Ganglion Cyst of Temporomandibular Joint: A Rare Case Report and Literature Review

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1. Abstract
Ganglion cysts are the most common of the more superficially located myxoid lesions and represent the most common soft tissue tumors that develops in the hand and wrist region. They are fluid-filled lesions that arise from the capsule of a joint or sheath of a tendon. There is no single theory that fully explains their pathogenesis, but trauma is a common factor in their development. Repetitive trauma to the joint capsule and ligamentous supporting structures results in degeneration of the connective tissue of the capsule and stimulates the production of tissue lubricants by fibroblasts. The resultant fluids accumulate and develops into a ganglion cyst. However, ganglion cysts lack a lining epithelial layer, distinguishing it from synovial cyst. The following is a case report of ganglion cyst of the temporomandibular joint (TMJ), which are rare with 50 case reports from 1977 to present. This report describes a ganglion cyst emanating from the medial portion of the right TMJ capsule, and reviews surgical treatment and management of this case.

2. Background
Ganglion cysts are the most common of the more superficially located myxoid lesions and represent the most common soft tissue tumors that develops in the hand and wrist region. They are fluid-filled lesions that arise from the capsule of a joint or sheath of a tendon. They most commonly occur in women in the third to fifth decade. There is no single theory that fully explains their pathogenesis, but trauma is a common factor in their development. Repetitive trauma to the joint capsule and ligamentous supporting structures results in degeneration of the connective tissue of the capsule and stimulates the production of tissue lubricants by fibroblasts. The resultant fluids accumulate and develops into a ganglion cyst. However, the ganglion cyst lining lacks an epithelial layer, distinguishing it from synovial cyst. This is a case report of a ganglion cyst presenting within the TMJ, which are rare with 50 case reports from 1977 to present [6-20, 29-49]. This report describes a ganglion cyst emanating from the medial portion of the right TMJ capsule, the surgical approach required to access the lesion, and a review the literature on ganglion cyst. Ganglion cysts rarely affect the temporomandibular joint and typically present with swelling in the preauricular region, pain, trismus, and difficulty with mastication. Diagnostic imaging tools, such as computed tomography and magnetic resonance imaging, have aided in diagnosis; however, only histopathologic examination will lead to a definitive diagnosis. The precise management of ganglion cysts of the TMJ remains uncertain owing to the rarity of these lesions. Treatment has focused on surgical excision without regard for lesion size or symptoms. Here we report a case of a patient with left ganglion cyst of the TMJ with persistent symptoms of pain and dysesthesia.

3. Case Report and Result
A 60-year-old Caucasian male initially presented to an outside clinic with chief complaints of left temporomandibular joint pain, left auriculotemporal (AT) nerve dysesthesia, and numbness (paresthesia) to both the left maxillary and mandibular molars. His initial work-up included a laryngoscopy and a computed tomography (CT) with contrast. The laryngoscopy revealed unremarkable findings and the CT scan showed only mild mucosal thickening of the sinus membrane but failed to demonstrate any pathology or lesions that could be clinically correlated with his symptoms. Without definitive etiology, his symptoms were treated pharmacologically. With the presentation of persistent left TMJ pain, AT nerve dysesthesia, and altered sensation, an MRI was obtained to rule out a neurologic etiology. The MRI showed an 11 x 7 mm high T2, low T1 signal mass aris-
ing from the ventral margin of the medial pole of the left temporomandibular joint. The lesion was seen in the region of mandibular branch of the fifth cranial nerve possibly resulting in compressive neuropathy (Figure 1).

At this time, he was referred to the Oral and Maxillofacial Surgery clinic at Dwight D Eisenhower Army Medical Center for further evaluation. His medical history was significant for depression, hyperlipidemia, GERD, low testosterone, and seasonal allergies. His medication list included: Zoloft, Flonase, Viagra, Ambien, and AndroGel. He was allergic to penicillin and denied the use of tobacco and alcohol.

