



Rare Case of Limited Mouth Opening due to Sphenomandibular Ligament Calcification: Case Report and Literature Review

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Abstract

Objective: To report a case of a 7-year-old male with restricted mouth opening of sudden progression, and to discuss the diagnostic and therapeutic approach for this condition.

Materials and Methods: An initial orthopantomogram radiograph showed no signs of Temporomandibular Joint (TMJ) problems. Physiotherapy was attempted, but did not improve the case, and mouth opening construction increased by the time. Three-dimensional computed tomography scans revealed a linear radiopaque region along the sphenomandibular ligament. The patient underwent intraoperative resection of the calcified ligament by osteotome and mallet.

Results: The procedure resulted in an automatic improvement of the mouth opening.

Conclusion: The case highlights the importance of careful radiological evaluation of the oral cavity and TMJ in patients with restricted mouth opening and presents a successful therapeutic approach to the condition.

Keywords: Calcification; Limited mouth opening; Ossification; Sphenomandibular ligament; Trismus

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Received Date: 20 Feb 2023

Accepted Date: 09 Mar 2023

Published Date: 16 Mar 2023

Citation:

Alrhabi LM, Altawili ZM. Rare Case of Limited Mouth Opening due to Sphenomandibular Ligament Calcification: Case Report and Literature Review. *Am J Otolaryngol Head Neck Surg.* 2023; 6(2): 1228.

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Introduction

The Sphenomandibular ligament is a band of connective tissue that runs from the spine of the sphenoid bone to the lingula of the mandible [1]. Its function is to provide stability and support to the Temporomandibular Joint (TMJ) by limiting excessive movement of the mandible.

During jaw movement, the sphenomandibular ligaments work in conjunction with other ligaments and muscles to control the motion of the mandible. Specifically, it helps to prevent the mandible from moving too far forward or backward, which could lead to dislocation or damage to the joint. Additionally, it helps to maintain the alignment of the mandible with skull, ensuring that the teeth of the upper and lower jaw remain in proper occlusion [2].

While the sphenomandibular ligament is not the only structure that contributes to TMJ stability, it plays an important role in maintaining proper joint function. Dysfunction or damage to sphenomandibular ligament can lead to a variety of TMJ disorders, such as trismus [3].

Trismus which the mouth is unable to open fully. It effects on the patient's quality of life as well as complicate the medical procedures [4,5]. Trismus can make intubation more challenging [5]. Treatment of mouth locking depends on underlying cause, and can include physiotherapy, medication, and surgical intervention [6-8]. Early diagnosis and prompt treatment of trismus can help improve outcomes and minimize the impact on the patient's quality of life.

Case Presentation

A 7-year-old male presented to the maxillofacial department, General Military Hospital, Sana'a, Yemen with his parents who complained from progressive constriction of their child's mouth opening lasted for three years. The boy was not complained from pain. Parents reported gradual limitation of their child's mouth opening since he was four-year-old, and not mentioned any facial trauma. Parents declared admission to private dental clinic, where advised to conservatively manage



Figure 1: Patient unable to open the mouth.

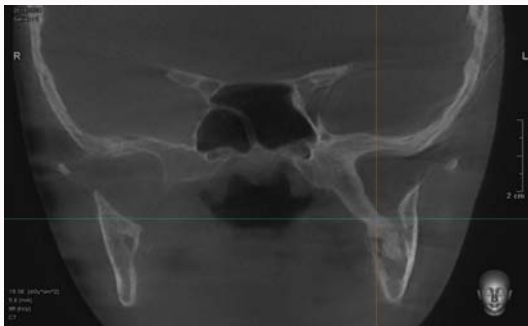


Figure 2: Coronal view of CT showing ossification of left sphenomandibular ligament.

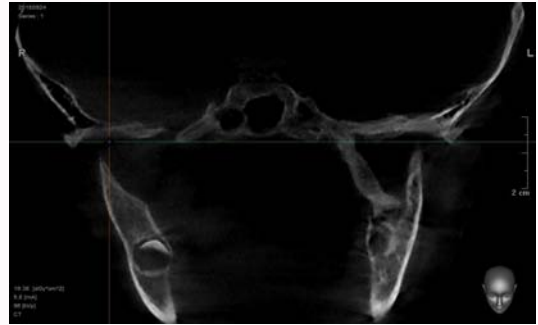


Figure 3: Coronal view of CT showing calcification extend through the whole body of left sphenomandibular ligament.



Figure 4: General anesthesia accomplished using blind nasal intubation through.



