Neuropathic pain after orthognathic surgery

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Objective. This study assesses the incidence of neuropathic pain after orthognathic surgery at our center and briefly reports the cases found.

Study Design. All records of orthognathic surgical patients between 2001 and 2011 were exported from the hospital information system into a Portable Document Format platform (Adobe Acrobat) to simplify keyword searching. Records of patients that developed debilitating chronic pain were isolated and examined.

Results. Records included 982 bilateral sagittal split osteotomies, 536 LeFort I procedures, and 335 surgically assisted rapid palatal expansion procedures. Six records were identified that described cases in which patients developed debilitating chronic neuropathic pain after orthognathic surgery (mean age at surgery, 43 years).

Conclusions. The exposure of the inferior alveolar nerve or partial axonal injury together with a disruption of the bony environment of the inferior alveolar nerve is a risk factor after bilateral sagittal split osteotomy. (Oral Surg Oral Med Oral Pathol Oral Radiol 2014;117:e102-e107)

Neuropathic pain after orthognathic surgery

Neuropathic pain of the IAN is a disabling condition that interferes with speaking, eating, drinking, kissing, shaving, applying make-up, and tooth brushing.2,3 After iatrogenic damage of the IAN, neuropathic pain severely affects the quality of daily life, often leading to litigation and patient complaints about their treatment.4 Renton and Yilmaz5 analyzed 90 patients with iatrogenic IAN injuries and 90 patients with iatrogenic lingual nerve (LN) injuries; all injuries resulted from an oral and maxillofacial procedure (dental implants, third molar surgery, endodontic treatment, or local anesthesia). Approximately 70% of their patients presented with neuropathic pain concurrent with anesthesia or paresthesia, and all patients exhibited neuropathy with varying degrees of loss of mechanosensory function, paresthesia, dysesthesia, allodynia, and hyperalgesia. In a similar review, Jääskeläinen et al.4 observed a 45% incidence of neuropathic pain in 58 patients with iatrogenic sensory deficits of the IAN and LN. Teerijoki-Oksa et al.6 prospectively followed 19 patients after BSSO and observed a 5% overall occurrence of neuropathic pain at 1-year follow-up, which is similar to the overall estimated incidence of neuropathic pain after traumatic and iatrogenic nerve injuries.7 In contrast, Borstlap et al.8 prospectively followed 222 patients after BSSO and reported no incidence of neuropathic pain. In the literature, information about neuropathic pain after orthognathic surgery is sparse. Therefore, we performed a retrospective review of the orthognathic population at our center to establish the incidence of neuropathic pain after orthognathic surgery and to briefly report the cases that we found.

Materials and Methods

All records of orthognathic surgical patients between 2001 and 2011 were exported from the hospital information system into a Portable Document Format platform (Adobe Acrobat) to enable keyword searching. These records included 982 BSSO procedures, 536 LeFort I procedures, and 335 surgically assisted rapid palatal expansion (SARPE) procedures. We examined all records in which the word “pain” was encountered during the postoperative course to isolate those records.
in which patients developed debilitating chronic pain that interfered with their professional, social, or psychologic well-being.

RESULTS

Six records were identified that described cases in which patients developed debilitating chronic neuropathic pain after orthognathic surgery. The diagnosis was based primarily on history and findings on physical examination.9,10 The surgical and demographic characteristics of these subjects are presented in Table I.

No case of neuropathic pain was observed as a result of upper jaw surgery, after a LeFort I osteotomy, or after a SARPE procedure. Neuropathic pain only developed after a surgical procedure in the lower jaw.

Patient 1 underwent BSSO advancement. The operative chart indicated that it was difficult to free the IAN from the proximal fragment bilaterally. Although no nerve transection was noted, the operative chart indicated that the nerve had some degree of axonal damage. The patient developed severe bilateral hypesthesia, which became disabling and painful at the left side after a few months. Based on the hypothesis that the pain might be due to compression, a reoperative procedure was performed on the left side after more than 12 months; however, this procedure did not improve the patient’s symptoms. Hyperbaric oxygen therapy was administered but did not provide relief. The patient was sent to different tertiary centers for additional evaluations, but no improvement was achieved. The patient was contacted 10 years postoperatively, the patient still felt pain and rapid development of severe depression. Pharmacologic management of the condition proved unsatisfactory, as did 20 sessions of hyperbaric oxygen. This patient was also a smoker and had a preoperative temporomandibular joint dysfunction at the left side. She had reoperative procedures several times at other centers without relief. Finally, the clinicians at one center decided to resect the part of the mandible between the mental foramen and the angle of the left mandible without reconstruction. At the last follow-up, 5 years postoperatively, the patient still felt pain and "attacks of needles" at the left side. She also felt tired rapidly after talking. The occlusion shifted after the partial mandibulectomy, causing temporomandibular joint problems on the contralateral side.

