REVIEW ARTICLE

Pterygoid Hamulus Bursitis: The Diagnosis and the Relation of Orofacial Pain

Haitham Abdullah Bin Turayki[1], Faisal Suhaim[2], Saud Abdullah AlMoumen[2], Ali Montashery[2], Muslat A Bin Rubaiaan[2], Abdulrhman Mohammed Aldawsari[2]

1. *Haitham Abdullah Bin Turayki, Head, Consultant oral and maxillofacial surgery in KSMC, King Saud Medical City, Riyadh, Saudi Arabia*
2. *Faisal Suhaim, King Saud Medical City (KSMC), Riyadh, Saudi Arabia*
3. *Saud Abdullah AlMoumen, King Saud Medical City (KSMC), Riyadh, Saudi Arabia*
4. *Ali Montashery, King Saud Medical City (KSMC), Riyadh, Saudi Arabia*
5. *Muslat A Bin Rubaiaan, King Saud Medical City (KSMC), Riyadh, Saudi Arabia*
6. *Abdulrhman Mohammed Aldawsari, King Saud Medical City (KSMC), Riyadh, Saudi Arabia*

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ABSTRACT

The pterygoid hamulus (PH), a miniature yet significant structure of the sphenoid bone, has been linked to orofacial discomfort referred to as "pterygoid hamulus syndrome." Pterygoid hamulus syndrome, is an uncommon etiology of orofacial pain with multiple differential diagnosis. In this article we present a case report of a 47 -year-old female patient who reported to King Saud Medical City (KSMC) with chief complaint of pain on right side of the face for 2–3 years. She was diagnosed with "pterygoid hamulus syndrome" and surgically operated, the same being documented in this article. We have provided a literature appraisal regarding the incidence, diagnosis, and management of this condition. Given the distinct treatment approach required for this condition compared to other pain syndromes in the same area, clinicians should consider the possibility of a diagnosis of pterygoid hamular syndrome.

INTRODUCTION

Orofacial discomfort presents a diagnostic challenge due to its diverse sources. Historically, various authors have noted that irritation in the palate associated with the pterygoid hamulus (PH) might cause referred symptoms in the head-and-neck region. (1,2) The pterygoid hamulus (PH), a miniature yet significant structure of the sphenoid bone, has been linked to orofacial discomfort referred to as "pterygoid hamulus syndrome." This term describes pain or discomfort in the palate and pharyngeal area resulting from inflammation of the hamular region caused by bursitis or an enlarged hamulus. Shankland (1996), Salins and Bloxham (1989) described the inflammation of the bursa covering the tensor veli palatini tendon as hamular bursitis.(3,4) Frequently, it is inappropriately identified as temporomandibular disorder (TMD), impacted third molars, trigeminal and glossopharyngeal neuralgia, anomalies of the styloid process and its accompanying ligaments, tumours, cysts, and infection of the middle ear.(5) By means of this article, our objective is to underscore the significance of PH syndrome in the differential diagnosis of orofacial pain and discomfort with an unknown aetiology.

CASE REPORT

A 47 -year-old female patient reported to King Saud Medical City (KSMC) with chief complaint of pain on right side of the face for 2–3 years. She is medically and allergic free and she has a history of nasal reduction after trauma with no nasal obstruction.The pain was severe, radiating to the ipsilateral ear and temporal region. The pain was aggravated on swallowing and relieved on medications. The patient was referred to the oral and maxillofacial surgery department by the ear, nose and throat (ENT) department after multiple attempts to identify the source of pain. They performed computed tomography(CT) of the paran-

-asal sinus with fine slice non-contrast CT images obtained through the paranasal sinuses. Their finding was bilateral mild mucosal wall thickening of the maxillary, sphenoid, ethmoid and frontal sinuses. There was bilateral blockage of the hiatus semilunaris attributed to the wall thickening noted in the maxillary sinuses. Part of the bilateral inferior nasal turbinates are not visualized, most likely related to previous surgery (turbinectomy). The nasal septum was central with intact bony outlines of the paranasal sinuses. No suspicious bone lesion was found. No abnormalities were seen in orbits, orbital cavities and visualized parts of the brain. Tiny calcification was noted in the right palatine tonsil, likely representing tonsillolith. The patient had visited multiple clinicians and had undergone various treatments for the same pain. The diagnosis was changed from temporomandibular joint (TMJ) disorder to pharyngitis to neuralgia.

The patient was intermittentently on pain relief medications such as benzodiazepam and was earlier diagnosed with myofascial pain dysfunction syndrome. The patient's medical, dental, and family history was unremarkable.

