Case Report
Surgical orodental implications in ankylosing spondylitis

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ABSTRACT
Temporomandibular joint and the pelvic complex are bidirectionally related. Ankylosing spondylitis (AS) is a seronegative arthropathy with the key feature of bony fusion of lumbar vertebrae. A 39 year old known case of AS was presented to private office for left lower impacted third molar surgical removal. Previously, he was rejected to receive oral care for pulpectomy and extraction due to limited mouth opening. Prior to the surgery, lateral neck radiography was obtained to exclude any subluxation of fracture of cervical vertebrae. Neck was supported to insure neck stability during surgical forces. In addition, considering consumption of immunosuppressive medications including corticosteroids, procedure was performed with a great care, with attention to higher possibility of infection and fracture. Access to the surgical site was not desirable, though surgery accomplished without any significant event and the patient discharged with routine analgesic and antibiotics recommendation. Sometimes, impaired access to the oral cavity in patients with AS leads to receive suboptimal or minimal orodental care. Long list of dental implications in these patients may be simplified by considering of careful neck and jaw support, applying at least possible forces and great attention to the infection control rules. It is wised to be performed under patient and skilled hands.

Key Words: Ankylosing spondylitis, immunosuppressant, oral surgery, temporomandibular joint

INTRODUCTION
About 200 years AC, Galen first had differentiated a debilitating disease from rheumatoid arthritis, presumably Ankylosing spondylitis (AS).[1] Later in 1961, Bernard Conner described pathologic skeletal changes in AS.[2] AS is a chronic inflammatory disease from the family of seronegative spondiloarthropathies (i.e., negative serum rheumatoid factor) in which, mainly axial skeletal and to a lesser extent peripheral joints are affected. It may also exhibit extra articular manifestations with ophthalmic, cardiac, and renal involvement. Prevalence of 0.1-1% of general population is reported, with male dominancy (male to female ratio 1:3).[3] Typically, a young 20-40 year old male person is affected. Average age of diagnosis is around 23-28 years with a 5.3-11.4 years delay from the onset of symptoms.[4,5] A strong genetic predisposition with environmental modulation is described. Also, it is proposed that hormonal imbalance may have a crucial role to change the pro-and anti-inflammatory cytokines.[6] More than 92% cases with SA are HLA-B27 positive. This is variable in different races and geographical locations. HLA-B27 positive rates are 8%, 40%, 50%, and 80% in healthy white, healthy African American, African American with AS, and Mediterranean Asians with AS, respectively.[3,7] Notably, only 5% of HLA-B27 positive population progress to AS.[8] A common initial complaint is a radiating low back pain to the buttocks that is alleviated by NSAIDs and exercising.[9] On examination, sacroiliac joints are tender and pain radiates to the back thigh.

A key pathologic change is an ongoing inflammatory process at ligaments and tendons insertion sites with ultimate fibrosis and ossification, so called
enthesis.\textsuperscript{13} Progressively, along with ossification induction at fibrous annulus of vertebrae a bony bridge forms and fusion occurs (bamboo spine). This adversely limits spinal flexion and extension that become worsen as disease progresses. Importantly, temporomandibular joint may be involved with consequent ligament ossification, bony erosions, condylar flattening, disk destruction, and reduced range of motion.\textsuperscript{10} Although true ankylosis is rare and to date ten cases are reported\textsuperscript{11,12} yet subjective symptoms and complaints are frequent and may be present in up to 60% of cases.\textsuperscript{13} Reduced mouth opening, osteoporosis, possible atlantoaxial subluxation, and impaired immune system due to immunosuppressive medications may be challenging to any maxillofacial surgeon when he or she decides to perform even a simple extraction or biopsy from a discolored plaque within the oral cavity. We aimed to highlight especial concerns with emphasis on dentistry considerations for minor oral surgeries in AS cases.

**CASE REPORT**

A 39-year-old Iranian male patient, a known case of AS since 15 years ago, referred to our clinic for his impacted mandibular wisdom tooth. He was operated for lumbar scoliosis and he had total hip replacement. He took 25 mg indomethacin daily, calcium 1000 mg daily, metothrexate 2.5 mg 4 days a week, prednisolone 5 mg daily, and folic acid 1 mg daily. There was no other systemic illness except mentioned including negative history of uveitis. Laboratory data revealed a positive HLA-B27, \textsuperscript{1}st hour ESR = 24 and \textsuperscript{2}nd hour ESR = 50, +++ CRP and mild hypochromic RBCs. Other data were within normal limits. Last bone densitometry was performed 10 years ago which was not significant regarding osteopenia or osteoporosis, though recent lateral neck radiography was suggestive of osteopenia of cervical vertebrae [Figure 1]. Oral panogram (OPG) revealed a left mandibular mesio-angular third molar [Figure 2]. Disk space was reduced and the borders of glenoid fossa and condylar head were sclerotic, yet no erosion and osteophyte was evident [Figure 3]. Mandibular border thickness was within normal limits (4-5 mm thick).

In oral examination, patient had class I angle occlusion. Mandible was limited in laterusive movement. No click or tenderness was noted examining temporomandibular joint. A 26-mm of maximum mouth opening was clarified. There were no other significant soft-and hard-tissue pathoses.

