Case Report

Treatment of tophaceous pseudogout with custom-fitted temporomandibular joint: a two-staged approach

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ABSTRACT

Tophaceous pseudogout, a variant of calcium pyrophosphate dihydrate deposition, is a relatively rare juxta-articular disease. It is a metabolic condition, in which patients develop pseudo-tumoral calcifications associated with peri-articular structures secondary to calcium pyrophosphate deposition into joints with fibrocartilage rather than hyaline cartilage. These lesions are reported in the knee, wrist, pubis, shoulder, and temporomandibular joint (TMJ) and induce a histiocytic foreign body giant cell reaction. We report a case of tophaceous pseudogout affecting the left TMJ with destruction of the condyle and glenoid and middle cranial fossa that was reconstructed with a TMJ Concepts (Ventura, CA) custom-fitted prosthesis in a 2-staged surgical approach using a silicone spacer. The surgical management using a patient-specific TMJ is a viable option when the fossa or condylar component has been compromised due to breakdown of bone secondary to a pathologic process. Our case describes and identifies the lesion and its rare occurrence in the region of the temporomandibular region. The successful management of tophaceous pseudogout of the TMJ must include a thorough patient workup including the involvement of other joints as well as the modification of bone of the glenoid fossa and condylar relationship of the TMJ. © 2015 The Authors. Published by Elsevier Inc. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

1. Introduction

Tophaceous pseudogout is a variant of calcium pyrophosphate dihydrate deposition (CPPD), a relatively rare juxta-articular disease. It is a metabolic condition where patients develop pseudo-tumoral calcifications associated with peri-articular structures secondary to CPPD deposition into joints with fibrocartilage rather than hyaline cartilage [1,2]. These lesions are reported in the knee, wrist, pubis, shoulder, and temporomandibular joint (TMJ), and induce a histiocytic foreign body giant cell reaction [1,2]. We report a case of tophaceous pseudogout affecting the left TMJ with destruction of the condyle, glenoid fossa, and middle cranial fossa that was reconstructed with a TMJ Concepts (Ventura, CA) patient-specific prosthesis [3] in a 2-staged surgical approach using a silicone spacer.

2. Case report

A 67-year-old female presented to the outpatient clinic of Geisinger Medical Center. The patient’s chief complaint was pain of the left TMJ and ear region of 5 years’ duration with increasing pain of 2 months’ duration. The patient also reported an inability to clean her left ear due to increasing difficulty of inserting cotton tip applicators in her ear. The patient denied any history of stress, tension, or bruxism. Her medical history was significant for uncontrolled type II diabetes mellitus, hypertension, hyperlipidemia, and an incarcerated incisional hernia, which was status post surgical repair. The patient was informed that her lack of glycemic control could impact her health as well as any surgical outcomes, and as such, the patient was referred to her primary care physician for optimization and control of her type II diabetes mellitus.

On clinical examination, the patient had a maximum incisal opening of 35 mm with no associated clicking or popping. She displayed deviation of the mandible 5 mm to the left on opening with point tenderness of the left TMJ upon palpation. A radiographic evaluation by panoramic exam revealed a diffuse radiopacity of the left mandibular condyle, warranting further investigation [Figures 1,2]. The patient was subsequently scheduled for a maxillofacial computed tomography (CT) scan without contrast, which revealed a calcified lesion without cortical borders encompassing the left condylar head with posterior extension to the external acoustic meatus. The lesion also extended superiorly to the skull base with possible perforation into the middle cranial fossa [Figures 3,4,5]. To rule out intracranial involvement, a neurosurgeon was consulted.
who advised that the patient obtain a magnetic resonance imaging of the brain to further elucidate the extent of the potential perforation. The magnetic resonance imaging revealed a mineralized left TMJ space abnormality consistent with calcification, along with superimposed degenerative and erosive changes at the left mandibular condyle. Extension of the lesion involved the medial margin of the mandibular fossa with possible direct contact with the middle cranial fossa [Figures 6, 7, 8]. The radiographic imaging was inconclusive as to the precise extent and involvement of the temporal bone. Bilateral wrist and knee radiographs were also obtained per recommendations by radiology to rule out CPPD. The radiographs returned negative for CPPD.

