CASE SERIES

Minotaur Syndrome (Masticatory Muscle Hypertrophy): Insights from a Case Series

Praveen K Sharma[1], Ajay Lucas[2], Jasvant Ram Ananthasayanam[2], Aashika Amir[2], and Sam Raja[3]

[1] Professor, Department of Radiology, Saveetha Medical College and Hospital, Saveetha Institute of Medical and Technical Sciences (SIMATS), Saveetha University, Chennai, Tamil Nadu - 602105, India
[2] Post-graduate Resident, Department of Radiology, Saveetha Medical College and Hospital, Saveetha Institute of Medical and Technical Sciences (SIMATS), Saveetha University, Chennai, Tamil Nadu - 602105, India
[3] Senior Resident, Department of Radiology, Saveetha Medical College and Hospital, Saveetha Institute of Medical and Technical Sciences (SIMATS), Saveetha University, Chennai, Tamil Nadu - 602105, India

ABSTRACT

Background: Minotaur Syndrome (MS, or Masticatory Muscle Hypertrophy) is a rare disease that affects mastication muscles. Temporalis and masseter muscle hypertrophy result in pseudo-masses or significant facial asymmetry. It occurs in both genders equally and is widely seen in anxious individuals with stressful lifestyles with bruxism, clenching, and chronic gum chewing. MS is usually asymptomatic. Symptoms include pain, mouth-opening limitations, and tension in the hypertrophied muscle region. Exostoses at the mandible angle give the face a "threatening" appearance, creating morpho-psychological conflict. Computed tomography (CT) and magnetic resonance imaging (MRI) are the prevailing imaging modalities employed to diagnose Minotaur syndrome, also known as masticatory muscle hypertrophy. The typical findings of MS on computed tomography (CT) and magnetic resonance imaging (MRI) are bilateral symmetrical or asymmetrical temporalis and masseter muscle hypertrophy. **Case series:** We report 5 cases of Minotaur syndrome in this article. **Conclusion:** Minotaur syndrome (or masticatory muscle hypertrophy) is a rare disease, diagnosed mainly by clinical and imaging findings, and it helps in the differential diagnosis of other conditions. Management of MS depends on who developed morpho-psychological conflict due to their facial appearance.

Malaysian Journal of Medicine and Health Sciences (2024) 20(SUPP13): 66-70. doi:10.47836/mjmhs.20.s13.14

Keywords: Bruxism, Masticatory Muscles, Hypertrophy, Facial Asymmetry, Computed Tomography, Magnetic Resonance Imaging

Corresponding Author:
Jasvant Ram Ananthasayanam, Postgraduate Resident
Email: jasvanttejas7@gmail.com
Tel : +91 9094774699

INTRODUCTION

The "Minotaur" is a mythical being described as a chimera-like creature featuring a bovine head and a human torso, as narrated by the Roman poet Ovid within the context of Greek mythology.

Minotaur syndrome is a relatively uncommon condition involving isolated or combined hypertrophy of all masticatory muscles that gives a threatening appearance to the face and creates morpho-psychological conflict in a gentle individual. The occurrence of idiopathic masseter muscle hypertrophy (IMMH) and idiopathic temporalis muscle hypertrophy (ITMH) was initially documented by Legg in 1880 in a case involving a 10-year-old female patient [1]. Isolated bilateral hypertrophy is caused by the masseter and temporalis muscles [2]. Benign masseteric hypertrophy is an infrequent pathological condition characterized by the unilateral or bilateral enlargement of the masseter muscle, leading to the formation of a pseudo-mass. An increase in the overall size of the muscle distinguishes the condition of unilateral or bilateral hypertrophy of the masseter muscle. Usually, asymptomatic differentials include odontogenic conditions, parotid gland pathology, and rare muscular tissue neoplasms—unilateral masseteric hypertrophy results in evident asymmetry of the mandibular region [3]. The condition known as masseter hypertrophy can lead to changes in facial contours, resulting in discomfort and undesirable esthetic effects [4]. Isolated unilateral temporalis muscle hypertrophy (IUTMH) was reported by Wilson and Brown in 1990 [5]. This condition does not appear to be an age preference [6].

CASE PRESENTATION

These cases were presented at Saveetha Medical College and Hospital. Case 1 was presented in the emergency department; Case 2 in the medicine outpatient department (OPD); Case 3 in the emergency department; Case 4 in the neurology department; and Case 5 in the medicine department. Written informed consent was obtained from all patients prior to the inclusion of their clinical data and imaging results in this publication. Additionally, ethical approval was secured from the ethical review board of Saveetha Medical College and Hospital to publish these case presentations.

