Isolated unilateral temporalis muscle hypertrophy

Case report

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Isolated unilateral temporalis muscle hypertrophy is an extremely rare cause of swelling in the temple region, with only 7 cases reported in the literature. The authors report the eighth case of this unique occurrence in a 17-year-old boy and review the current literature.

(https://thejns.org/doi/abs/10.3171/2013.1.PEDS12534)

KEY WORDS • unilateral temporalis muscle hypertrophy • botulinum toxin type A • temporal swelling

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Case Report

This 17-year-old Caucasian boy presented with the chief symptoms of hemicranial headache and swelling of the right side of his head for approximately 6 months' duration. He denied any head trauma, fever, or other complaints. He had a medical history of migraines as well as occasional tension headaches relieved by Excedrin or ibuprofen. Results of the physical and neurological examination were normal, with the exception of a large, painful, readily palpable swelling in the right temporal region.

Results of the patient’s laboratory evaluations were unremarkable. Cranial MRI studies revealed an enlarged asymmetrical prominence on the right temporalis muscle with no abnormal contrast enhancement or intracranial abnormalities (Fig. 1). An incisional biopsy demonstrated unremarkable skeletal muscle with mature adipose tissue, establishing the diagnosis of IUTMH. He was offered a variety of treatment options, including BtA injections or temporalis muscle reduction surgery. He has elected to undergo BtA injections at the Oral and Maxillary Facial Surgery Clinic.

Discussion

Masticatory (temporalis, masseter, pterygoid) muscle hypertrophy is a rare clinical phenomenon. The first reported case of masticatory muscle hypertrophy, albeit of a bilateral nature of the temporalis and masseter muscles, was described by Legg in 1880 in a 10-year-old girl.7 Numerous cases of bilateral masticatory muscle hypertrophy have been reported in the literature since then.4,5 The majority of clinical reports of masticatory muscle hypertrophy involve primarily the masseter rather than the temporalis muscle, and also this condition occurs bilaterally more often than unilaterally, with parafunctional jaw habits as one of the main causes.8,10,19

Abbreviations used in this paper: BtA = botulinum toxin type A; IUTMH = isolated unilateral temporalis muscle hypertrophy.
However, masticatory muscle hypertrophy can occur in a variety of combinations of hypertrophy affecting the temporalis, masseter, and medial pterygoid muscles.\textsuperscript{13,15} Whereas the first incidence of masticatory hypertrophy was reported in 1880, the first report of IUTMH was published more than a century later by Wilson and Brown in 1990.\textsuperscript{19} Previous case reports of temporalis muscle hypertrophy were always accompanied by associated clinical findings of massteric hypertrophy. To date, only 7 clinical cases of IUTMH have been reported (Table 1).

Masticatory muscle hypertrophy is primarily reported with bilateral masseteric muscle hypertrophy; therefore the exceedingly rare occurrence of isolated unilateral hypertrophy occurring in the temporalis muscle, as seen in our patient, cannot be overemphasized. Because there is no clear cause of IUTMH, patients in whom this phenomenon is suspected should be examined carefully for other pathological entities, particularly in a patient without prior medical history of bruxism or masticatory hypofunction as well as parafunctional jaw habits.\textsuperscript{8} Patients who have been successfully treated for temporalis muscle hypertrophy have experienced diminishing headaches and migraines, demonstrating a potential link between the onset of headache and IUTMH.\textsuperscript{14,15,18} The associative mechanism between headache and IUTMH, however, is not yet well understood. Although migraines are associated with a range of cranial and cervical muscle outcomes, primarily muscle tenderness,\textsuperscript{10} there is no reported association of migraines with IUTMH.

The differential diagnosis comprises mainly neoplastic processes or inflammatory processes, including benign muscle hypertrophy, lipomatosis, liposarcoma, idiopathic inflammatory myopathy, rhabdomyosarcoma, proliferative myositis, muscular dystrophy, and infiltrative leukemias and lymphomas.\textsuperscript{8} Rare cancers such as liposarcomas and rhabdomyosarcomas may present with swelling in the temporal region, and inflammatory processes such as proliferative myositis or idiopathic inflammatory myopathy may cause similar symptoms of headache in patients. The general clinical presentation of headache may also mask serious diseases such as leukemias and lymphomas, which can rarely involve the temporalis muscle, and would have to be diagnosed quickly for expeditious treatment.