Differential diagnosis included osteochondroma, chondrosarcoma, CPPD deposition, synovial chondromatosis, ossifying fibroma, and osteosarcoma. Following all diagnostic imaging, surgical options were discussed with the patient that included an excisional biopsy of the left TMJ mass with condylectomy, reconstruction of cranial base, immediate reconstruction of the left TMJ with a stock prosthesis or delayed reconstruction with a patient-specific prosthesis. Navqi et al. in 2008 [4] described the use of aspirate cytology as an effective diagnostic tool for those patients suspected to have tophaceous pseudogout of the TMJ. He described tophi in cytologic views for those afflicted with tophaceous pseudogout. In this case, a decision was made to biopsy the lesion with evaluation of both frozen and final histologic specimens. The patient was advised preoperatively that the treatment plan could be modified pending the results and aggressiveness of the lesions following frozen section.

Stage I surgery consisted of the condylectomy with excisional biopsy of the mass and placement of maxillomandibular fixation. A silicone spacer was used to prevent post-resection fibrosis following the unilateral condylectomy. A preauricular incision was made to gain access to the lesion and mandibular condyle. The lesion was a white, crystalloid mass that appeared to be granulated, crumbling easily to touch [Figure 9]. A wedge of the lesion was sent for intraoperative frozen histologic examination. The lesion was determined to be benign, displaying a foreign body giant cell reaction with evidence of tophi [Figures 10, 11, 12]. An intraoperative diagnosis of tophaceous pseudogout of the TMJ was the initial
impression. The lesion was removed in its entirety by performing a condylectomy to 25 mm below the condylar head. No clinical perforation into the middle cranial fossa was noted; however, marked degenerative and erosive changes of the bony fossa were present. The patient was then placed in maxillomandibular fixation with placement of an appropriately sized silicone spacer [Figures 13,14]. The patient remained in maxillomandibular fixation after completion of the procedure to prevent migration of the spacer. A postoperative maxillofacial CT scan was performed per TMJ Concepts’s protocol for patient-specific prosthesis fabrication [Figure 15].

The patient was admitted to the hospital for postoperative monitoring and pain management. The only complication noted in the immediate postoperative period was weakness of the temporal branch of cranial nerve VII. The patient was subsequently discharged.
after a short hospital stay and followed up as an outpatient for the duration of recovery after stage I surgery without further complication.

Eight weeks later, stage II surgery was performed. Maxillomandibular fixation wires were removed as part of the non-sterile portion of the surgical procedure. The silicone spacer was removed via the previous preauricular incision, and a retromandibular incision was utilized for access to the mandibular ramus. The patient-specific prosthesis was then placed with intimate adaptation to both the glenoid fossa and the ramus. The preoperative occlusion was confirmed intraoperatively. The patient tolerated the procedure well without complications and was extubated and brought to the PACU in stable condition. Postoperative maxillofacial CT scan confirmed appropriate placement of the prosthesis without any indication of perforation into the middle cranial fossa [Figure 14]. In the postoperative phase, physiotherapy was implemented to enhance controlled motion following TMJ replacement surgery.

Figure 8. Three dimensional reconstruction of the maxillofacial computed tomography scan without contrast from posterior-lateral view showing medial extent of the mass.

Figure 9. Majority of the mass resected from the left temporomandibular joint. Histopathology reveals a diagnosis of tophaceous pseuodgout.

Figure 10. 40x crystals with compensated polarized light used to identify calcium pyrophosphate crystals.

Figure 11. 40x crystals with decompensated polarized light.

Figure 12. Histologic exam showing foreign body giant cell reaction.
The patient is currently 1 year status post surgery with normal function of the prosthetic joint, with a maximum incisal opening of 40 mm without complaints.