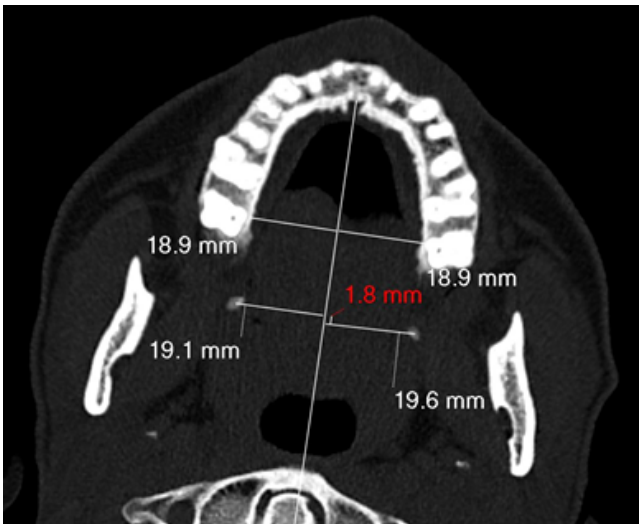
Extraoral examination revealed no abnormalities. Bilateral TMJ examination was normal, with no prominent signs and symptoms of TMJ disorder. Intraoral examination showed focal areas of slight bony enlargement with typical blanching palatal to the maxillary tuberosity on the right side. The swelling was hard on palpation, suggesting bony enlargement, with typical blanching erythema surrounding the swelling. Marked tenderness could be elicited on slight provocation in the same area. The differential diagnosis included elongated pterygoid hamular process, glossopharyngeal neuralgia, and idiopathic orofacial pain.

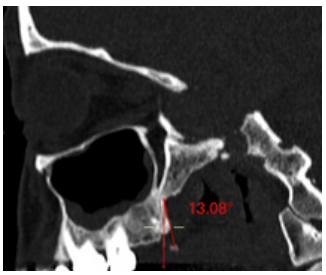
Further evaluation with cone-beam computed tomography (CT) was performed for precise location, and measurements in both axial and coronal sections were advised, which demonstrated a

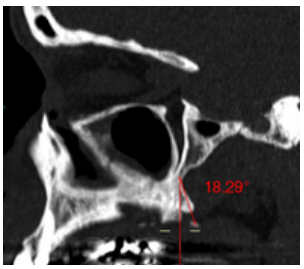
prominent right pterygoid hamulus. Both radiographic and clinical findings led to the final diagnosis of PH syndrome.

Surgical resection of the elongated hamuli was planned under general anaesthesia in this case. Intraorally, a longitudinal incision over the bony prominence, posteromedial to the maxillary tuberosity, was made. Dissection was done to isolate and expose the elongated hamulus on the right side, which was located along the palatal mucogingival line.
The hamulus was excised with rongeur forceps, and bony margins were smoothened by rounded bur. Primary closure was obtained. Postoperative healing was uneventful. The patient reported relief of symptoms after that and were kept on regular follow-ups with no recurrence until now.

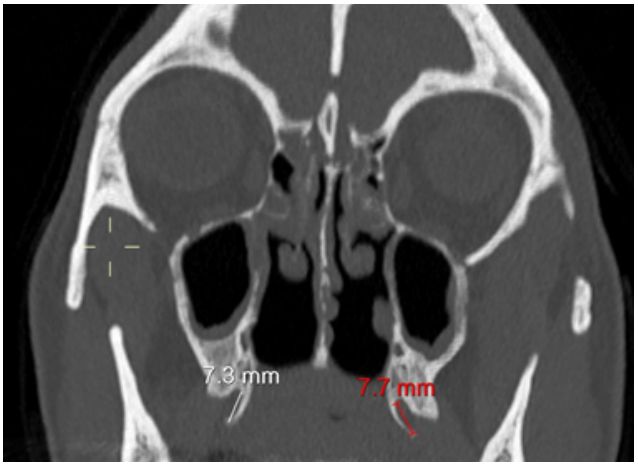
Radiological perspective:

