Unilateral Temporalis Muscle Hypertrophy: A Rare Cause of Swelling of the Temple

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Introduction

Isolated unilateral temporalis muscle hypertrophy (IUTMH) is an extremely rare cause of swelling in the temple, with only seven cases reported in the literature. We report the eighth case of this unique condition in a 17 year old white male, who presented initially with hemicranial headache.

Case Presentation

History

•17 year-old Caucasian male, with PMH of migraines and occasional tension headaches

•Chief complaint: 6 months of unilateral headache and swelling in the right side of the head

Evaluation

•Physical exam demonstrated a large, painful, readily palpable swelling in the right temporal region

•Neurologic exam and laboratory data were unremarkable

•Cranial MRI revealed a much enlarged right temporalis muscle with no abnormal contrast enhancement (Fig.1)

•Diagnosis of IUTMH confirmed with incisional biopsy, showing unremarkable skeletal muscle with mature adipose tissue

Treatment

•Patient was offered either Botulinum toxin type A (BtA) injections, or temporalis muscle reduction surgery - chose BtA injections

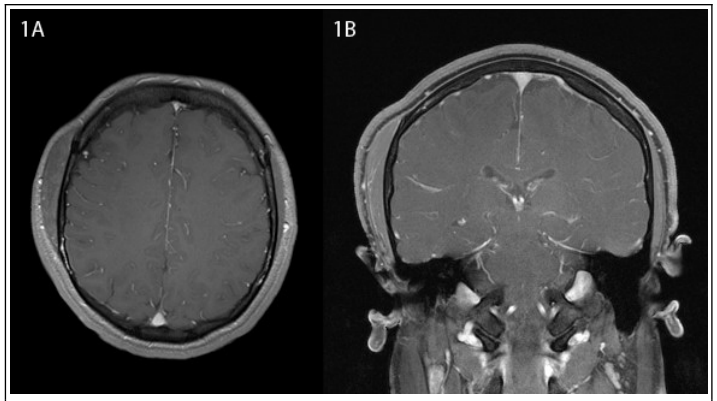

Fig.1 axial (1A) and coronal (1B) contrast cranial MRI demonstrating an enlarged nonenhancing right temporalis muscle

Conclusions

IUTMH is a particularly rare cause of swelling in the temple. We report the eighth case of IUTMH to our knowledge. MRI studies demonstrate normal muscle signal intensity without abnormal enhancement and laboratory tests are typically within normal limits.

While biopsy may not be necessary given these findings it should be considered if there is any question about the diagnosis. Treatment can involve clinical management through medicines, the application of splints, surgical intervention, or Botulinum toxin injections.

TABLE 1. A summary of reported IUTMH cases.

Case (Age, Sex)	Race	Presentation	Site	Onset time	Treatment
Wilson and Brown 1990 (43 y, F)	White	Painless swelling	R	11 mo.	Supportive
Serrat and Garcia-Cantera 1998 (15 y, F)	–	Swelling, temporalis muscle contraction, limitation of mouth opening	L	12 mo.	Surgery
Isaac 2000 (35 y, M)	White	Painless swelling	L	8 mo.	BtA
Lowry and Helling 2003 (45 y, M)	Black	Swelling, recurrent headaches	L	12 mo.	Supportive
Prantl et al 2005 (48 y, F)	–	Painless swelling	R	12 mo.	Surgery
Prantl et al 2005 (57 y, F)*	–	Swelling, temporalis muscle contraction, headaches	R	–	BtA
Rokadiya and Malden 2006 (33 y, F)	White	Painful swelling, headaches	L	3 mo.	Amitriptyline, splint
Vordenbäumen et al 2009 (22 y, F)	White	Painful swelling, recurrent headaches	R	6 mo.	Acetaminophen
Wang et al 2012 (17 y, M)	White	Painful swelling, recurrent headaches	R	6 mo.	BtA

* same patient who presented 9 years later

Table 1. A summary of reported IUTMH cases.

Discussion

•IUTMH is an exceedingly uncommon clinical occurrence without a clear etiology (only 8 reported cases including our case), when it was first reported in 1990 (Table 1)

•Average age at presentation from the literature (including our case): 35 years.

•More common in Caucasians, with a slight female preference and no right/left preponderance

•Masticatory muscle (temporalis, masseter, pterygoid muscles) hypertrophy is a rare clinical phenomenon, with the majority of clinical reports involving the masseter muscle (more bilaterally than unilaterally) - first reported case was in 1880

Differential Diagnosis

•Neoplastic processes (lipomatosis, liposarcoma, rhabdomyosarcoma, infiltrative leukemias and lymphomas)

•Inflammatory processes (idiopathic inflammatory myopathy, proliferative myositis)

Treatment Options

•Symptomatic treatment with analgesics

•BtA injections (simple and less invasive, muscle is only temporarily paralyzed, but high cost and repeated dosage sometimes needed)

•Muscle reduction surgery (more conventional treatment, but with potential side effects such as trismus, fibrosis, and decreased range of motion, but only needs to be done once if surgery is successful)