Patient 2 developed painful hypoesthesia at the chin after BSSO. This condition interfered with his professional activities as a clarinetist, causing

<table>
<thead>
<tr>
<th>No.</th>
<th>Gender</th>
<th>Surgical site of lesion</th>
<th>Year of surgery</th>
<th>Age at surgery</th>
<th>Bone healing</th>
<th>Intraoperative nerve condition</th>
<th>Innervation territory</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>M</td>
<td>BSSO</td>
<td>2001</td>
<td>32</td>
<td>Normal</td>
<td>Axonal damage L+R</td>
<td>IAN L</td>
</tr>
<tr>
<td>2</td>
<td>M</td>
<td>Mandibular bone graft</td>
<td>2004</td>
<td>53</td>
<td>Normal</td>
<td>Normal</td>
<td>All over the face</td>
</tr>
<tr>
<td>3</td>
<td>W</td>
<td>BSSO</td>
<td>2007</td>
<td>48</td>
<td>Defect; pseudarthrosis</td>
<td>Elongation + axonal damage L</td>
<td>IAN L</td>
</tr>
<tr>
<td>4</td>
<td>M</td>
<td>BSSO</td>
<td>2008</td>
<td>43</td>
<td>Normal</td>
<td>Axonal damage L+R</td>
<td>IAN L+R</td>
</tr>
<tr>
<td>5</td>
<td>W</td>
<td>BSSO</td>
<td>2011</td>
<td>54</td>
<td>Defect; infection</td>
<td>Normal</td>
<td>IAN L+R</td>
</tr>
<tr>
<td>6</td>
<td>W</td>
<td>BSSO</td>
<td>2011</td>
<td>28</td>
<td>Defect; pseudarthrosis</td>
<td>Normal after detachment</td>
<td>IAN L</td>
</tr>
</tbody>
</table>

BSSO, bilateral sagittal split osteotomy; IAN, inferior alveolar nerve; M, man; W, woman; L, left side; R, right side.
distress and depression. Hyperbaric oxygen therapy resulted in sufficient improvement, although it did not completely relieve his symptoms. The patient also obtained relief with antidepressants and antiepileptic drugs. He regained his capacity to play the clarinet, but he still had difficulty drinking liquid, owing to loss of temperature perception in the lower lip. The operative chart described a difficult BSSO, with the nerve close to the lower and buccal borders. This patient was a nonsmoker. Remarkably, the width of the mandibular canal had decreased at the 12-month postoperative panoramic radiograph (Figure 2).

Patient 5, a nonsmoker, underwent a technically unremarkable BSSO procedure with a large advancement. Nearly full recovery of sensation was reported in the weeks after the procedure. Perioperatively, the nerve did not need to be detached from the proximal segment of the mandible; it was suspended in the osteotomy gap without the protection of the mandibular canal. No visible nerve damage was perceived during the surgical procedure. Because the mesiodistal and buccolingual gap was large, it was decided to fill the gap with HydroSet (Stryker, Kalamazoo, MI, USA), which is an injectable, sculptable, and fast-setting bone substitute (calcium phosphate cement) that converts to hydroxyapatite (Figure 3). The patient developed bilateral osteomyelitis, which was complicated by chronic incapacitating pain that did not respond to medication. Hyperbaric oxygen did not improve the infection but did improve the pain. Surgical debridement was performed several times, but no final pain relief was observed. After surgical debridement with plate removal and removal of the hardened cement, the patient developed a hypoesthesia that became permanent. Two years after surgery, the patient reports satisfactory pain relief in the mandible but describes pain at the temporomandibular joints.

Patient 6 underwent an uneventful BSSO procedure with a large advancement. Although a freer was used to free the nerve from the proximal fragment, the operative chart described no particular difficulty. This patient was a heavy smoker and a bruxist; she developed an infection of the osteosynthesis plates, which were removed 3 months after BSSO. The occlusion at the left side progressively shifted to become a class II malocclusion, which was accepted by the patient. It was
initially thought that the infection caused the pain at the left lower jaw, but this pain persisted after plate removal; it became disabling, and a sensory impairment developed at the left side of the lower jaw. Cone beam computed tomography (CBCT) revealed a pseudarthrosis at the left side. Successive panoramic radiographs revealed an upward rotation of the proximal fragment (Figure 4). CBCT confirmed the left-side pseudarthrosis. An early reoperative procedure and bone grafting of the affected side with iliac crest bone led to significant improvement of all symptoms.

DISCUSSION

In the present study, the 0.51% incidence (5/982) of neuropathic pain after BSSO was far lower than the incidences of postoperative neuropathic pain observed after other surgical procedures outside the neurosensory distribution of the trigeminal nerve. This incidence was also far below that reported by Jääskeläinen\(^4\) after BSSO. Benoliel et al.\(^1\) attribute this relative resistance of the trigeminal nerve to trauma-induced hyperactivity to the fact that the trigeminal nerve displays significantly less ectopic discharge than other peripheral nerves. This difference might explain the low incidence of neuropathic pain in the trigeminal system.