Figure 5: Immediate opening occur after calcified ligament excision.

with physiotherapy including use of wooden tongue depressor blade to increase opening by 1 mm. 'This treatment was not helpful to maximize mouth opening' as they mentioned. However, parents stated that by doing physical therapy the child complained from pain; therefore, they decided to terminate the treatment and after that the restriction was in progression.

After three-year of problem progression, child and his parents admitted to the local Maxillofacial Department of General Military Hospital, Sana'a, and Yemen. In clinical examination, edge-to-edge occlusion an inter-incisal distance of 1 mm (Figure 1), left lateral extrusion of 0 mm, and right lateral extrusion of 0 mm. Previous TMJ symptoms or relevant medical history were denied from patient's parents. A further referral to Maxillofacial Radiology Center for three-dimensional Computed Tomography (CT) scans revealed radiopacity of left Sphenomandibular ligament (Figure 2). This was managed by surgical intervention.

A diagnostic radiograph of three-dimensional Computed Tomography (CT) scan (Figure 2, 3) was initially reported to show linear radio-opaque within the left sphenomandibular ligament. General anesthesia accomplished using blind nasal intubation (Figure 4). A left triangular mucosal incision at molar area was planned through an intraoral approach. Elevation of mucoperiosteal flap using molt #9 periosteal elevator and detection of calcified ligament. By using osteotome and mallet, the calcified ligament dissected at the lingula. Immediately, the mouth opened with help of mouth gag (Figure 5). Dissected ossified ligament is shown in Figure 6. Flap closed by using 3.0 Vicryl interrupted sutures.

Parents instructed to keep their child under nothing be oral (NPO) until full anesthesia recovery. Then resume soft diet. For maintaining good oral hygiene, mouth washes and teeth brushing started day two after operation. Antibiotics and analgesics (Augmentin 312 mg syrup 10cc, Ibuprofen 200 syrup 10cc, and Dexamethasone 4 mg) were prescribed. Patient discharged on third day after operation.

The histopathological test confirmed that the radio-opacity was represent calcification affecting the majority of the Sphenomandibular ligament.

Discussion

Trismus a restricted mouth opening is rarely seen with children [9]. It can be caused by congenital or acquired disorders. Congenital



Figure 6: The excised part of calcified ligament.

causes include various syndromes and diseases, while acquired causes can be due to infection, iatrogenic, trauma, or oral care [10]. Trismus can present a challenge for intubation and have serious implications for feeding and breathing, particularly in newborns.

A very rare situation reported, trismus due to skull ligament ossification [11]. Sphenomandibular ligament ossification is a very rare cause of trismus, and its pathogenesis is not well understood. Sphenomandibular ligament is a thin, flat, fibrous band that limits the inferior movement of the mandible during mouth opening [12]. Sphenomandibular calcification is very rare, only one case reported in past 20 years combined by mastication muscle ossification [13]. Its detection through two-dimensional radiographs is difficult due to bony superimposition. CT or MRI scans are more effective at revealing soft tissue calcification [14].

Myositis ossificans is an uncommon disease characterized by the formation of mature bone on muscles or related soft tissues [15]. There are two forms of myositis ossificans: Munchmeyer's disease and # (MOT) [16-18]. Munchmeyer's disease may occur after trauma or develop in infancy as an autosomal dominant disease. MOT is localized and attributed to violent or repeated trauma. The pathogenesis of myositis ossificans is poorly understood, but metaplasia of connective tissue cells due to exposure of bone morphogenic proteins is a widely accepted theory [19].

In present case, diagnostic CT scans show faint linear calcification along the sphenomandibular ligament confirmed by the clinical finding of a dissected bony-like tissue then confirmed by histopathologic report. The initial confusion of the MRI and CT reports can be explained by the ligament's proximity to the lateral pterygoid and temporalis muscles, and that this condition is unfamiliar to surgeons and radiologists alike.

When patients with limited mouth opening undergo oral and maxillofacial surgery, several methods can be used for general anesthesia, including blind nasal intubation, retrograde intubation, and McCoy Laryngoscope [20-23]. In present case, general anesthesia was achieved through blind nasal intubation (Figure 5).

Surgical intervention is necessary for ossification of soft tissue ligaments. In the present case, physiotherapy did not improve the patient's condition, and gradual constriction progressed. Excision of the calcified ligament immediately resolved the problem.

Acknowledgement

The authors would like to express their gratitude to all the doctors and assistants whose valuable participation during the surgery contributed to the smooth execution of the procedure.

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