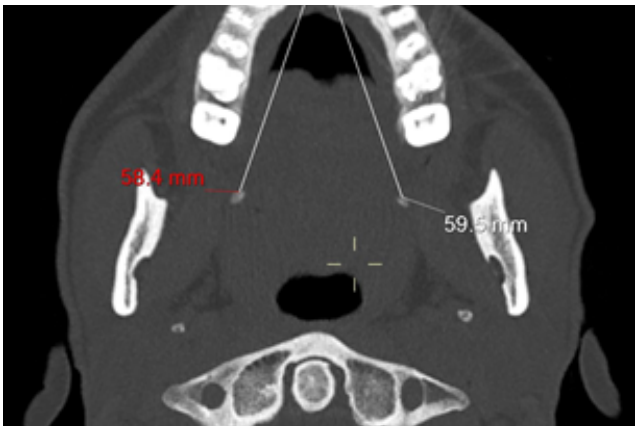






We used an imaginary line as a reference line, which was drowned from the midline of the incisive foramen perpendicular toward the vertebra, to locate the area and the angulation of the tip of the Hamulus. We named that line as Bin Turayki's line.





The angle of the pterygoid hamulus can play a significant role in the development of hamular bursitis. The orientation and angulation of the hamulus can affect how the tendon of the tensor veli palatini muscle interacts with the hamulus, potentially leading to increased friction or pressure. This can cause inflammation of the bursa, resulting in hamular bursitis.
Factors Influencing Bursitis
Increased Friction: A sharper angle may increase the contact and friction between the tendon and the hamulus.
Mechanical Stress: Certain angles may cause abnormal mechanical stress on the surrounding tissues during activities such as swallowing or speaking.
Anatomical Variations: Variations in the angle and length of the hamulus can contribute to an individual's susceptibility to bursitis.

DIAGNOSIS AND TREATMENT

Diagnosing the impact of the hamulus angle on bursitis typically involves:
Clinical Examination: Assessing symptoms and conducting a physical examination.
Imaging Studies: CT scans or MRIs to evaluate the structure, length, and angle of the pterygoid hamulus and surrounding tissues.
Treatment options, if the angle is a contributing factor, may include:
Non-surgical Approaches: Rest, anti-inflammatory medications, and physical therapy to manage symptoms.
Surgical Intervention: In severe cases, surgical correction of the hamulus angle or removal of the bursa may be considered.
Consulting with a specialist in maxillofacial surgery or an otolaryngologist is essential for a proper diagnosis and tailored treatment plan.
Review of Literature:
Within the context of literature, numerous anatomic and radiographic investigations have been conducted to determine the location and angulation of the pterygoid hamulus.(6-8) Anatomical implications of this structure in relation to age and function have been investigated.(9) Certain authors have also associated the length of the pterygoid hamulus with obstructive sleep apnea.(10) There are a limited number of instances of pterygoid hamulus syndrome documented in the literature. A comprehensive exploration was conducted in the PubMed/Medline database utilizing MeSH terms like "Pterygoid Hamulus," "Pterygoid Hamulus Syndrome," and "Hamular Bursitis," using a range of Boolean operators such as "AND" and "OR." As far as the author is aware, a total of 32 instances of this phenomenon, including the current occurrences, have been identified so far (Table 1).

Authors	Years	Diagnosis	Number of cases	Age/sex	Management
Gores(11)	1964	Elongated hamulus (edentulous)	2	-	Surgical resection
Hertz(1)	1968	Elongated hamulus (edentulous)	1	-/female	Surgical resection
Wooten et al(2)	1970	Elongated hamulus presenting as an asymptomatic mass	3	19/male 20/male 5/male	Surgical exposure, no resection. Reassurance
Panzoni and Clauser(12)	1978	Pterygoid hamular syndrome	1	-	-
Charbeneau and Blanton(13)	1981	Pterygoid hamular syndrome	1	22/male	Patient education
Hjørting-Hansen and Lous(14,15)	1987a.b	Coined the term “pterygoid hamulus syndrome”	2	-	-
Salins and Bloxham(3)	1989	Hamular bursitis (no enlargement of pterygoid hamulus)	1	Fifth decade/female	1 ml of dexamethasone (decadron) 4 mg/ml
Kronman et al.(16)	1991	Hamular bursitis (with osteophyte)	1	60/female	Excision of fibrous tissue with removal of osteophyte
Eyrich et al.(17)	1997	Pterygoid hamular syndrome	1	25/male	Surgical resection
Dias(18)	1997	Hamular bursitis	1	-	-
Sasaki et al.(19)	2001	Pterygoid hamular syndrome	1	47/male	Surgical resection
Fu et al.(20)	2004	Pterygoid hamulus syndrome	9	-	Surgical resection
Ramirez et al.(5)	2006	Hamular bursitis	2	43/female 52/female	Infiltration of 1 ml of synthetic cortisone; NSAIDs; soft diet
Dupont and Brown(21)	2007	TMD with pterygoid hamular pain	92	-	No treatment
Cho et al.(22)	2013	Pterygoid osteophyte	1	62/female	Surgical resection
Roode and Bütow(23)	2014	Pterygoid hamulus syndrome	1	50/male	Surgical resection

