

## CASE REPORT



## Orthognathic Surgery in a Patient with Multiple Sclerosis

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### ABSTRACT

**Aim:** The aim of this paper was to report a case of orthognathic surgery successfully done in a patient with multiple sclerosis.

**Background:** Multiple sclerosis (MS) is a chronic, progressive inflammatory disorder of the central nervous system affecting young adults, characterized by lymphocytic infiltration of the brain and spinal cord leading to demyelination and focal axonal damage. Clinically, MS patients present with reversible neurological dysfunction in the early stages, which progresses to irreversible neurological disability and deficit. Oral manifestations of MS include facial numbness or pain, neuralgias, facial paralysis, dysarthria and dysphagia. While dental treatment is not contraindicated in MS patients, it is, however, limited to preventive and supportive dental care.

**Case description:** A 23-year-old Saudi male patient with a diagnosis of MS since 2008 reported to the oral and maxillofacial surgery (OMFS) department for correction of dentofacial deformity. The patient was under follow-up with the neurology department and was being treated with interferon beta-1a. Following consent from the neurologist and the patient, a Lefort 1 segmental osteotomy was done under general anesthesia. The patient was stable throughout the surgical procedure and during the postoperative period. The patient was discharged upon complete surgical recovery and no acute exacerbations of MS were reported during the perioperative period.

**Conclusion:** Based on our observations, orthognathic and maxillofacial surgical procedures can be safely carried out in patients with MS, provided a strict perioperative prophylactic regimen for stress reduction and prevention of acute attacks of MS is adhered to.

**Clinical significance:** Due to the stressful nature of dental treatment and oral and maxillofacial surgical procedures, acute exacerbations of MS are very much likely. Hence, it is imperative

that dental and oral surgical practitioners are aware of the manifestations of MS and are able to manage such patients with suitable treatment modifications.

**Keywords:** Multiple sclerosis, Orthognathic surgery, Dentofacial deformity, Interferon beta-1a, Lefort 1 osteotomy

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### BACKGROUND

Multiple sclerosis (MS) is a chronic, progressive inflammatory disorder of the central nervous system affecting young adults and more commonly males.<sup>1,2</sup> It is characterized by lymphocytic infiltration of the brain and spinal cord leading to demyelination and focal axonal damage.<sup>1-3</sup> While the etiology of MS is not precisely known, studies have reported autoimmune etiopathogenesis along with a pre-disposition to environmental factors, viruses and heredity.<sup>2,4-6</sup> Over the course of the disease, there is transient remyelination of the axons, which however, is not enduring due to the chronic inflammatory nature of the disease.<sup>1</sup> Clinically, MS patients present with reversible neurological dysfunction in the early stages, which progresses to irreversible neurological disability and deficit with time.<sup>1,2</sup> Clinical complications of MS include motor organ dysfunction, sensorineural deficits, like paresthesia, dysesthesia and numbness in addition to cognitive disability and behavioral problems in about 10% of patients.<sup>4,7-9</sup> Diagnosis of MS is based on history, clinical, laboratory and radiographic findings.<sup>2</sup>

Oral manifestations of MS include paroxysmal attacks of facial numbness or pain (neuropathic or neuralgic), facial paralysis, dysarthria (inability to articulate words) and dysphagia (inability to swallow) during the initial stages of the disease.<sup>2,9</sup> In addition, trigeminal neuralgia,

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temporomandibular disorders and paroxysmal orofacial pain syndromes have also been reported during later stages of MS.<sup>1-4</sup> Moreover, lack of motor coordination could affect oral hygiene practices.<sup>2-4</sup> While dental treatment is not contraindicated in MS patients, it is, however, limited to preventive and supportive dental care, preferably delivered during stages of remission.<sup>2</sup> A comprehensive literature search revealed no evidence of association between dental or oral surgical procedures and clinical manifestations of MS. However, stress related to surgical procedures and anesthesia (general, regional and local anesthesia) has been implicated in exacerbation of clinical manifestations and aggravation of existing symptoms in patients with MS.<sup>10-15</sup>

We report a case of maxillary orthognathic surgery done under general anesthesia, for correction of maxillo-mandibular deformity, in a patient with MS. Based on evidences available in the published literature, this is the first report of a case of orthognathic surgery in a patient with MS.

