

# Neurogenic Heterotopic Ossification within the Temporomandibular Joint in a Stroke Patient

Z Ayyoub, GDouglas, M Khan, B Carroll

Rancho Los Amigos National Rehabilitation Center

## Abstract

A 50 year-old male with a history of atrial fibrillation on Coumadin developed an acute infarct of the right Middle Cerebral Artery (MCA) involving the frontal and temporal lobes. The patient developed cerebral edema with a midline shift and hemorrhagic conversion. A ventriculostomy was performed followed by an emergent frontotemporal decompressive craniotomy. The patient developed uncal herniation and underwent a redo frontotemporal craniectomy with right temporal lobectomy and decompression of midbrain with evacuation of the basal ganglia hematoma. On admission to an inpatient rehabilitation unit, approximately 20 months after onset of injury, the patient was found to have diffuse spasticity with multiple severe contractures, limited range of motion, hemifacial spasms, bruxism and cervical muscle dystonia. Patient had limited opening of his mouth both actively and passively, work up revealed mature

heterotopic ossification (HO) of the left temporomandibular joint (TMJ) at the condyloid process at the level of the condyloid head and neck of the mandible.

**Key Words:** Heterotopic ossification; Temporomandibular joint; Stroke; Bruxism

## Introduction

Heterotopic ossification has been a well-documented issue for patients with spinal cord injury (SCI), traumatic brain injury (TBI) and burns<sup>1</sup> It is defined as the extra-articular formation of lamellar bone within the soft tissue surrounding a joint but outside the joint capsule.<sup>2</sup> While its etiology remains largely unknown, common sites of development and presenting symptoms have been discovered with associated treatments. Traumatic HO and neurogenic HO have been delineated as two common variants, both with associated patterns of development. HO usually is found in the proximal joints and limbs with a peak occurrence at 2 months from original injury.<sup>3-4</sup> Roentgenographic confirmation usually occurs at 2 months and the usual sites affected are the hips, shoulders and elbows. In this report we are presenting a case of a HO which is unusual in its site of development and its associated condition.

## Case Report

On September 7, 2003, a 50 year-old male with a history of atrial fibrillation on Coumadin developed loss of consciousness and left sided hemiparesis. He was first treated at Whittier hospital where he was diagnosed with stroke and then transferred to UCLA medical center where he was followed by neurology. An MRI of the brain on the same day revealed an acute infarct of the right MCA artery involving the frontal and temporal lobes. The patient began to deteriorate two days later and was diagnosed with cerebral edema and a ventriculostomy was performed, after which the patient began extensor posturing. He was treated with fresh frozen plasma to reverse the anticoagulation effect of Coumadin but CT scan demonstrated MCA infarct with severe edema and a 1cm midline shift with a hemorrhagic component. As a result the patient underwent an emergent frontotemporal decompressive craniotomy. Post-operative CT scan demonstrated marked worsening with more bleeding,

## Authors and their Affiliations

**Ziyad Ayyoub**, MD, Chief of Adult Brain Injury, Rancho Los Amigos National Rehabilitation Center, Assistant Professor of PM&R Loma Linda University, Clinical Assistant Professor of PM&R Western University of Health Sciences, Clinical Assistant Professor of Medicine, David Geffen School of Medicine at UCLA, USA.

**Garland Douglas**, MD, Clinical Professor of Orthopedics at USC/Rancho Los Amigos Hospital, Downey, CA, USA.

**Mohammed Khan**, MD, Chief Resident Physician, Loma Linda University Medical Centre, Loma Linda, CA/Rancho Los Amigos Hospital, Downey, USA.

**Breda Carroll**, Medical Student 4 th Year, Western University of Health Sciences, USA.

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## Correspondence

Ziyad Ayyoub, MD  
Rancho Los Amigos National Rehabilitation Center  
7601 East Imperial Highway  
Downey, CA 90242 USA

Phone: 562 (401) - 6073

FAX: 562 (401) - 6247

Email: zayyoub@dhs.lacounty.gov

particularly in the basal ganglia and caudate. He was diagnosed with uncal herniation and underwent a redo frontotemporal craniectomy with right temporal lobectomy and decompression of midbrain with evacuation of the basal ganglia hematoma. Subsequently patient recovered from surgery but remained minimally responsive. He had a PEG tube placed on September 20, 03; percutaneous tracheostomy on the September 22, 03 and was weaned from a ventilator and ultimately discharged on October 3, 03 to Shea Rehabilitation hospital in Whittier in a comatose state.

On October 6, 03, three days after his discharge, the patient was brought to the emergency room at Cedars-Sinai due to a high grade fever. He was in atrial fibrillation with rapid ventricular response and was admitted to the ICU with a fever of 104°F. CT of the brain demonstrated a right to left midline shift of 0.6cm secondary to edematous changes caused by the subacute infarcts seen in the right frontal, temporal and occipital lobes. His rate was controlled and it was discovered that he was growing coagulase negative staphylococcus in his blood. Subsequently a central line from his left subclavian was removed and he was treated with vancomycin, ceftazidime and flagyl. An EEG was performed on October 9, 03 showed diffuse generalized slowing and flattening, consistent with nonspecific generalized encephalopathy or toxic-metabolic encephalopathy. He was discharged in stable condition on October 15, 03 to the Barlow unit of Whittier Presbyterian hospital for long term care.

Subsequently patient had progressed from a comatose state in 2003 to being awake and alert and able to follow one step commands and communicate via hand gestures in 2005. These advances occurred while the patient was being cared for in the nursing facility at Whittier Presbyterian.