Walter and Gregg\(^1\) followed 36 patients who had a sagittal split osteotomy (13 patients), an intraoral vertical ramus osteotomy (10 patients), or a mandibular vestibuloplasty (13 patients). One patient (1/36; 2.8% of the total group) developed a neuropathic pain condition after an intraoral vertical ramus osteotomy. In this series, out of the 10 patients with an intraoral vertical ramus osteotomy, 7 (70%) experienced acute postoperative paresthesia and continued to have paresthesia at the 6-month postoperative follow-up. One developed neuropathic pain.

Steinhauser\(^1\) reexamined the long-term results of 282 patients after a vestibuloplasty in the lower jaw with skin graft and reported that 1% of the patients experienced “neuralgia pains.”

In 5 out of 6 patients, the neuropathic pain arose as a direct consequence of a lesion or disease affecting the somatosensory system.\(^14\) In patient 2, the diagnosis of neuropathic pain was less clear-cut, because the reported sensory abnormalities extended far beyond the neuroanatomic level. Neurologic investigation pointed to a psychogenic pain condition rather than a neuropathic pain state.

In the present series, late reintervention did not appear to improve patients’ pain, with the exception of patient 5, in whom early correction of pseudarthrosis with an iliac crest bone graft immediately improved the developed sensory disturbance and the experience of pain. Yates et al.\(^15\) reported on 41 patients with microsurgical trigeminal nerve repair and found that 11 patients exhibited neuropathic pain before nerve repair. Neuropathic pain persisted in all 11 patients after microsurgical nerve repair. The other 30 patients in the study were pain-free before the microsurgical repair and remained pain-free after the nerve repair. Yates et al. concluded that the presence of neuropathic pain before trigeminal nerve repair is the major risk factor for experiencing postoperative neuropathic pain after nerve surgery, which is in agreement with our present findings that neither additional surgery nor late reintervention on the nerve improved neuropathic pain.

Three patients exhibited a disturbed bone healing together with an intraoperatively exposed IAN, suggesting...
that some degree of axonal damage might trigger the neuropathic pain condition, as well as that the simultaneous presence of a mandibular bone defect sometimes creates the necessary environment to cause neuropathic pain. Early repair of the bony environment in patient 6 proved beneficial.

In patient 5, the nerve was exposed and HydroSet was used. Benoliel et al. reported that perineural inflammation without damage to the axonal nerve trunk elevates spontaneous activity. Consequently, the presence of inflammation alone can induce ectopic activity and spontaneous pain, which might explain the symptoms in this patient. The “inflammatory soup” may directly activate or indirectly sensitize nociceptors. An alternative mechanism to explain the condition in this patient is offered by a similar finding described by Gregg, in whose study perineural calcium hydroxide was associated with severe and refractory dysesthesia.

The BSSO procedure was uneventful in patients 1 and 4. Still, it is possible that some axonal damage occurred during a difficult detachment maneuver. Additional compression of the nerve by the proximal fragment is also possible but cannot be seen on panoramic radiographs. Tay and Zuniga reported on 59 patients referred with 73 injured trigeminal nerves. In their study, neuropathic pain occurred in 14.9% of IAN injuries, and only in those with mild or no sensory impairment. Out of 73 injured nerves, 1 LN and 7 IANs were injured during orthognathic surgery. Out of the 7 IANs injured during orthognathic surgery and examined with clinical neurosensory testing, 2 exhibited no sensory impairment, 3 exhibited mild sensory impairment, and 2 exhibited severe sensory impairment. One of these patients had a painful trigger at the injury site, 2 reported pain, and 2 others had neuropathic pain. Obviously, there is no direct correlation between the extent of the nerve damage and the occurrence of neuropathic pain. The mean age in this patient group was 43 years, which is well above the mean age of our BSSO population (26.3 years).

The diagnosis of neuropathic pain was only established in a timely manner for patients 5 and 6. Because preoperative pain has consistently been a good predictor of persistent postoperative pain, it seems advisable to implement a preventive strategy in these patients to block the pain as rapidly and completely as possible. This must be confirmed for trigeminal neuropathic pain after orthognathic surgery.

In conclusion, the records that we examined revealed that the diagnosis of “neuropathic pain” was often not established in a timely manner; it usually took some time for this diagnosis to be reached. It was apparently difficult for patients to communicate unfamiliar symptom qualities, and it was also apparently difficult for the surgeon to accept neuropathic pain as a working diagnosis. The mean age at surgery was 43 years in the subgroup with neuropathic pain. In comparison, the mean age of the entire patient group was 26.3 years, suggesting that age is a risk factor when combined with axonal IAN damage and possibly a bone deficit. The neuropathic pain after orthognathic surgery was observed only after BSSO, not after maxillary orthognathic surgery. Total nerve transection (neurotmesis) was not an intraoperative finding in any case, although the simultaneous occurrence of an exposed nerve or partial axonal IAN injury together with a disruption of the bony environment of the IAN is a risk factor after BSSO. Early repair of the bony environment may prove beneficial; recognition of the condition after the optimal treatment period of 6 months often yields poor results.

REFERENCES