Case presentation 1:A 20-year-old male presented to emergency department with a workplace head injury. CT Brain with 3D (SSD) shows bilateral temporalis muscles asymmetrical hypertrophy (right TMT = 8.8 mm < left TMT = 9.6 mm) and bilateral masseter muscles asymmetrical hypertrophy (right MMT = 21.8 mm > left MMT = 20.2 mm) and facial asymmetry, respectively. (Figure 1)

Case presentation 2:A 27-year-old male who came to medicine OPD presented with headaches for three months. CT Brain with (3D) SSD shows bilateral temporalis muscles symmetrical hypertrophy (right TMT = 9.4 mm & left TMT = 9.4 mm), bilateral masseter muscles asymmetrical hypertrophy (right MMT = 18.6 mm < left MMT = 18.9 mm) and facial asymmetry respectively. (Figure 2)

Case presentation 3:A 28-year-old male presented in emergency department with slip and fall complaints. CT Brain with (3D) SSD shows bilateral temporalis muscles

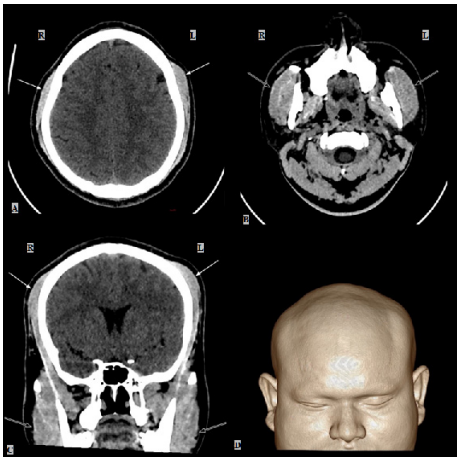

Figure 1: (A, B, C, D): A 20-year-old male presented with a workplace head injury. CT Brain (A, B - Axial, C - Coronal sections in brain window) shows bilateral temporalis muscles asymmetrical hypertrophy (R<L) (long white arrows) and bilateral masseter muscles asymmetrical hypertrophy (R>L) (long black arrows) at the levels of centrum semiovale, rami of the mandible and the frontal horns of the lateral ventricles, respectively and (D - 3D Coronal SSD) shows facial asymmetry.

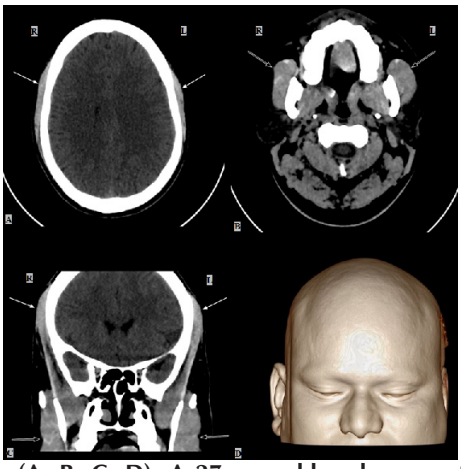

Figure 2: (A, B, C, D): A 27-year-old male presented with headaches for three months. CT Brain (A, B - Axial sections, C - Coronal section in brain window) shows bilateral temporalis muscles symmetrical hypertrophy (long white arrows), and bilateral masseter muscles asymmetrical hypertrophy (R<L) (long black arrows) at the levels of centrum semiovale, rami of the mandible and frontal horns of lateral ventricles respectively, and (D - 3D Coronal SSD) shows facial asymmetry.

Figure 3: (A, B, C, D): A 28-year-old male presented with slip and fall complaints. CT Brain (A, B - Axial sections, C - Coronal section in soft tissue window) shows bilateral temporalis muscles asymmetrical hypertrophy (R<L) (long white arrows) and bilateral masseter muscles asymmetrical hypertrophy (R<L) (long black arrows)at the levels of corona radiata, rami of the mandible and frontal horns of lateral ventricles respectively and (D - 3D Coronal SSD) shows facial asymmetry.

asymmetrical hypertrophy (right TMT = 13.3 mm < left TMT = 13.6 mm) and bilateral masseter muscles asymmetrical hypertrophy (right MMT = 21.1 mm < left MMT = 21.8 mm) and facial asymmetry respectively. (Figure 3)

Case presentation 4:A 49-years-old male came to neurology department presented with giddiness for one week. MRI Brain shows bilateral temporalis muscles asymmetrical hypertrophy (right TMT = 11.7 mm > left TMT = 10.4 mm) and bilateral masseter muscles asymmetrical hypertrophy (right MMT = 17.6 mm < left MMT = 19.3 mm). (Figure 4)