On June, 6, 05 the patient was admitted from home to Rancho Los Amigos National Rehabilitation Center (RLANRC) for inpatient rehabilitation. On admission he was found to have multiple severe contractures, limited ROM, hemifacial spasms, bruxism and cervical muscle dystonia/spasticity. The patient had been completely dependent in all activities of daily living with a tracheostomy and G-tube in place. On admission to RLANRC it was noted that the patient has severe bruxism, hemifacial spasm with an inability to open his mouth actively and limited opening of mouth passively by examiner. There was no prior history of trauma or TMJ joint or any prior dysfunction. It was also noted that the patient demonstrated twitching of the periorbital muscles, right platysmal muscle and diffuse twitching on the right side of face.

After careful evaluation and work up, taking in to consideration over all condition of the patient, on 6/7/05 EMG guided chemodenervation with Botox A was performed to the bilateral mentalis, medial pterygoid, temporalis and masseter muscles to treat severe bruxism. Right scalene, sternocleidomastoid and trapezius muscles were also injected with Botox A to treat his torticollis. Also muscles in the upper and lower extremities were injected due to spasticity. Following Botox injection patient had several episodes of oxygen desaturation. Workup to evaluate for possible aspiration pneumonia was conducted, blood culture was positive for gram positive cocci. He was subsequently treated with appropriate antibiotics as per infectious disease recommendations. Post Botox injection the patient continued to have an inability to open his mouth actively and passively by examiner.

The patient was transferred to the intensive care unit on June 27, 05 due to increased secretions and worsening oxygen desaturation. Work up revealed congestive heart failure with bilateral pleural effusions and he was placed on a ventilator and aggressively diuresed. After significant improvement in his condition the patient was transferred to step down unit but in view of earlier aspiration episodes a jejunostomy tube placement via endoscopic guidance was planned. The placement could not be carried out, however, due to extreme difficulty with passive opening of the patient's mouth in order to introduce the endoscopy tube. An X-ray and CT scan of the TMJ (Figures 1) were ordered on July 15, 05 to evaluate for possible causes for this limitation. The studies demonstrated mature heterotopic changes to the left TMJ at the condyloid process at the level of the condyloid head and neck. Triple phase bone scan was unnecessary test to confirm this finding. On August 28, 05 Patient was subsequently transferred to skilled nursing facility in stable condition.



Figure 1. CT of the TM Joint showing HO.

## Discussion

While there are numerous case reports and larger studies that address the issue of HO in the case of SCI and TBI, there are relatively few cases of HO in patients with stroke. Currently there are 3 published case reports documenting HO in patients with stroke, none of which involve the TMJ.<sup>5-6</sup> At the time of this review only one case report directly addressing the issue of HO in the jaw was found and it involved the anterior maxilla of a 13 year-old boy with no history of trauma or complaints of pain.<sup>7</sup> A second case report tackles hyperproliferation of bone in the anterior maxilla of a 2 year old child and hypothesizes that bizarre parosteal osteochondromatous proliferation (BPOP) represents late stage HO.<sup>8</sup> Both cases address the specific histopathology of HO versus osteochondroma in order to better identify and diagnose cases of HO in the maxilla.

In addition, there are several articles reporting on the issue of TMJ ankylosis, the bony or fibrous adhesion of the joint components leading to a loss of function. A review of 204 patients in Egypt who underwent surgical correction of their jaws attributed 98% of its patient's TMJ ankylosis to precipitant trauma but did not specify the type of trauma or age at which it occurred.<sup>9</sup>

The development of HO in the bilateral TMJs has been reported in a patient who had suffered second and third degree burns on 30% of his body (none affecting his head or neck). Over the subsequent 8 years he underwent a bilateral TMJ arthroplasty with two subsequent revisions secondary to reossification, leaving the authors to conclude that surgical resection in the case of HO may be of little value.<sup>10</sup> A second report involving the case of a 10 year old female with second and third degree burns over 40% of her body (including the right side of her face) developed HO of the right TMJ 2 years after her initial injury. She also underwent surgical excision of the joint with subcondylar ostectomy and arthroplasty but this report does not include long-term follow-up.<sup>11</sup>

HO has been found to occur at rates of 10-20% in the closed-head brain injury population with known contributing factors such as spasticity, fracture, infection and DVT. Stroke associated HO is quite rare with very few case reports in the literature.<sup>12</sup> While the etiology of HO is not clearly known, its pathogenesis is distinct from metastatic calcification and theorized to include the transformation of primitive cells of mesenchymal origin into osteogenic cells.<sup>13</sup> HO is most commonly found in the proximal joints and limbs and, in the case of SCI, below the level of lesion. Rarely has HO been demonstrated in the maxillary region and those reported cases are associated with burns or no trauma at all.

Clinically HO usually presents with limited range of motion, patient complaints of pain and localized swelling at the affected area. Lab work may reveal elevated

alkaline phosphatase and by two months post injury most patients with HO will have evidence on roentgenography. Treatment and prevention of HO ranges from bisphosphonates, NSAIDs (indomethacin and ibuprofen), radiation therapy to surgical resection in certain cases.<sup>14, 15</sup> The lack of post-stroke HO reported might indicate difficulty associated with diagnosis due to issues such as multiple pain sources and additional impairments both of which frequently affect stroke patients. This study reports a presentation of HO which is unusual in its location (TMJ) and associated comorbidity (stroke).

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