Clicking hyoid: A rare case report and review

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ABSTRACT

Painful conditions of unexplained origin pose a challenging task for the clinician from both the diagnosis and treatment perspective. This might be accounted due to overlapping symptomatology; moreover, nonspecific subjective findings push the diagnostic process to a more perplexing direction. The main loophole in their management lies in difficulty to make an appropriate diagnosis. Clicking hyoid is an extremely rare anomaly of hyoid bone which produces painful clicking in throat. Here, we report a case of a young patient presented with pain and clicking in throat which aggravated on swallowing. After a series of examinations, it was diagnosed as clicking hyoid and was managed successfully by surgical treatment. The objective of this article is not only to increase awareness of the ailment so as to validate its existence but also demonstrate the significant utility of the surgical management.

Key words: Clicking, cornua, hyoid, pain, swallowing

INTRODUCTION

Painful clicking in the throat is a rare entity to be encountered in the clinical settings. Its diagnosis is equivalent to solve a puzzle and is primarily based upon exclusion criteria. Clicking hyoid is an abnormal condition of hyoid bone which is characterized by enlargement of greater cornua of hyoid bone. These enlarged cornua impinge on cervical vertebrae. This impingement of cervical vertebrae produces interosseus friction. This frictional relation between the two bones is responsible for pain and clicking sound which is perceptible to the patient. The pain and clicking usually aggravates on swallowing as well as on neck movements. The abnormal enlargement can be a sequel of trauma.

Clicking sensation and pain while deglutition is an unpleasant condition, which produces physiological and psychological bearings. This dictates the need for accurate diagnosis and proper management of the condition. Different modalities of treatment have been reported in the literature. These include conservative and surgical approaches. Here, we report a case of a young female who visited to seek treatment for suspicion of some cancerous lesion causing pain and clicking in throat. Due to long standing history of problem, the patient started developing loss of interest in surroundings and sleep disturbances lending her to life of senility and seclusion. The patient was diagnosed as a case of clicking hyoid. The case was managed surgically with resection of greater cornua of hyoid. The purpose of this case report is to discuss the etiology, clinical presentation, treatment options, and to validate the existence of the unusual anomaly “clicking hyoid” in the differential diagnosis of chronic pain in the maxillofacial region.

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CASE REPORT

A woman aged 35 years presented to our department with a 1-year history of clicking noise in her throat on swallowing [Figures 1 and 2]. This was associated with pain but no difficulty on swallowing. She denied any previous history of trauma. She did not have any respiratory symptoms or jaw-locking. Taking into consideration, the patient’s symptom logy; it was suspected to be a case of the salivary gland sialolith. On sialography and ultrasonography, no such finding was evident.

On clinical examination, a trigger point was palpable on anteroinferior aspect of masseter muscle. Patient had an associated sleep disturbance and loss of interest in surroundings. Beck’s depression index was used to assess the depression associated with pain. Patient was found to have moderate depression. She was given trigger point injection with 0.5 ml of 0.5% injection bupivacaine. However, patient had no relief. For alleviation of pain associated depression, tricyclic amitriptyline antidepressant 75 mg was used for 3 weeks. Patient got no relief in pain and clicking. On further examination of throat, CT scan was done [Figure 3] greater horn of hyoid bone was found large in size with abnormally long greater cornua. While swallowing, a significant click was as greater cornua were impinged, caught, and slipped back the spine. Subsequently, the diagnosis was made as “clicking hyoid.”

Patient was explained of the diagnosis and offered the options of conservative and surgical modalities of treatment. However, patient elected the conservative one. To commence the conservative treatment, hyoid bone was palpated digitally as advised by Brown. The patient was given local lignocaine plus steroid injections weekly for 1 month on the tip of the affected cornua. However, patient reported with no relief. Thereafter, the patient was planned for surgical resection of greater cornua under general anesthesia. An informed written consent was obtained from the patient for the surgery.

Superior limb of Mcfee incision was given. Skin, subcutaneous tissue, platysma were incised. Blunt dissection was done through deep cervical fascia. Middle pharyngeal constrictor was bluntly dissected from the greater cornua to expose it [Figure 4]. After the exposure, greater cornua was resected from its junction to the body [Figure 5]. Layer wise closure was done. The patient was followed up with an uneventful healing with complete resolution of clicking and pain [Figure 6].

DISCUSSION

The hyoid is horseshoe shaped bone which is suspended from the tips of the styloid processes of the temporal bones, straddled by the stylohyoid ligaments. It consists of five segments. These constitute a body, two greater cornua, and two lesser cornua. The hyoid is ossified from six centers: Two for the body and one for each cornu. Ossification commences in the greater cornua toward...
the end of fetal life, in the body shortly afterward, and in the lesser cornua during the 1st or 2nd year after birth.\textsuperscript{[1]} It has no direct contact with any other bone in the human body. The hyoid bone forms a movable base for the tongue and its varied movements and is held in position by a large number of muscles. The hyoid bone has connections with muscles to the mandible (mylohyoid), tongue (hyoglossus), skull (stylohyoid), thyroid cartilage (thyrohyoid), sternum (sternohyoid), to the medial border of the scapular notch (omohyoid), and to the pharyngeal median raphe (middle pharyngeal constrictor muscle).\textsuperscript{[2]}

Diagnostic testing included digital or bimanual palpation with the index finger on the greater cornua of the nonaffected side. This procedure directed the entire hyoid toward the surface of the skin of the affected side, with the thumb stabilizing the affected cornua at the site of injury, as recommended by Brown.\textsuperscript{[3]} When pressure was applied to the left cornua, the radiation of painful symptoms was intensified when pressure was maintained.