Figure 1: Lateral neck radiography revealed no fracture and subluxation of cervical vertebra

Figure 2: Impacted mandibular mesio-angular third molar

Figure 3: Right temporomandibular joint. Note reduced space and sclerotic border of glenoid fossa and condylar head
A lateral neck X-ray was taken prior to the operation to detect any possible luxation of fracture of the cervical vertebrae which was normal. He got anesthetized using total 4 carpules each containing 1.8 cc of 2% lidocaine with 1:100,000 epinephrine solution. Patient had moderate limitation in cervical flexion and extension and a cushion was inserted behind his head to prevent subluxation or fracture of upper cervical vertebrae while the surgeon exerts unintentional heavy forces during the surgery. During a routine procedure, a flap was elevated and impacted right mesio-angular third molar was removed. A great care was made to avoid any heavy forces and rapid movement of the jaws and neck with regards to antiseptic and low traumatizing maneuver. Afterward, the surgical site was sutured and packed. Surgery was uneventful except for difficulty in accessing the site due to the limited range of mouth opening. The patient was discharged with prescriptions for oral analgesic and 400 mg cefixime for 7 days and 4 mg dexamethasone for injection.

DISCUSSION

A case of AS is presented with emphasis on orodental pre-and intraoperative concerns. Our patient did not complaint about temporomandibular joint (TMJ) problem and had normal occlusion of class I angle classification and canine rise anterior guidance. Attention to the occlusion and teeth intercuspation is of a great interest. A new emerging theory of bidirectional cause–effect relationship between temporomandibular/cranio mandibular and pelvic systems is described. Evaluating 100 patients with SA, Weneberg in 1982 concluded that severity of stomatognathic symptoms was correlated with general subjective joint symptoms.[14] Various authorities believe that orodental complex stability dictates postural position and vice-versa is adversely affected by pelvic instability.[15,16] Temporomandibular factors may be attributed to the sacroiliac dysfunction and sprains. [16,17] Fink et al. investigated the effect of artificial dental occlusal interference in healthy individuals and figured out a significant hypomobility of upper cervical spine and sacroiliac joint.[18] A combined orodental rehabilitation with orthodontic/prosthodontic and chiropractic cotreatment is promising to improve both TMJ and pelvic pain and function.[10,16] Prior to each oral manipulation, obtaining a lateral cervical radiography is wised. This simple radiography gives informative clues about cervical fusion, atlantoaxial subluxation, and relative bone density. In the case of suspicious findings, a neurologic consult and further radiologic examinations are recommended. Protecting head in a slight flexion by cushion put behind the head and avoiding maneuver with extensive neck flexion and extension is critically important. Another barrier to appropriate access into the oral cavity is limited mouth opening as a consequence of damaged TMJ. A careful manual examination with further computed tomography imaging would provide the clinician to how extent the TMJ is damaged secondary to AS.[11] Simple X-rays and even CT and MRI imaging remain intact early in the course of disease. A stand up MRI is valuable to detect cervicothoracic injuries.[19] Joint fissure, pseudodilation, periaricular osteosclerosis, ligament ossification, or early ankylosis without predestruction may be evident in imaging.[13] In addition, tissue samples may reveal vascular proliferation, leukocyte infiltration, or disk destruction, which is not contrasting pathologic features from other inflammatory diseases of TMJ.[20]

Moreover, clinician should be aware of increased risk of bleeding [nonsteroidal anti inflammatory drug (NSAIDs) consumption], fracture, and infection (corticosteroids and immunosuppressive medications). Generally, treatments include medication, surgical manipulation and physical therapy. Among medications, NSAIDs are prescribed more often. More specific biologies such as infliximab and anakinra that targets TNFα and interleukin 1 receptor respectively may be used when previous medications are failed.[3,21] Other medications with various success and conflicting results are azathioprine, methotrexate, cyclophosphamide, cyclosporine, and topical and systemic corticosteroids.[2] Such medication put the patient at the risk of serious infection. A case of mandibular osteomyelitis following tooth extraction in a patient with AS taking Infliximab is reported.[21] There are initial successful diskectomy and synovectomy results.[22] More recently, a bilateral alloplastic TMJ replacement may be helpful in the case of ankylosis and severe disk destruction.[23]

Antigenic mimicry between HLA-B27 and Klebsiella pneumonia raise concerns that individual tendency to express manifestations of AS in heavily modulated by intestinal bacterial load.[24] Meanwhile Toivanen et al. argued this possible microbial role in exciting immune and inflammatory cascades.[25] It has been postulated that eliminating starch from daily diet as a prime
nutrient for Klebsiella may be promising, yet needs further confirmatory investigations. Moreover, a 12-week trial of moxifloxacin conferred significant and sustained improvement in inflammatory signs of AS. The possible mechanism is antibacterial property of such medication. Besides, slow-stretching masseteric exercises could increase mouth opening. Generally, local anesthetics and conscious sedation are safe for patients with AS, while general anesthesia is not recommended because of the following reasons: 1 – cervical fusion prone this part of vertebrae to fracture and serious neurologic impairment. 2 – Osteopenia or osteoporosis resulting from either long standing disease or corticosteroid consumptions increases the risk of fracture. 3 – Limited mouth opening and calcified ligaments make the intubation difficult. Hence, forceful intubation should be avoided to prevent luxation or fracture of cervical vertebrae. 4 – Stiff thoracic cage due to acromioclavicular and sternoclavicular joint damages decreases the efficacy of ventilation.

Up to 10% of patients have aortic regurgitation which needs endocarditis antibiotic prophylaxis prior to invasive intraoral procedures. Long standing disease may predispose to secondary amyloidosis with resultant renal failure. An impaired renal function test should hazard the clinician to adjust certain antibiotics and analgesics.

CONCLUSION

Conclusively, prior to office oral surgeries in cases with AS, a lateral neck radiography should be taken to ensure stability of cervical vertebrae. In addition, clinician should always remember the systemic consequences of AS and specifically modify any surgical or nonsurgical remedies according to the concurrent direct or indirect comorbidities.

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