

The Pterygoid Hamulus Syndrome – An Important Differential in Orofacial Pain

Abstract

The purpose of this review was to document cases of pterygoid hamulus (PH) syndrome and to describe the various etiology, differential diagnosis, and management strategies so far reported in literature. Here, we also present two case reports of PH syndrome. A comprehensive search in PubMed/Medline database was done using MeSH terms such as “Pterygoid Hamulus,” “Pterygoid Hamulus Syndrome,” and “Hamular Bursitis” using various Boolean operators such as “AND” and “OR”. Till date, 31 cases of this entity including the present cases have been found. Conservative management was followed in the earlier reported cases; however, most cases were treated by surgical resection.

Keywords: Bursitis, facial, hamular, pain, syndrome

Introduction

Orofacial pain is a diagnostic challenge because of its varied origins. In the past, different authors have observed that irritation in the palate corresponding to the pterygoid hamulus (PH) has referred symptoms in the head-and-neck area.^[1,2] The PH, a small but significant process of the sphenoid bone, has been associated with orofacial pain that is termed as “pterygoid hamulus syndrome.” The term is used to describe pain in the palatal and pharyngeal region that is due to inflammation of the hamular region on account of bursitis or an elongated hamulus. Shankland in 1996 and Salins and Bloxham in 1989 explained the inflammation of the bursa covering tensor veli palatini tendon as hamular bursitis.^[3,4]

More often than not, it is misdiagnosed as temporomandibular disorder (TMD), impacted third molars, trigeminal and glossopharyngeal neuralgia, abnormalities of the styloid process and its associated ligaments, tumors, cysts, and infection of the middle ear.^[5] Through this review article and case series, we aim to highlight the importance of PH syndrome in the differential diagnosis of orofacial pain of unknown origin.

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Case Reports

Case 1

A 45-year-old male patient reported with the chief complaint of pain on swallowing for 2 years.

The pain was insidious in onset, progressed gradually from mild to severe over 2 years. The pain was typically located in the palatal and pharyngeal area. The patient had visited multiple clinicians and had undergone various treatments for the same. The diagnosis was changed from temporomandibular joint (TMJ) disorder to pharyngitis to neuralgia. The patient's medical, dental, and family history was unremarkable. On extraoral examination, no tender areas could be localized. Bilateral TMJ examination was normal. Intraoral examination showed bilateral focal areas of swelling palatal to the maxillary tuberosity [Figure 1]. The swelling was hard on palpation suggestive of bony enlargement, with typical blanching erythema seen surrounding the swelling. Marked tenderness could be elicited on slight provocation in the same areas.

The differential diagnosis included elongated pterygoid hamular process, glossopharyngeal neuralgia, and idiopathic orofacial pain.

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Radiographic examination with cone-beam computed tomography (CBCT) was performed. CBCT revealed bilateral enlarged hamuli with a lateral inclination. Precise location and measurements were obtained [Figure 2].

All the features seen on radiographs with clinical correlation led to the final diagnosis of PH syndrome.

Case 2

A 23-year-old male patient reported with the chief complaint of pain on the left side of the face for 2–3 years.

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 on intermittent pain relief medications 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 left side. 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.

Radiographic investigations with intraoral periapical radiograph were advised, which demonstrated a prominent left PH [Figure 3]. Further evaluation with CBCT was performed for precise location and measurements in both axial and coronal sections. Both radiographic and clinical findings led to the final diagnosis of PH syndrome.

Surgical resection of the elongated hamuli was planned under local anesthesia in both cases. 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 left side, which was located along the palatal mucogingival line [Figure 4]. The hamulus was excised with a bur, and bony margins were smoothed. Primary closure was obtained. Postoperative healing was uneventful. The patients reported with relief of symptoms thereafter and were kept on regular follow-up up to 2 years with no recurrence [Figure 5].

Review of Literature

In literature, there have been various anatomic and radiographic studies to locate the spatial position and angulation of PH.^[6-8] Anatomic implications of this structure with respect to age and function have been studied.^[9] Some authors also correlated PH length to obstructive sleep apnea.^[10] There are very few cases of PH syndrome reported in literature. A comprehensive search in PubMed/Medline database was done using MeSH terms such as “Pterygoid



Figure 1: Clinical photograph showing bilateral swellings in the hamular region

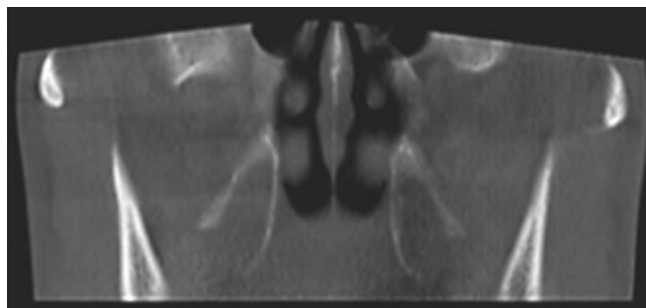


Figure 2: Coronal section of cone-beam computed tomography showing elongated hamuli with a lateral inclination

