Case Report

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The Value of SPECT CT in Patient Management of Osteoarthritis of the Temporomandibular Joint

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Abstract

A 30 year old female patient presented in 2009 with severe bilateral temporomandibular joint (TMJ) pain, restricted mouth opening and tenderness over the masseter muscles. She was initially diagnosed with Wilkes Stage IV of the right and left TMJ. A diagnostic arthroscopy performed bilaterally failed to relieve her pain. She was diagnosed with an isolated spondyloarthropathy of TMJ based on a positive test for HLA B. Further medicamentous treatment was unsuccessful over subsequent months and years. CBCT of the TMJ showed discrete sclerosis of the right TMJ. Based on an additional SPECT-CT with ^{99m}Tc hydroxymethylene diphosphonate with significant increased uptake of the tracer in the right TMJ, the right TMJ was considered as end stage disease and a right total joint replacement (TJR) using a Zimmer Biomet custom made prosthesis was performed. A left TJR was performed 2 years later.

Introduction

Osteoarthritis (OA) is the most common form of arthritis affecting the human body. It is characterized by gradual degradation of the cartilage with collapse and resorption of underlying articular bone. It ultimately results in severe morphological changes of the condylar head and inflammatory reaction in the disc and synovial tissues.

In the early stage of this disease, there are no changes seen on X-ray. It is generally accepted that the sensitivity and specificity of an orthopantomogram (OPG) is too low to detect early signs of OA of the temporomandibular joint (TMJ). MRI of the TMJ is the gold standard to diagnose defects in the cartilage and displacement of the disc, but osteoblastic activity or bone remodelling in the underlying bone cannot be evaluated with this imaging modality.

In that respect, ^{99m}Tc-labeled diphosphonates used for bone scintigraphy are incorporated by chemisorption on the surface of hydroxyapatite crystals of the bone, and the uptake of this tracer mainly depends on osteoblastic activity and to a lesser extent, to regional blood flow related to inflammation induced hyperaemia. The introduction of hybrid imaging, combining tomographic scintigraphic imaging (SPECT) with anatomical imaging (CT) allows a more precise localisation of the scintigraphic findings and hence to a higher accuracy. SPECT-CT has been proven to be very valuable in detecting altered bone metabolism in a patient group suspected of OA of peripheral extremities [1]. Kim et al. [2] evaluated the usefulness of bone SPECT-CT imaging in the diagnosis of osteoarthritis of the TMJ by depicting higher uptake ratio in the condyle of patients with osteoarthritis.

In this case report, a young female patient with HLA

B27 positive spondyloarthropathy of the TMJ presented with poor initial outcome after arthroscopy and open joint surgery. Prior to total joint replacement (TJR) of the condyle, bone SPECT-CT was performed to detect increased bone metabolism in the condyle. The imaging findings on bone SPECT-CT were correlated to the histological findings on the resected condyle.

Case Report

A 30 year old female patient presented to the clinic in 2009 with severe bilateral TMJ pain, restricted mouth opening (MO) of 29 mm and tenderness over her masseter muscles bilaterally. She was initially diagnosed with Wilkes Stage IV of the right and left TMJ. Conservative treatment with nonsteroidal anti-inflammatory drugs (NSAID) and physiotherapy failed. After 3 months, a diagnostic arthroscopy was performed bilaterally and revealed apparent overall fibrosis of the capsule. Postoperatively, she developed increasing pain and tenderness over both joints which remained unresponsive to NSAID, paracetamol and methylprednisolone 16 mg.

After 3 months, arthroscopy of both TMJs was repeated, demonstrating grade IV synovitis. Rheumatological counselling revealed a positive test for HLA B 27 and the diagnosis was made of spondyloarthropathy of the TMJ without involvement of other joints. Whole body bone scintigraphy with SPECT-CT showed no evidence for sacroiliitis but revealed increased tracer uptake in the right TMJ. Lab results showed no anaemia or raised ESR. The patient was treated with salazopyrine 100 mg twice a day without success. There was no improvement in pain levels in the right TMJ and further deterioration in MO to 19 mm. In September 2010, an open joint procedure with plication of the right TMJ articular disc was attempted without noticeable improvement in pain levels or MO. Over the subsequent months and years, the patient was seen by a neurologist, rheumatologist and maxillofacial surgeon, and was treated unsuccessfully with methylprednisolone, pregabalin 75 mg, amitriptyline 50 mg and methotrexate 15 mg.