## CASE DESCRIPTION

A 23-year-old Saudi male patient reported to the oral and maxillofacial surgery (OMFS) department at King Faisal specialist hospital and research center (KFSHRC), Riyadh, Saudi Arabia in January, 2013 for correction of dentofacial deformity. Medical history revealed a diagnosis of MS in 2008. The diagnosis of MS was based on 'McDonald criteria', and since then the patient has been under treatment with interferon beta-1a (AVONEX, Biogen Idec, NC, USA)—30 micrograms administered intramuscularly once a week.<sup>16</sup> The patient was under regular follow-up at the neurology department at KFSHRC and had not reported any neurological adverse events related to MS in the last 3 years. General examination of the patient was unremarkable.

Upon dental evaluation, the patient complained of difficulty in chewing and recurrent tongue biting. Clinical examination revealed the following (Figs 1 and 2):

- No obvious extraoral facial asymmetry.
- Concave facial profile with normal appearing malar eminences, orbits and nose.
- Clicking of the left temporomandibular joint on protrusion.
- Upper lip incompetence by 4 mm and a 90° nasolabial angle.
- Intraoral maxillary dental midline coincident with the facial midline.
- Mandibular dental midline shifted by 1 mm to the left of maxillary dental midline.
- Prominent incisal show at rest and smile with an overjet of 2 mm and no occlusal canting.



**Figs 1A to D:** Facial photographs: (A) preoperative frontal facial view, (B) preoperative facial profile view, (C) postoperative frontal facial view and (D) postoperative facial profile view

- Anterior openbite of 5 mm, bilateral posterior crossbite and retroclined lower anterior teeth.
- Class 1 canine and molar relationship on the left side, and class 3 canine and molar relationship on the right side.
- Fair oral hygiene and evidence of recurrent aphthous ulcers.

Cephalometric analysis revealed a high mandibular plane angle along with a facial depth of 91°, SNA–83.5° and SNB–83°.

The patient was referred for orthognathic surgical correction and subsequently a treatment plan was formulated. In February, 2013, pre-surgical orthodontic correction was initiated for maxillary and mandibular occlusal plane leveling, dental alignment and decompensation; without compromising the root divergence between the maxillary canine and 1st premolar teeth. This was to be followed by double jaw surgery involving segmental Lefort 1 osteotomy with differential impaction to correct anterior open bite and transverse discrepancy, along with mandibular setback by bilateral sagittal split osteotomy to follow occlusion and correction of dental midline. Considering the underlying neurological condition of the patient, any prolonged surgical stress could have adversely induced or exacerbated an acute attack of MS. The surgical treatment plan was thus suitably modified to



**Figs 2A to F:** Dental occlusion photographs: (A), (B), (C) preoperative occlusion and (D), (E), (F) postoperative occlusion

minimize operating time and surgical insult to the patient. Accordingly, it was decided to avoid mandibular surgery without compromising the final esthetic and functional outcomes. This was achievable by a Lefort 1 osteotomy with segmental advancement of 3 mm and impaction of 2 mm, in addition to transverse maxillary expansion by 3 mm and shifting the maxillary midline by 3 mm.

The final treatment plan was explained to the patient and consent was obtained for the surgical procedure in October, 2013. In addition, the surgical treatment plan was discussed with the neurology and anesthesia teams to obtain necessary clearances before surgery. Preoperative laboratory findings were within normal limits. On the day of surgery, the patient was premedicated with cefazolin 1 gram intravenous (IV), dexamethasone 8 mg IV and midazolam 5 mg intramuscular. Under routine monitoring, the patient was induced with etomidate 15 mg, remifentanyl 1 $\mu$ g/kg and vecuronium bromide 7 mg, and naso-endotracheal intubation was done. The patient was mechanically ventilated and anesthesia was maintained with 0.2  $\mu$ g/kg of remifentanyl and sevoflurane at a concentration of 1.5% along with N<sub>2</sub>O and O<sub>2</sub>. Remifentanyl was discontinued prior to surgical wound closure. Following infiltration with lidocaine and epinephrine (xylocaine 2% with epinephrine 1:80,000, Astra-Zeneca, Saudi Arabia), a maxillary buccal vestibular incision was made to expose the maxilla. Lefort 1 segmental osteotomy was done with a surgical saw (Stryker TPS™, Stryker instruments, Kalamazoo, Michigan, USA) under saline irrigation. The maxillary segment was repositioned and fixed with 1.5 mm titanium plates and screws (Matrix orthognathic plating systems, DePuy Synthes, Zuchwil, Switzerland); and mucosal wounds were closed with 4 to 0 vicryl resorbable sutures (Ethicon, Johnson and Johnson, Norderstedt, Germany). The patient was stable