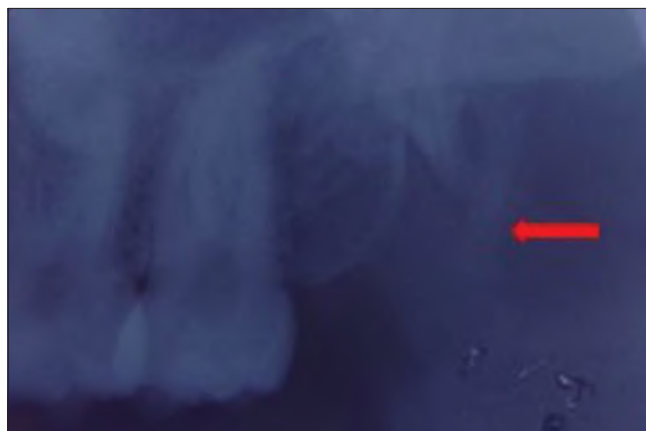


Figure 3: Intraoral periapical radiograph showing an elongated hamulus

Hamulus,” “Pterygoid Hamulus Syndrome,” and “Hamular Bursitis” using various Boolean operators such as “AND” and “OR”. To the author’s knowledge, till date, 31 cases of this entity including the present cases have been found [Table 1].^[1-5,11-24] The age group in our review ranged from 5 to 62 years. Male preponderance was seen. All cases reported were symptomatic except the two cases reported by Wooten *et al.* who showed elongated hamulus as an asymptomatic mass in the soft palate region. Conservative

Table 1: Literature review

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 <i>et al.</i> ^[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
Hjørtting-Hansen and Lous ^[14,15]	1987 ^{a,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 <i>et al.</i> ^[16]	1991	Hamular bursitis (with osteophyte)	1	60/female	Excision of fibrous tissue with removal of osteophyte
Eyrich <i>et al.</i> ^[17]	1997	Pterygoid hamular syndrome	1	25/male	Surgical resection
Dias ^[18]	1997	Hamular bursitis	1	-	-
Sasaki <i>et al.</i> ^[19]	2001	Pterygoid hamular syndrome	1	47/male	Surgical resection
Fu <i>et al.</i> ^[20]	2004	Pterygoid hamulus syndrome	9	-	Surgical resection
Ramirez <i>et al.</i> ^[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 <i>et al.</i> ^[22]	2013	Pterygoid osteophyte	1	62/female	Surgical resection
Roode and Bütow ^[23]	2014	Pterygoid hamulus syndrome	1	50/male	Surgical resection
Bandini <i>et al.</i> ^[24]	2017	Hamulus hypertrophy	1	36/female	Conservative, followed by surgical
Present case		Pterygoid hamulus syndrome	2	45/male 23/male	Surgical resection

NSAIDs: Nonsteroidal anti-inflammatory drugs; TMD: Temporomandibular disorder

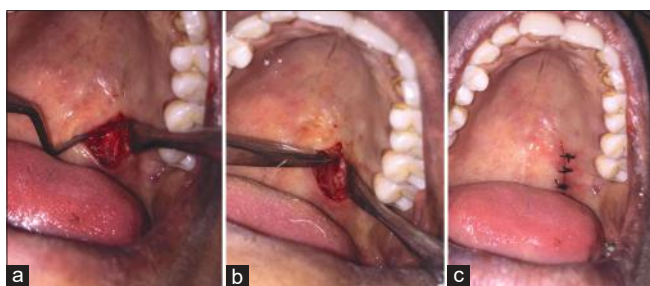


Figure 4: (a) Intraoperative photograph showing the exposed hamulus. (b) Intraoperative photograph showing the excised hamulus. (c) Intraoperative photograph after primary closure

management was followed in the earlier reported cases; however, most cases were treated by surgical resection. Similar technique was followed in our cases owing to the elongated hamulus. There is also a variation in terminologies of the entity such as “Pterygoid Hamular Syndrome,” Hamular Bursitis,” and “Hamulus Hypertrophy.”

Discussion

The PH can be responsible for atypical pain in the oral cavity and pharynx. Its close topographical relationship to



Figure 5: Intraoral post operative photograph showing good healing

the upper dental arch and pharynx renders it of interest to all specialties that are involved with this region. There have been various efforts to describe the mechanism of hamular pain, but the precise etiology is not known.