The anomalies of hyoid bone are of congenital as well as acquired variety [Table 1]. Since these are infrequently encountered in clinical practice, only a few anomalies of hyoid bone and its associated structures has been reported in the literature so far. In the present case, the condition was of acquired variety. However, there was no history of trauma or inflammation which could be suspected for condition.

**Clicking hyoid**

Congenital enlargement of hyoid bone was reported by Morrissey and Alun-Jones\textsuperscript{[4]} in 1989. The patient reported with the chief complaint of pain on swallowing in addition with jaw locking. Radiographic examination revealed large and splayed greater horns impinging inside of the mandible. The author stated that partial excision of bilateral greater cornua of hyoid provide complete resolution of the problem.

A similar case of painful clicking was documented by Makura and Nigam,\textsuperscript{[5]} where patient was diagnosed as clunking neck with characteristic finding of noisy painful neck movements. Greater horn of hyoid was found to be enlarged, and complete resolution of symptoms was achieved by surgical excision of greater cornua of hyoid bone.

| Table 1: Flow chart visualizing the causes of clicking hyoid |
|--------------------------------------------------------------|
| **Congenital**                                               |
| **Acquired**                                                 |
| **Post Surgical**                                            |
| **Inflammatory**                                             |
| **Oral Habits**                                              |
| **Radical Neck Dissection**                                  |
| **Mandibular Advancement**                                   |
Another milestone of painful clicking was added in the literature by Ilankovan. He reported a case of clicking aggravated on neck movements. The hyoid was normal with no previous history of trauma. However, on further detailed examination, an accessory abnormal bone was found. Initially, the patient was managed conservatively. However, the symptoms worsened. Hence, on surgical exploration, a long bone articulating by synovial joint with superior cornua of thyroid cartilage and greater cornua of hyoid bone. In the present study, the patient denied of any previous history of trauma. However, on palpation, greater horn of hyoid bone was found to be enlarged.

In the 17th century, stylohyoid ossification was diagnosed as a result of the great work of Marchetti of Padua. Weinlecher has been credited for reporting a case treated with removal of styloid process. Stylohyoid ligament ossification has been documented by Demanchetis in 1652. Later, in the 20th century, Eagle and Durham as well as Moffat et al. as reported a syndrome associated with dull throat pain which is usually confined to tonsillar fossa. The pain was aggravated on deglutition, neck movements, and radiating to ear. Dolan in 1949 supported Moffat et al. that Styloid syndrome should be considered in the differential diagnosis of atypical pain.

The list of acquired roots of hyoid anomalies is replete of diversity. Myriad of individual causes and plethora of surgical procedures are associated with the condition.

Bibby studied the effect of oral habits such as mouth breathing and tongue thrusting on the hyoid bone position in 18 patients. Mouth breathers may have altered the position of hyoid bone. In our patient, there was no characteristic oral habit considered responsible in the literature.

The hyoid bone syndrome was first described in 1954. The syndrome is characterized by regional tenderness in proximity to the greater cornua of the hyoid bone. After that, in 1968, the syndrome was considered to be a form of insertion tendinosis of stylohyoid tendon, which normally suspend the hyoid bone from styloid process. Moreover, transient alleviation of pain was reported with injection of procaine HCl and corticosteroid at the tip of the greater horn. Lim in 1987 reported 50 patients suffering from the hyoid bone syndrome. All patients had excision of the involved greater horn after 1 month of failure of conservative medical treatment. The surgical excision promptly relieved pain in 90% of the patients. Moreover, he claimed that an understanding of the intimate relationship of greater horn of the hyoid with the posterior pharynx and carotid sinus is essential in detecting the hyoid bone syndrome. In our case, the condition was managed by surgical excision of greater horn.

Radical neck dissection is one of the reported factors associated with hypertrophy of sternohyoid muscle. This hypertrophied infrahyoid in turn produces troublesome ailment of hyoid. It can originate from some orthognathic surgical procedures employed for the management of facial asymmetry. The commonly associated procedures documented in the literature are for mandibular advancement. In the present case, patient had never undergone any of the above mentioned surgical procedure. It was very difficult to diagnose the condition in the absence of any congenital involvement, history of previous trauma and inflammation, surgical treatment, habit.

Conclusion
Clicking hyoid is an unusual anomaly and rare anomalies not only present an intriguing quest to solve but also provide positive reinforcement to diversify the diagnostic eye field of the clinician. Clicking hyoid is more intriguing from diagnostic prospective. An appropriate treatment modality cannot be instituted in lieu of improper diagnosis. A various number of root causes have been suggested at varying periods of time. The workhorse of treatment is surgical excision of greater cornua of hyoid bone. Hence, we believe that it should be considered in the diagnosis of painful deglutition.

Declaration of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/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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