She was eventually referred for further assessment and treatment. A repeat open joint disc repositioning procedure with an Arthrex corkscrew micro-anchor was attempted but proved unsuccessful. MO was 19 mm postoperatively with pain scores of 10/10 on the visual analogue scale for pain. She was on continuous oxycodone 10 mg twice a day.

In 2014, cone beam CT of the TMJ showed discrete

sclerosis of the right TMJ. MRI of TMJ showed anterior luxation of the disc and mild degradation of the cartilage. Bone SPECT-CT showed significantly increased tracer uptake in the right TMJ predominantly affecting the condylar head. No increased tracer uptake was noticed in the left TMJ (Figure 1) at that time.

Figure 1: Bone SPECT-CT showing significantly increased (>50%) tracer uptake in the right TMJ predominantly in the condylar head.

Based on this imaging finding and the clinical deterioration, the diagnosis of a drugs refractory end stage OA of the right TMJ was made and a right total joint replacement (TJR) using a Zimmer Biomet custom made prosthesis was performed. The procedure was uneventful, producing a highly significant improvement in pain levels and function, with a MO exceeding 30 mm.

Histopathological analysis of the resected condyle showed fragmentation of the articular cartilage with an irregular surface contour and remodelling of the subchondral bone, (grade IV according to the classification of Toller (Figure 2) and confirming end stage TMJ disease.

Figure 2: Histopathological analysis of the specimen of the condyle showing fragmentation of articular cartilage with an irregular surface contour and remodelling of the subchondral bone. An occasional intra-osseous subchondral cyst is seen.

There was further deterioration of the left TMJ approximately 1 year after the right TJR. A discectomy was subsequently performed in February 2016; this relieved her symptoms for 3 months but without overall improvement in pain levels. A left TJR was therefore performed 6 months later. The patient recovered fully with complete resolution of her pain and a stable MO of 35 mm. She was reviewed in clinic in September 2017 and remains symptom free.

Discussion

Treatment of temporomandibular joint (TMJ) disorders can be a challenging condition for patient and surgeon. This case highlights such a challenge.

The patient was initially treated for internal derangement of the TMJ and later diagnosed with HLA B27 positive spondyloarthropathy, affecting only the TMJ. Although arthritis of the TMJ is often involved in systemic inflammatory arthritic disease, it is rarely the initial symptomatic manifestation of the disease. In a large cohort study of paediatric rheumatology centers, 55 patients with isolated TMJ arthritis were described [3]. TMJ arthritis can be the first and only manifestation of inflammatory arthritic disease. Those patients are seen by

dentists or practitioners unfamiliar with the condition and are often erroneously treated for internal derangement of the TMJ or idiopathic condylar resorption.

In all racial groups HLA B27 antigen is associated with ankylosing spondylitis. In the United Kingdom, HLA B 27 is present in 90 to 95% of patients with ankylosing spondylitis [4]. The incidence of TMJ involvement varies between 3% and 21%. Often there is severe destruction of the TMJ with ankylosis. Worldwide, only 8 cases of bilateral TMJ ankylosis have been described [5] and only 1 of these was a case of fibrous ankylosis similar to this case report. On the other hand, this case report is unusual because there are no other signs of the disease like rigidity of the cervical spine or “bamboo spine”, raised ESR and sacroiliitis.

Since the TMJ is a synovial joint, it can be affected by any rheumatological disease. The ongoing synovial activity in these joints will make successful treatment of the TMJ complaints in this group of patients more challenging than in patients with no rheumatic disease. Therefore, a different approach has to be considered.