Bandini et al.(24)	2017	Hamulus hypertrophia	1	36/female	Conservative, followed by surgical
Kende, et al. (25)	2019	Pterygoid hamulus syndrome	2	45/male 23/male	Surgical resection
KSMC case	2023	/////	1	00/female	Surgical resection

Table 1

The age range in our analysis varied from 5 to 62 years. There was an overrepresentation of males seen. All cases that were documented exhibited symptoms, with the exception of the two cases detailed by Wooten et al., where an extended hamulus was observed as an asymptomatic mass in the soft palate area. Conservative care was employed in the previously documented instances; yet, the majority of patients underwent surgical excision. Comparable methodology was employed in our instances due to the extended hamulus. There is also a discrepancy in the terminology used to refer to this condition, with variations such as "Pterygoid Hamular Syndrome," "Hamular Bursitis," and "Hamulus Hypertrophia."

DISCUSSION

The pterygoid hamulus may be responsible for unconventional pain in the oral cavity and pharynx. Its proximity to the upper dental arch and pharynx makes it a subject of interest for all disciplines dealing with this area. There have been multiple efforts to clarify the mechanism of hamular pain, although the exact etiology remains unknown. Diligent examination of this area enabled us to identify and address the vague and contradictory symptoms associated with extension of the pterygoid hamulus in the oral cavity. Given the infrequency of the conditions, excluding it from the diagnosis is more of a standard practice than an exception. This results in a delay of treatment. In this case, the patient endured a period of 2–3 years before receiving a definitive diagnosis. Frequently, it was erroneously identified as Eagle's syndrome, temporomandibular disorders (TMDs), geniculate ganglion neuralgia, glossopharyngeal neuralgia, cysts and tumors, otitis media, foreign substances, burning mouth syndrome, and impacted third molars.

In our case, the diagnosis was established relying on clinical and radiological findings. Typical clinical manifestations often observed include palatal pain with one side being more erythematous than the other, firm swelling or enlargement, and redness of the palatal mucosa over the hamulus, intense localized pain in the hamular region, ear pain, and difficulty and pain while swallowing. In addition to these clinical characteristics, conventional radiographic techniques including lateral cephalometric, submentovertex, and tomography, as well as sophisticated imaging methods such as axial and coronal plane computed tomography scans with three-dimensional perspectives, can be employed.

The response to nonsteroidal anti-inflammatory medications and a detailed history of symptoms helped us in excluding neuralgias. The lack of any additional external muscle or temporomandibular joint (TMJ) observations has eliminated the possibility of TMJ diseases. Nevertheless, Dupont and Brown discovered positive findings for hamular pain in a study involving 493 patients with temporomandibular disorders. (21) Certain authors have proposed the administration of local anesthetic in the area as an excellent diagnostic aid. In our instances, the existence of a sensitive red bone protrusion eliminated the necessity for local anesthetic injection, yet examination was done by using the recommended above mentioned diagnostic aid. Moreover, the simple existence of a raised soft-tissue region is insufficient for diagnosis, as researchers have previously identified asymptomatic elongated hamulus as a swelling in this area. (2) Authors have proposed numerous etiologies of pterygoid hamulus syndrome. Sasaki et al. proposed that the anomalous pterygoid hamulus is responsible for for mechanical stimulation to the surrounding tissues, consequently disrupting the typical function of the tensor veli palatini muscle, which can further be leading to bursitis. (19) In addition, these events might also stimulate the lesser and greater palatine nerve, glossopharyngeal nerve, and facial nerve, which may cause symptoms resembling neuralgia.

Furthermore, dysfunction of the tensor veli palatini muscle may also result in symptoms in the meatus, a phenomenon observed in case of our patient. Ramirez et al. proposed that the mentioned expression emphasizes the shared neuronal connectivity between the oro-masticatory and the otic systems. (5) In their two case reports, they suggested a bio-psychosocial perspective, which detailed associated TMD symptoms and the treatment provided for both.