Case presentation 5:A 26-years-old male came to medicine department presented with an occipital headache for one year. CT Brain shows bilateral temporalis muscles asymmetrical hypertrophy (right TMT = 10.4 mm < left TMT = 10.6 mm) and bilateral masseter muscles asymmetrical hypertrophy (right MMT = 22.4 mm > left MMT = 22.1 mm) (Figure 5)

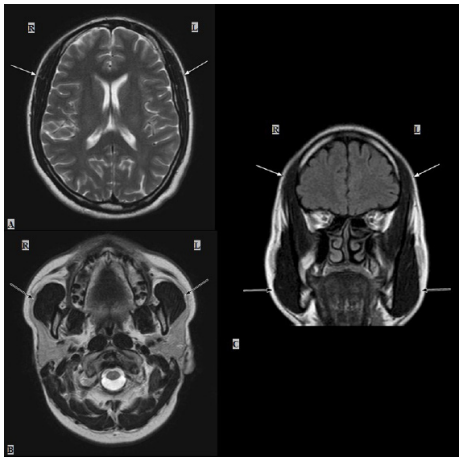

Figure 4: (A, B, C): A 49-years-old male presented with giddiness for one week. MRI Brain (A, B - T2W Axial, C - FLAIR Coronal) shows bilateral temporalis muscles asymmetrical hypertrophy (R>L) (long white arrows) and bilateral masseter muscles asymmetrical hypertrophy (R<L) (long black arrows) at the levels of corona radiata, rami of the mandible, and frontal lobes respectively.

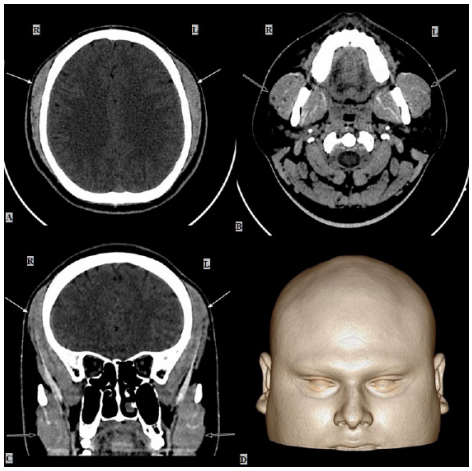

Figure 5: (A, B, C, D): A 26-years-old male presented with an occipital headache for one year. CT Brain (A, B - Axial sections, C - Coronal section in brain window) shows bilateral temporalis muscles asymmetrical hypertrophy (R<L) (long white arrows) and bilateral masseter muscles symmetrical hypertrophy (R>L) (long black arrows) at the levels of centrum semiovale, rami of the mandible and frontal lobes respectively and (D - 3D Coronal SSD) shows facial asymmetry.

DISCUSSION

Anatomy:

The masticatory muscles are the masseter, temporalis, medial, and lateral pterygoids. It is responsible for adduction and lateral motion.

The masseter muscle is considered the primary muscle responsible for mastication. The object in question has a rectangular shape and comprises three distinct layers, namely the superficial, middle, and deep layers. It is from zygomatic arch origin and to mandible ramus and angle insertion. The masseteric nerve, the mandibular (anterior) division of the trigeminal nerve, supplies the muscle. The primary function is to elevate the mandible and clench the teeth. Additionally, it serves to extend the angle of the mandible [7].

Located laterally on the head is a muscle called the temporalis. The origin of the temporal fossa on the parietal bone is located between the infratemporal crest and the inferior temporal line. The temporalis muscle is inserted into the mandible's coronoid process and ramus. The nerve supply of the temporalis muscle is provided by the deep temporal nerve and the trigeminal nerve's mandibular (anterior) division. Elevating and retracting the mandible is the primary purpose [8].

Etiology:

The common causes of Minotaur syndrome are psychological factors, particularly emotional stress, anxiety, personality disorders, bruxism, clenching, and gum chewing, with a theoretical explanation that is secondary to parafunctional jaw movements [9]. Other causes are dental malocclusion, bony prominences leading to trauma, and reactive hypertrophy [10], which occurs primarily in the young. However, inflammation, trauma, neoplasm, myopathy, and muscular dystrophy were excluded [11]. The histological examination of the affected muscle confirms a definitive diagnosis.

Clinical and imaging findings:

The most common clinical presentation of Minotaur syndrome is facial asymmetry. The condition often presents as a square-shaped face with a rectangular arrangement and is generally characterized by a lack of noticeable symptoms. But in certain situations, specific symptoms may also appear, including bruxism, protrusion, and trismus. The masseteric thickness is located anatomically in the inferior region of the mandibular ramus [12].