3. Discussion

CPPD is a metabolic arthropathy characterized by intra- and periarticular calcium crystals [5,6] that primarily affects late middle-aged and elderly patients. CPPD has also been identified as pseudogout and chondrocalcinosis. CPPD is believed to be caused by a metabolic disturbance of phosphate metabolism. The cause of acute attacks is unknown; however, it is believed that crystals shed from the articular cartilage surface secondary to trauma, degradation of cartilage by localized infection or inflammation, or alterations in inorganic pyrophosphate concentrations in synovial fluid [1,5].

Tophaceous pseudogout is an uncommon variant of CPPD. In comparison to other joints, the TMJ is the most affected site by tophaceous pseudogout [2]. A comprehensive medical history and clinical and radiographic examinations are imperative in diagnosis and treatment planning of this condition. Associated symptoms may include facial pain, otalgia, preauricular swelling, trismus, and tenderness to palpation [6]. Systemic conditions that have been found in association with tophaceous pseudogout include: rheumatoid arthritis, gout, hypomagnesemia, hypothyroidism, amyloidosis, hypophosphatemia, hyperparathyroidism, hemosiderosis, hemochromatosis, and familial hypocalciuric hypercalcemia [1,2,5]. Histologic examination is needed for final diagnosis of tophaceous pseudogout.

Treatment of tophaceous pseudogout ranges from conservative pharmacotherapy with nonsteroidal anti-inflammatory drugs and colchicine to surgical debridement and resection [1,5,7]. If surgical intervention is utilized, meticulous removal of crystal deposits should be performed due to potential for significant bony involvement. In a review of the literature, in 2011 Ascani et al. found 58 patients with reported cases of tophaceous pseudogout of the TMJ. Of these 58 patients, 33 patients received total removal of the lesion. Two of these patients received reconstruction status post resection, one with a costochondral graft and one with a TMJ prosthesis. The TMJ prosthesis placed for reconstruction was a Christensen TMJ implant [8].

Our patient was offered two treatment options: (1) single-stage surgery using a stock TMJ prosthesis; and (2) two-stage surgery with a patient-specific TMJ prosthesis. When presenting the options to the patient, the risks and benefits of option #1 were described as follows: overall less morbidity due to single stage procedure with...
no need for maxillomandibular fixation postoperatively. Possible contouring of bone to ensure the proper fit of the prosthesis would be determined intraoperatively. The risks and benefits of option #2 were described as follows: increased morbidity of two surgical procedures with maxillomandibular fixation and placement of a silicone spacer during the fabrication of the patient-specific prosthesis (possibly 8 weeks or longer). Intraoperative contouring of the mandible would not be necessary with the patient-specific prosthesis. The usual surgical postoperative sequelae associated with preauricular and submandibular approaches (pain, swelling, bleeding, infection, scarring, cranial nerve VII deficits, and sensory deficits) were discussed in detail, as they were relevant to both approaches. After careful discussion and consideration of all parameters, both the patient and the surgeon agreed to opt for a patient-specific TMJ prosthesis.

This case demonstrated reconstruction of the temporomandibular joint using a two-staged approach. Stage I of the protocol involved a preoperative CT scan, followed by a gap arthroplasty of 2-3 cm and placing the patient in maxillomandibular fixation. A silicone spacer was then fitted appropriately to preserve the anatomic surgical site for future placement of the patient-specific prosthesis. A CT scan was then obtained immediately postoperatively and submitted for fabrication of the patient-specific prosthesis while in maxillomandibular fixation. Stage II consists of the removal of the silicone spacer and placement of the patient-specific prosthesis with an abdominal fat graft. The autologous fat graft was placed deep to and around the prosthetic condyle to prevent heterotopic bone formation [9,10]. Postoperatively, the patient was given a regimen of physiotherapy and jaw exercises. The two-stage technique for placement of a patient-specific prosthesis proved to have clinically satisfactory results. This case report describes the use of a two-staged process and placement of a patient-specific TMJ prosthesis as a consideration in reconstruction of bony defects after surgical resection in patients with tophaceous pseudogout.

Consent

Written informed consent was obtained from the patient(s) for publication of this case report and case series and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Conflict of interest

The authors declare there are no conflicts of interest.

References