hemodynamically throughout the surgical procedure, which was carried out as planned. The immediate postoperative period was uneventful and analgesia was achieved with non-steroidal anti-inflammatory drugs (NSAIDs). Following recovery, the patient was discharged on the first postoperative day and followed up in the OMFS out-patient clinic after 5 days and once every week for 6 weeks thereafter. The patient was advised to use guiding elastics and a maxillary occlusal stent for 2 weeks and 6 weeks respectively. Following this, the patient was referred for post-surgical orthodontic treatment for settling of occlusion and restorative treatment for closure of spaces between the maxillary anterior teeth.

All through the orthognathic planning and treatment stages, the patient continued his medication for MS. The patient had no aggravation of symptoms or acute attack due to MS during his period of hospitalization. The patient has been under continuous follow-up with the neurology department since then and remains asymptomatic.

## DISCUSSION

Multiple sclerosis is a progressive disease with manifestations ranging from a benign illness to an incapacitating disease.<sup>1,2</sup> Clinically, the course of the disease may be characterized into four major types: (1) the most common relapsing/remitting type with episodic attacks followed by periods of remission; (2) primary progressive type with increasing deterioration of neurological function; (3) secondary progressive type wherein the relapsing/remitting MS changes course to a progressive disease; (4) progressive/relapsing type with increasing deterioration of neurological function aggravated by episodic attacks.<sup>2,17</sup> While acute attacks of MS may occur spontaneously, it has also been reportedly associated with stressful conditions, such as infection, pregnancy and surgical procedures

under anesthesia.<sup>15</sup> Aggravation of clinical manifestations in MS patients following surgical procedures have been reported as early as 1976.<sup>18</sup> The postoperative milieu of stress, infection and inflammation rather than the surgical procedure has been considered as the reason for aggravation of MS symptoms.<sup>1,10,11,15</sup> Similarly, anesthesia, irrespective of whether it is regional, local or general, has also been implicated in aggravating clinical manifestations of MS.<sup>12-15</sup> Nevertheless, it remains unexplained, whether the exacerbation in symptoms is related to the anesthetic agent or the stress of the procedure.<sup>15</sup>

There are at least two reports in the literature, which have resulted in acute attacks in previously undiagnosed MS patients following oral surgical procedures under local anesthesia.<sup>12,19</sup> Concomitantly, there are also reports of major surgical procedures done in MS patients ranging from obstetric to orthopedic and cardiothoracic surgeries without any acute exacerbations of the disease.<sup>13-15,20,21</sup> While such procedures have been reportedly done under both general and regional anesthesia, there are studies indicating a preference for general anesthesia over regional or local anesthesia.<sup>13-15</sup> In light of these evidences, it is alluring to hypothesize that the emotional, physical and nociceptive stresses associated with surgical procedures are the major contributing factors for acute attacks, following surgery in MS patients. Furthermore, infection, pain and thermal stimuli due to changes in body temperature have resulted in acute attacks of MS.<sup>13,14,18</sup> All of this reiterates the need for adequate preoperative preparation, premedication and good anesthesia for perioperative stress reduction and prevention of surgery related acute attacks in MS patients.