The prevailing imaging techniques employed for the diagnosis of minotaur syndrome (also known as masticatory muscle hypertrophy) are computed tomography (CT) and magnetic resonance imaging (MRI) [13,14]. Seltzer and Wang (1987) assert that CT and MRI scans are employed to diagnose diverse masseter muscle diseases. The characteristic findings of this syndrome, as observed using computed tomography (CT) and magnetic resonance imaging (MRI), are hypertrophy or enlargement of the temporalis and masseter muscles, which can occur unilaterally or bilaterally and exhibit either symmetrical or asymmetrical patterns. The median

thickness of the transverse metatarsal tunnel (TMT) on the right side was 4.22 mm, with a 2.4–7.7 mm range. On the left side, the median TMT thickness was 3.98 mm, with a 2.2–6.2 mm range. In the relaxation, the MMT was 11.3±1.2 mm in males and 9.8±1.3 mm in females [15].

Management:

The medical management of Minotaur syndrome (or masticatory muscle hypertrophy) involves botulinum toxin A injections [16,17]. Botulinum toxin, sometimes BTX or Botox, is a neurotoxic protein the bacterium produces. The anaerobic bacteria Clostridium botulinum produces a neurotoxin known as botulinum toxin A [17]. When injected into the muscle, this neurotoxin disrupts the neurotransmitter pathway, resulting in targeted paralysis and progressive muscular atrophy. Furthermore, there was a noticeable amelioration in symptoms related to the accompanying headache. The analgesics were used successfully to address the hypertrophy of the masseteric and temporalis muscles [18]. In the present study, surgical interventions were considered primarily for cosmetic and aesthetic factors in patients with Minotaur Syndrome (Masticatory Muscle Hypertrophy). The first surgical technique consisted of partial resectioning of the muscle and bulk through an extra-oral approach published in 1947 [18,19]. In 1959 the intraoral approach was described, and removing bone at the mandibular angle was difficult. The widespread utilization of the intraoral approach has been made possible by advancing surgical retractors explicitly designed for saw procedures [19]. The treatment of choice is the excision of the masseter muscle's internal layer, which reduces thickened bone in the mandibular angle region via an intraoral approach. The observed results consist of the presence of bone spurs located at the angle of the mandible. Guggenheim and Cohen reported the presence of bony spurs that develop due to periosteal irritation and subsequent deposition of new bone, which is a response to heightened muscular forces exerted by the bundles [19,20]. The surgical technique used in our cases involved partial resectioning of the muscle through an intraoral approach, was favored in our study due to its efficacy in reducing thickened bone in the mandibular angle region, and because it allows for direct access to the masseter muscle's internal layer, minimizing visible scarring. During the procedures, specific attention was paid to the presence of bony spurs at the angle of the mandible, which were observed in some cases. This was consistent with the findings reported by Guggenheim and Cohen, who described such spurs as resulting from periosteal irritation and subsequent new bone deposition in response to increased muscular forces. The surgical outcomes in our study were generally favourable, but several challenges were noted, including the risk of hematoma formation, transient facial nerve paralysis, and trismus. These potential complications were carefully managed by utilizing advanced surgical retractors and employing meticulous surgical techniques. Patients were informed of the risks associated with general anaesthesia, and postoperative care included monitoring for infection and other sequelae [20]. For the cases presented, specific post-operative suggestions included the use of botulinum toxin A injections to further reduce muscle hypertrophy and alleviate any residual symptoms. This was particularly recommended for patients with significant aesthetic concerns or those experiencing ongoing pain and discomfort.

Differential diagnosis:

The differential diagnosis of Minotaur syndrome (or masticatory muscle hypertrophy) includes:

-Salivary gland: benign and malignant neoplasms.
-Inflammation or infection: an abscess or cellulitis.
-Lymphatic system: metastatic lymph node, Lymphadenitis, lymphoma, lymphangioma.
-Connective tissue: lipoma, fibroma.
-Myopathy: Masseter tumor, myositis ossificans, periostitis.
-Vascular system: hemoma, A-V malformations, or pseudoaneurysm.

CONCLUSION

Minotaur syndrome (or masticatory muscle hypertrophy) is a rare disease, diagnosed mainly by clinical and imaging findings, and it helps in the differential diagnosis of other conditions—management in cases of those who developed morpho-psychological conflict due to their facial appearance.

Take home message

-Although rare, Minotaur syndrome is diagnosed mainly by clinical and imaging findings.
-Chronic bruxers are managed with psychiatric care and psychotherapeutic medications.
-A proper diagnosis of this condition avoids aggressive and unwarranted surgical therapy.

ACKNOWLEDGEMENT

I would like to express my sincere gratitude to the handloom weavers for taking part in the study and the department of community medicine in helping me to carry out this project.

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