Clinical examination revealed a maximal incisor opening of 45 mm without deviation on opening. He demonstrated normal lateral excursive and protrusive movements. There were no clicks nor pops in the TMJ, and no crepitus, bruits, nor pulsations were appreciated on auscultation and palpation. He reported previous trauma to his left joint. His dental exam was unremarkable with only his wisdom teeth absent with no active caries. He had stable and reproducible class I molar and canine occlusion and had no signs of parafunctional habits. His tympanic membranes were clear and intact. There was no palpable mass or swelling in the pre-auricular region and no lymphadenopathy. Cranial nerves II-XII were intact bilaterally, except for his complaints of left TMJ pain, left AT dysesthesia, and paresthesia to left maxillary and mandibular molars. A panoramic radiograph was obtained and did not show any abnormalities (Figure 2). Based on the clinical exam and the imaging the surgeons determined the lesion most likely represented a ganglion or synovial cyst.

Figure 1: Axial T2-weighted MRI obtained preoperatively showing a well-circumscribed lesion with a homogenous hyperintensity that measures 11x7mm in size located on the medial pole of the left TMJ.

Figure 2: Pre-operative panoramic radiograph without any significant remarkable findings.

4. Diagnosis and Management

Following a detailed discussion with the patient concerning various treatment options he elected for surgery. With the patient under general anesthesia, a pre-auricular incision was performed to access the left temporomandibular joint. Once dissection was completed down to the zygomatic arch, it was extended anteriorly in a subperiosteal plane to expose the lateral aspect of the articulating eminence. An attempt was then made to dissect medially and anterior to the condyle but, due to limited access, the team was unable to visualize the lesion.

A left submandibular incision was then made 2 centimeters below the inferior border of the left mandible. Dissection was carried through the subcutaneous fat, platysma, and superficial layer of the deep cervical fascia until identification of the facial vein and artery
was completed. These vessels were ligated, cut, and retracted superiorly. The position of the nerve was identified with the use of a nerve stimulator and retracted superiorly. The pterygomasseteric sling was then excised and dissection was carried up to the sigmoid notch exposing the lateral border of the ramus. Utilizing a reciprocating saw, a vertical ramus osteotomy was then completed 1 cm anterior to the posterior border. The proximal mandibular segment was then rotated laterally, and the lesion was then accessed from the pre-auricular incision.

The cystic lesion was found and identified; however, it decompressed prior to its complete removal expressing a thick gelatinous substance. The decompressed tissue and a small cuff of surrounding muscle was removed and sent to pathology for histological examination. The mandible was then fixated using a strut plate oriented in a vertical fashion with monocortical screws and the incisions cleaned and closed in a standard fashion (Figure 3).

Histologic evaluation of the hematoxylin and eosin stained slide confirmed the diagnosis of ganglion cyst. The slides demonstrated dense fibrous connective tissue aggregates with focal myxoid change surrounding multiple, irregular cyst-like spaces in a background of skeletal muscle and adipose tissue (Figures 4A and 4B). No definitive epithelial lining is noted. These microscopic findings are characteristic of a ganglion cyst.

The patient was discharged from the hospital on post-operative day two and had an unremarkable healing process. His pre-operative symptoms of left TMJ pain, left AT nerve dysesthesia, and paresthesia resolved within several weeks. A repeat MRI was obtained four months following surgery, showing no lesion around the vicinity of the left TMJ. The articulating disc was noted to be in a normal location and with appropriate movement during functioning. The patient’s presentation and improvement following surgery suggest that the location of the cyst in the TMJ and its proximity to the course of the auriculotemporal nerve may have caused auriculotemporal dysesthesia along with the constellation of the other symptoms.

**Figure 3:** Panoramic radiograph obtained immediate postoperatively after removal of cystic lesion along with fixation with a strut plate.

**Figure 4:** Photomicrographs of the hematoxylin and eosin-stained specimen demonstrate multiple compartments of cyst-like spaces surrounded by dense fibrous connective tissue with focal myxoid change in a background of skeletal muscle and adipose tissue (4A). A higher magnification view demonstrates spindle cells in the dense fibrous connective tissue wall with no definitive epithelial lining (4B).
5. Discussion

The location of the ganglion cyst in this report is note-worthy due to it being the first documented case where the lesion was located along the medial aspect of the joint capsule. All reported ganglion cysts associated with the TMJ usually have occurred lateral to the joint. The most common symptoms are swelling in the pre-auricular region and pain confined to the location of the cyst that is often accentuated by movement of the joint [3,7] If the mass is palpable, it is generally firm, smooth, and slightly moveable. The initial differential diagnosis often includes a parotid mass, sebaceous cyst, branchial cleft cyst, neuroma, lipoma, vascular lesion, and lymphangioma [11] Typically, ganglion cyst will be omitted from the differential diagnosis due to its uncommon occurrence. In fact, there are case reports in the literature where ganglion cyst in the preauricular region have been treated initially as parotid tumors [8]. Besides this reported case, other cases with atypical presentations include extension of the TMJ ganglion cyst into the middle cranial fossa causing facial nerve palsy [19], and a report a case with auricle paresthesia that was thought to have risen from compression of the auriculotemporal nerve [6].