When disease modifying anti-rheumatic drugs are failing to relieve TMD symptoms, a minimal invasive approach can reasonably be adopted.

Injection of corticosteroids into inflamed joints has been used for decades. In a younger patient with juvenile idiopathic arthritis, intra-articular injection into the TMJ may be considered although there is little evidence to suggest improvement of MO or reducing pain levels. There are still concerns that the beneficial short-term anti-inflammatory effect could be outweighed by the effect on further condylar growth in children in the longer term. In children with juvenile idiopathic arthritis, intra-articular steroid injection caused worsening of the arthritic changes in 15 patients (32%).

The ongoing synovial activity in these joints can explain the poor response to minimally invasive techniques like arthroscopy of the upper joint compartment which will cause only transient pain relief. In a recent publication on the outcome of temporomandibular joint arthroscopy, rheumatic disease [6], restricted MO and bilateral muscle pain correlated significantly with a poor outcome. All of these clinical signs were present in this patient.

The cartilage of the condyle and the synovium of the lower joint compartment will be affected by the

inflammatory arthritic disease. In addition, there is evidence from a cadaver study [7] of a greater prevalence of morphological changes in the inferior surface of the disk of the TMJ. Although recently reported in 1 patient [8], arthroscopy of the lower joint compartment is extreme demanding due to possible damage to the cartilage of the condyle during puncturing of the compartment. In general, an open procedure of the TMJ will more easily allow access to the lower compartment to correct bony deformity of the condyle or remove hyperplastic synovium. There is however no clear evidence in the literature of the efficacy of this procedure in relieving pain in rheumatic disease. Since the aetiology of the joint destruction is inflammatory in nature and thus different from non-inflammatory joint disease, as seen in osteoarthritis, the next step in surgical intervention in patients with inflammatory arthritic disease of the TMJ will be total joint replacement (TJR). At this stage, the use of bone SPECT-CT may be helpful in detecting increased bone metabolism in the condyle, as illustrated in this case.

Although TJR in these patients can be expected to be more complicated by altered anatomy, inflamed tissues, ankylosis and use of systemic medications, O'Connor et al. [9] reported excellent results of TJR of the TMJ in 46 patients with inflammatory arthritic disease with significant improvement in pain, mouth opening and dietary function, and good long-term outcomes up to 104 months in 14 patients.

There is evidence that the outcomes of TMJ arthroscopy are better in patients treated in the early phase of their disease than in patients treated in a later phase [10]. Long term "conservative" non-surgical treatment is not conservative since the intra articular disease will progress with more pain and restricted MO due to fibrosis. In addition, neuropathic pain will compromise the outcome of any surgical procedure.

It follows that early intervention of TMD, including in patients with inflammatory arthritic disease, may be the most appropriate treatment.

Long term conservative treatment of these patients will ultimately result in a chronic pain condition due to central and peripheral sensitization. Due to this condition, anxiety and depression may develop and both are well known to promote even further, the perception of pain.

In this case report, the correlation between histopathological features of a condyle with end stage

cartilage degeneration and the imaging findings on bone SPECT-CT is reported for the first time, and confirms the potential of bone SPECT/CT for the diagnosis and evaluation of degenerative changes of the TMJ, reported in previous studies highlighted in this case report. Paul et al. [11] studied the correlation of the histopathological features of end stage ankle osteoarthritis and bone SPECT-CT images. In SPECT positive specimens, a number of histological characteristics were described: a specific osteoblast-mediated bone formation in the absence of functional osteoclasts and increased cellularity and collagen deposition in bone marrow. ^{99m}Tc-labeled diphosphonates bind selectively to mineralized bone and not to organic bone material which supports the observation of increased tracer uptake and the number of osteoblasts.

The clinical value of bone SPECT/CT in the diagnostic work-up of OA of the TMJ warrants further investigation and has to be viewed together with a careful clinical examination and additional OPG x-ray, CBCT and MRI imaging of the condyle.

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