The location, dimensions, and medio-lateral inclination of the hamulus are important for the proper functioning of various muscles, including the tensor veli palatini, palatopharyngeus, and pharyngeal constrictors.(24) Due to the presence of sophisticated imaging techniques like CBCT, it has been possible to determine the dimension and inclination. This not only aids in diagnosis but is also of great utility in treatment. In our study, the measurements of the pterygoid hamulus fell within the range of 0.0–0.0 mm, with the orientations of the PHs being laterally inclined in the coronal plane. The mean measurement of the left hamulus was 5.0 mm and that of the right was 4.9 mm according to measurements made by Eyrich et al. in normal individuals, however in the CBCT investigation conducted by Orhan et al., it was recorded as 5.40 mm and 5.48 mm on the right and left sides, respectively.(17, 10) The elongation of the hamulus is not the sole causative cause in this syndrom. Charbeneau and Blanton suggest that alternative relationships may be present, such as: (1) the medial pterygoid plate and its hamular process perhaps being positioned more inferiorly in relation to the palate; or (2) the soft palate mucosa possibly being thinner.(13) However, the bilateral presence serves as compelling proof of the anatomical entity being the cause in one of our patient.

Treatment options may include pharmaceutical interventions, less invasive procedures like injections, or invasive measures such as surgical excision. For traditional minimally invasive therapy, it has been recommended to administer 1 ml of dexamethasone 4 mg per ml to reduce inflammation. Furthermore, anti-inflammatory drugs such ibuprofen, at a dosage of 600 mg to 800 mg, every 6 hours, should be recommended. Patients should undergo periodic reevaluation If there is improvement of symptoms, injections

should be repeated and the anti-inflammatory medicine ought to be extended and maintained, with periodic follow-ups.
If conservative therapy is ineffective, or if the length of the pterygoid hamulus exceeds the recognized normal limits, surgical intervention should be considered. If osteophytes, a prominent hamular process, or bursa fibrosis are detected, surgical intervention should be followed. If there are no inflammatory or fibrotic changes in the bursa, as is the situation in our case, a hamulotomy should be performed, or the hamulus should be excised, using a Rongeur's forceps. An identified complication of hamulotomy is the disruption of the tensor veli palatini function over the osseous hook, leading to Eustachian tube dysfunction expressed as hypoacusis, as well as the inability to form a proper seal between the palate and throat during speech and swallowing. Table 2 illustrates a clinical manual detailing the symptoms, differential diagnosis, and treatment options for this condition (25).

Table 2: Clinical guide for the diagnosis and management of pterygoid hamulus syndrome

	Features
Etiology	Injury, infection, or a preexisting condition. Anesthesia intubations, swallowing a big bolus, yawning, sustained intraoral auscultation, overextended maxillary prosthesis, the traumatic strike during toothbrushing, bulimic patients, "fellatio" in child sexual abuse.
Clinical features	Palatal pain with the offending side more erythematous than the opposite side. Swelling of the palatal mucosa over the Hamulus. Sharp localized pain in the hamular region. Elongated hamuli will be evident as a firm swelling or enlargement. Ear pain. Difficulty and pain with swallowing.
Diagnostic criteria	Reported history, clinical findings, and diagnostic anesthetic infiltration into the hamular region.
Differential diagnosis	Eagle's syndrome, TMDs, geniculate ganglion neuralgia, glossopharyngeal neuralgia, cyst and tumors, otitis media, foreign bodies, burning mouth syndrome, impacted third molars.

Investigations	Conventional radiographic imaging: The lateral cephalometric, submentovertex, and tomography. Advanced imaging: CT scan in axial and coronal planes with 3D views. CBCT should be preferred over a CT image
Management	Conservative treatment. The source of trauma or irritation should be removed. The infiltration of 1 ml of dexamethasone 4 mg per ml. Anti-inflammatory medications such as ibuprofen, 600 mg–800 mg, every 6 h. If conservative treatment proves unsuccessful, surgical management should be considered

CT: Computed tomography; 3D: Three dimensional; CBCT: Cone-beam CT, TMDs: Temporomandibular disorders

CONCLUSION

Pterygoid hamulus syndrome, an uncommon etiology of orofacial pain, was documented in this article. Within this article, we have provided a literature appraisal regarding the incidence, diagnosis, and management of this condition. The palate and pharyngeal regions necessitate specific clinical monitoring for differentiating orofacial pain. Given the distinct treatment approach required for this condition compared to other pain syndromes in the same area, clinicians should consider the possibility of a diagnosis of pterygoid hamular syndrome.

Declaration of patient consent:

The authors affirm that they have acquired all requisite patient permission paperwork. Within the document, the patients have provided their agreement for the publication of their photos and other clinical data in the journal. The patients comprehend that their names and initials will not be disclosed, and diligent measures will be taken to obscure their identification; yet, complete anonymity cannot be assured.

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