Ganglion cysts are found in the collagenous connective tissue of a joint capsule or tendon sheath. They arise from myxoid degeneration of the connective tissue surrounding the capsule of the joint and do not communicate with the joint cavity. They are filled with a clear, gelatinous fluid, and do not have an epithelial lining. In the past some authors have erroneously categorized ganglion cyst and synovial cyst to be synonymous.

Synovial cyst can communicate with joint space and are formed either by herniation of the synovial into the surrounding tissue or displacement during embryogenesis. Synovial cysts contain synovial fluid and have an epithelium lining containing synoviocytes [10]. Both ganglion cysts and synovial cysts can be a result of trauma, but only synovial cysts have an primary inflammatory process [4]. Despite the different causes and histological descriptions, the clinical characteristics and treatment of these two cyst are the similar.

The hematoxylin and eosin staining of ganglion cyst is straight forward and will show a cystic type of lesion that has a dense fibrous connective tissue resembling a cyst wall. No epithelial lining will be present. The immunohistochemical staining of the lining of ganglion cyst will be positive for vimentin [18]. Some authors have reported the use of chromatography in analysis of the fluid, but results were inconclusive and further studies of are needed to understand their significance [12].

There is no consensus regarding the best imaging technique for this lesion, but the use of MRI in the presented case was essential to detecting the lesion. MRI is very helpful in determining the lesions size, relationship to other structures, nature (cystic vs. non-cystic), and serve as a guide during the treatment planning process. Other pre-operative diagnostic imaging that has been used in the past include computer tomography (CT), arthrography [23], and ultrasonography. When obtained, plain radiographs are usually normal, but a noteworthy case of abnormal finding includes an intraosseous ganglion cyst within the mandibular condyle [9]. Lopes et al [24] found that ultrasound can help differentiate from other lesion or anatomic structures due to its anechoic picture. However, other reports have shown ganglion cyst to be hypochoic [7]. With these conflicting reports we can conclude that ultrasound is not reliable in the diagnosis of ganglion cyst.

Numerous forms of treatment have been considered for ganglion cyst including mechanical compression, aspiration, and injection with hydrocortisone, but surgical excision remains the treatment of choice for symptomatic lesions. Ganglion cyst can resolve spontaneously without treatment [13], however observation fails to provide a diagnosis and biopsy is not recommended when other tumors or malignancies cannot be ruled out. Mechanical crushing involves compressing the lesion until it burst. This technique has the highest recurrences rate and has not been used to treat any of the reported cases of TMJ ganglions. Aspiration with or without injecting the lesion with steroids are indicated by some orthopedic surgeons, but this treatment modality has a lower success rate then surgical excision and can cause thinning of the overlying skin. All ganglion cyst associated with the TMJ found in the literature have been treated with surgical excision and no recurrences have been reported. However, orthopedic literature has shown that lesion with complete surgical removal can reoccur.

Lesions located on the medial aspect of the condyle and ramus can create difficult surgical access. Due to the deep location of the lesion, nearby significant vessels, and limited field of view the surgeon must be very familiar with the anatomy this region to avoid significant complications. Yang et al [17] suggest using a preauricular incision and transcondylar approach. From his incision, without disrupting the capsule or pterygoid muscle attachments, he was able to expose the lateral condyle down to the sigmoid notch. The condyle was cut off at the level of the sigmoid notch, and rotated in a upward and counterclockwise direction. The lesion was removed, and the condyle was rotated back into position and fixated with 2 titanium plates. In this case report, a pre-auricular incision as well as a submandibular incision with a vertical ramus osteotomy was utilized to access the lesion. Despite the difficult location, the surgery team was able to obtain adequate surgical access and remove the lesion.
References