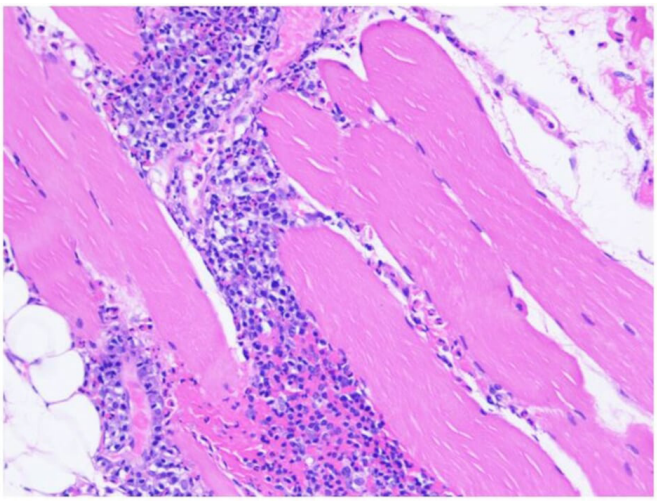

Figure 2: Hematoxylin and Eosin staining shows interstitial mononuclear and polymorphonuclear inflammatory infiltrates.

After the diagnosis, the patient started treatment with oral corticosteroids and showed a significant clinical improvement with reduction of pain and edema on the right side.

Discussion

Isolated unilateral TMH is an example of focal myositis. This disease is an inflammatory muscle disorder of usually unknown etiology. The diagnosis can only be established by exclusion of local infection, malignant tumors of the muscle, and connective tissue disease. The inflammatory cells represented mainly macrophages and CD8-positive cells. By electron microscopy, there is only minimal pathology of interstitial capillaries and no unequivocal evidence of undulating tubuloreticular profiles as often seen in dermatomyositis [2].

Isolated unilateral TMH is a rarely reported clinical entity. Consideration of a broad differential diagnosis combined with a detailed histological and radiologic work-up will help the physician diagnose the underlying pathology [3].

This disease is peculiar because there is no identifiable etiology, age category or side predominance. The potential etiological factors include local factors, such as bruxism, dental malocclusion, bony prominences leading to trauma and reactive hypertrophy ascribed to psychogenic factors [4].

There are different treatment modalities available for this disease. Some patients have not undergone any active intervention and were treated symptomatically [3].

Further treatment options include amitriptyline hydrochloride, corticosteroids, or the placement of a maxillary stent [5]. Botulinum toxin [6] may present an effective but less invasive alternative to surgical procedures [7] in cases not controllable by analgesic medication [5-7].

Author Contributions

Franzoi A.E.A designed the study, collected and analyzed the data, and wrote the article. Sotopietra B. collected the data. Monfredini N.H. collected, analyzed the data and reviewed the article. Magno M.V.M. reviewed the article. Wille P.R. reviewed the article. Roberge V.D. collected the data and reviewed the article.

Ethical Publication Statement

We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this report is consistent with those guidelines.

Disclosure and Permission

The authors report no disclosures. All authors and contributors agree to the conditions outlined in the Authorship and Contributor

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The authors take full responsibility for the data, the analyzes and interpretation, and the conduct of the research. There was no sponsorship for the scientific article and there was no conflict of interest with all the authors.

The author has received permission to cite any personal communications and has received and submitted a Patient Consent-to-Disclose Form for any figure or video of the patient.

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