After radiological examination, a definitive diagnosis can be arrived at via histopathological examination of biopsy samples to confirm IUTMH.\textsuperscript{3} Although IUTMH may certainly be suspected in patients with classic pre-

\begin{table}[h]
\centering
\begin{tabular}{|l|c|c|c|c|c|c|c|}
\hline
Authors & Year & Pt Age (yrs), Sex & Race & Presentation & Site & Onset Time (mos) & Treatment \\
\hline
Wilson & Brown, 1990 & 43, F & Caucasian & painless swelling & rt & 11 & supportive \\
Serrat et al., 1998 & 15, F & NR & swelling, temporalis muscle contraction, limitation of mouth opening & lt & 12 & symptomatic \\
Isaac, 2000 & 35, M & Caucasian & painless swelling & lt & 8 & BiA \\
Lowry & Helling, 2003 & 45, M & African American & swelling, recurrent headaches & lt & 12 & symptomatic \\
Prantl et al., 2005 & 48, F & NR & painless swelling & rt & 12 & surgery \\
Rokadiya & Malden, 2006 & 33, F & Caucasian & swelling, temporalis muscle contraction, headaches & rt & NR & BiA \\
Vordenbäumen et al., 2009 & 22, F & Caucasian & painless swelling, recurrent headaches & rt & 6 & acetaminophen \\
present study & 17, M & Caucasian & painless swelling, recurrent headaches & rt & 6 & BiA \\
\hline
\end{tabular}
\caption{Literature review of 8 reported cases of IUTMH\textsuperscript{*}}
\end{table}

\textsuperscript{*} NR = not reported; pt = patient.
\textsuperscript{†} The same patient, who presented 9 years after initial treatment.
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recomendation because it constitutes a low-risk procedure, and would affirm a precise diagnosis.

A variety of treatment options are available for IUTMH. These include symptomatic treatment with analgesics in those patients who have pain, muscle reduction surgery, or BtA injections.3,6,12,14,18

Treatment with BtA injections offers several benefits over surgical intervention—not only are the injections simple and less invasive than surgery, but surgery could potentially result in trismus, fibrosis, a decreased range of motion, and a decreased likelihood of reducing muscle hyperactivity. Additionally, BtA injections only temporarily paralyze the muscle, and increase the likelihood of decreasing the muscle’s tonic hyperactivity, thereby leading to atrophy of the muscle, improvement in the patient’s function, and a probable reduction in the frequency and severity of the patient’s associated headaches. However, the cost associated with BtA treatment remains high.1,9,11,17

To our knowledge, this is the eighth reported case of IUTMH, highlighting its exceedingly uncommon clinical occurrence. The average age at presentation was 35 years, and it occurred more commonly in Caucasians, with a slight female preference and no right/left preponderance (Table 1). We bring this case to the attention of the neurosurgical community because neurosurgeons are frequently consulted for swelling in the head region.

Conclusions

Isolated unilateral temporalis muscle hypertrophy is a particularly rare cause of swelling in the temple. Herein, we report the eighth case of IUTMH to our knowledge. The MRI studies demonstrate normal muscle signal intensity without abnormal enhancement, and results of laboratory tests are typically within normal limits. Although IUTMH may certainly be suspected in patients with classic presentation and imaging findings, a temporalis biopsy is recommended because it constitutes a low-risk procedure, and would affirm a precise diagnosis. Treatment can involve clinical management through analgesic medicines, the application of splints, surgical intervention, or BtA injections.

Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author contributions to the study and manuscript preparation include the following. Drafting the article: all authors. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Olivero.

References


Manuscript submitted August 15, 2012. Accepted January 7, 2013. Please include this information when citing this paper: published online February 8, 2013; DOI: 10.3171/2013.1.PEDS12534. Address correspondence to: William C. Olivero, M.D., Department of Neurological Surgery, University of Illinois College of Medicine at Urbana-Champaign, Carle Foundation Hospital, Urbana, Illinois 61801. email: olib@uic.edu.