Careful assessment of this region helped us to trace and manage the ambiguous and conflicting symptoms related

to PH elongation in the oral cavity. Due to the rarity of the disease, its exclusion from the diagnosis becomes a rule rather than the exception. This leads to delay in treatment. In both the cases, the patients suffered for 2–3 years before reaching a final diagnosis. Often, they are misdiagnosed as Eagle's syndrome, TMDs, geniculate ganglion neuralgia, glossopharyngeal neuralgia, cyst and tumors, otitis media, foreign bodies, burning mouth syndrome, and impacted third molars. In our cases, the diagnosis was made based on clinical and radiological findings. Clinical features commonly seen include palatal pain with the offending side more erythematous than the opposite side, firm swelling or enlargement and redness of the palatal mucosa over the hamulus, sharp localized pain in the hamular region, ear pain, difficulty and pain with swallowing. Along with these clinical features, conventional radiographic imaging such as lateral cephalometric, submentovertex, and tomography and advanced imaging including computed tomography scan in axial and coronal planes with three-dimensional views can be used.

Responsiveness to nonsteroidal anti-inflammatory drugs and a long history of symptoms helped us in ruling out neuralgias. Absence of any extraoral muscular or TMJ findings ruled out TMJ disorders. However, Dupont and Brown found positive findings of hamular pain in a study of 493 TMD patients.^[21]

Some authors have suggested infiltration of local anesthesia in the region as an excellent diagnostic aid.^[8] In our cases, the presence of a tender erythematous bony prominence excluded the need for local anesthetic infiltration. In addition, the mere presence of an elevated soft-tissue area is not enough for diagnosis as authors have previously found asymptomatic elongated hamulus as a swelling in this region.^[2]

There have been various pathogeneses of PH syndrome postulated by authors. Sasaki *et al.* suggested that the abnormal PH is the cause of mechanical invigoration to the adjacent tissues, thus interrupting the normal action of the tensor veli palatini muscle, which can further cause bursitis.^[19] Furthermore, these events also invigorate the lesser and greater palatine nerve, glossopharyngeal nerve, and facial nerve, which may mimic neuralgia-like symptoms. In addition, tensor veli palatini dysfunction may also lead to symptoms in the meatus, which was seen in both our cases.

Ramirez *et al.* suggested that such referred expression focuses on the common neural interconnection between the oro-masticatory and the otic system.^[5] They also suggested a bio-psychosocial aspect in their two case reports which reported with concomitant TMD symptoms and were managed for both.

The position, length, and medio-lateral inclination of the hamulus are of concern for the adequate function of several muscles such as tensor veli palatini, palatopharyngeus,

and pharyngeal constrictors.^[24] Due to availability of the advanced imaging modalities such as CBCT, measurements of the length and inclination have become possible. This not only assists in diagnosis, but is also of immense use in treatment. The length of the PH in our cases was found to be within the range of 4.9–7.2 mm, with the positions of PHs inclined laterally in the coronal plane. The average length of the left hamulus was 5.0 mm and the that of the right was 4.9 mm as found by Eyrich *et al.* in normal individuals, whereas in the CBCT study by Orhan *et al.*, it was found to be 5.40 and 5.48 mm on the right and left sides, respectively.^[17,10] Excessive length of the hamulus is not the only etiological factor in this syndrome. According to Charbeneau and Blanton, other relationships can exist, as follows: (1) the medial pterygoid plate and therefore its hamular process, may have been located more inferiorly than usual with respect to the palate; or (2) the mucosa of the soft palate, could be thinner.^[13] However, bilateral occurrence is a strong evidence of anatomic entity as the etiology in one of our cases.

Treatment may be medicinal, minimally invasive including injections, or invasive such as surgical resection. For conventional minimally invasive treatment, infiltration of 1 ml of dexamethasone 4 mg per ml has been advised to mitigate the inflammation. In addition, anti-inflammatory medications such as ibuprofen, 600 mg–800 mg, every 6 h should be prescribed. Patients should be re-evaluated on a regular basis. If there is improvement of symptoms, injections should be repeated and the anti-inflammatory medication should be extended and maintained, with regular follow-ups.

If conservative treatment proves unsuccessful, or the length of PH is above the normal defined limits, surgical management should be considered. If osteophytes, a prominent hamular process or bursa fibrosis, are present, the surgical treatment should be followed.

If inflammatory or fibrotic changes of the bursa are not present, such as in our case, a hamulotomy should be accomplished, or the hamulus should be removed, by the use of a rongeur. One reported complication of hamulotomy is interruption of the tensor veli palatini function over this osseous hook that results in tubic dysfunction with otic expression as hypoacusis besides loss of a palate-pharynx seal during speaking and swallowing. Table 2 describes a clinical guide enumerating the symptoms, differential diagnosis, and treatment options for this entity.

Conclusion

PH syndrome, a rare cause of orofacial pain, was reported in this article. Through this article, we have presented a literature appraisal on the incidence, diagnosis, and treatment of this condition. The palatal and pharyngeal areas warrant particular clinical surveillance during the differential diagnosis of orofacial pain. As the treatment for

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

Features	
Etiology	Injury, infection, or a preexisting condition
Clinical features	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 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

this entity is markedly different from that for the other pain conditions of this region, clinicians should contemplate a potential diagnosis of pterygoid hamular syndrome.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patients have given their consent for their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest

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