While intravenous anesthetic drugs like thiopentone sodium have been contraindicated in MS patients, inhalational agents, like sevoflurane and desflurane have been used safely.<sup>13-15</sup> Similarly, drugs like midazolam, propofol, fentanyl, remifentanyl, steroids, opioid analgesics and NSAIDs have been used perioperatively without exacerbating acute attacks of MS.<sup>13-15,21</sup> Even though epidural anesthesia has been administered safely using bupivacaine in MS patients,<sup>14,21</sup> mandibular block anesthesia with articaine has reportedly resulted in an acute attack of MS in one patient.<sup>19</sup> While literature regarding dental local anesthesia and its effect on MS are sparse, there are no reports contraindicating their usage. In the present case, the patient was pre-medicated with midazolam for anxiety and stress reduction, and a good depth of intra-operative anesthesia was maintained with the help of remifentanyl and sevoflurane. Infiltrations of local anesthetic lidocaine with epinephrine helped in blocking pain impulses in addition to vasoconstriction of the surgical site. Postoperative pain and pyrexia were effectively

countered by administration of NSAIDs. Moreover, in order to reduce surgical stress and operating time, the treatment plan was modified to achieve desired results with only maxillary orthognathic surgery.

It has been reported that MS has a high family recurrence rate, indicating a strong genetic predisposition.<sup>1,11</sup> However, there are no evidences of association between MS and dentofacial deformities. In the present case, the patient reported with severe maxillo-mandibular deformity in all the three planes, which required multimodality treatment in the form of orthodontics and orthognathic surgery. In a study evaluating hospitalization following orthognathic surgery, it was found that the length of hospitalization was directly related to the severity of the dentofacial deformity and complexity of the surgical procedure.<sup>22</sup> This, not only implies the degree of operative stress endured by orthognathic surgery patients, but also the risk of acute exacerbations due to orthognathic surgery in a patient with MS. Dickerman et al proposed a perioperative prophylactic regimen to avoid adverse events in MS patients undergoing surgical procedures; essential components of which include—premedication for anxiety reduction, prophylactic antibiotic and anti-inflammatory medications, smooth anesthetic induction and adequate depth of anesthesia.<sup>10</sup> Moreover, studies have suggested making a choice of general anesthesia over local or regional anesthesia whenever possible, while surgically treating MS patients.<sup>2,10</sup>

An estimated 2.5 million people are affected with MS worldwide, with a prevalence of 90 per 100,000 population.<sup>1</sup> With a prevalence of 25 per 100,000 population, MS is not commonly seen in Saudi Arabia.<sup>17</sup> Medical management of MS involves multi-modality treatment. While episodic attacks are symptomatically treated with steroids, benzodiazepines and centrally acting muscle relaxants; long-term treatment is primarily done by immune-modulant therapy using mitoxantrone (anti-neoplastic agent), interferon- $\beta$ , natalizumab (monoclonal antibody) and glatiramer acetate.<sup>1,2</sup> Although there is a risk of drug interactions in MS patients undergoing dental or surgical treatment, there were no such issues in the present case as our patient was under interferon therapy only.<sup>2</sup> Moreover, the neurology team was taken into confidence while performing all the treatment procedures.

## CONCLUSION

Maxillofacial surgery in MS patients is relatively uncommon, and to the best of our knowledge, this is the first time a case of orthognathic surgery in a patient with MS is being reported. Based on our observations, orthognathic and maxillofacial surgical procedures can be safely carried



out in patients with MS, provided a strict perioperative prophylactic regimen for stress reduction and prevention of acute attacks of MS is adhered to. Nevertheless, long-term multicenter studies of maxillofacial surgical treatment done in MS patients are required to formulate specific guidelines for their judicious management.

### CLINICAL SIGNIFICANCE

Multiple sclerosis is associated with a variety of orofacial manifestations. While there are prescribed guidelines for the general surgical management of MS patients, no such guidelines are available for oral, dental or maxillofacial surgical treatment.<sup>1,10</sup> Owing to the stressful nature of dental treatment and oral and maxillofacial surgical procedures, acute exacerbations of MS are very much likely. Hence, it is imperative that dental and oral surgical practitioners are not only aware of the manifestations of MS, but are also able to manage such patients with suitable treatment modifications.<sup>2,3</sup>

### Ethical Approval

Ethical approval for this case report was obtained from the ethics committee at KFSHRC, Riyadh, Saudi Arabia. Furthermore, patient consent was obtained for utilizing his photographs and dental records.

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