

Atypical Facial Pain in Silent Sinus Syndrome

Nirvi Shah* and Sanjog Pangarkar

Department of Physical Medicine and Rehabilitation, University of California Los Angeles/VA Greater Los Angeles Healthcare System, USA

*Corresponding author: Sanjog Pangarkar, Department of Physical Medicine and Rehabilitation, University of California Los Angeles/VA Greater Los Angeles Healthcare System, USA

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Abstract

Silent Sinus Syndrome is a disease process characterized by hypoglobus or enophthalmos secondary to collapse of the orbital floor in the presence of asymptomatic chronic maxillary sinusitis. The prevalence of this disease is unknown, however approximately 100 cases have been reported in the medical literature thus far. Although typically dominated by ophthalmic complaints, we present a patient with post-operative atypical facial pain. The pain was refractory to both conservative and common interventional treatments. In this case report, we present a potential treatment option that has not been described in the literature previously for this condition. IncobotulinumtoxinA was used off-label for treatment of neuropathic, facial pain. More research is needed to explore potential mechanisms of incobotulinumtoxinA, including its possible role in affecting the trigeminal nucleus or blocking autonomic pathways. This treatment was highly effective in not only symptomatically reducing severe, atypical facial pain but also in preventing another future surgical procedure for pain relief.

Keywords: Silent sinus syndrome; Atypical facial pain; IncobotulinumtoxinA; Botulinum toxin

Introduction

Silent Sinus Syndrome is a rare condition characterized by hypoglobus or enophthalmos secondary to collapse of the orbital floor in the presence of asymptomatic chronic maxillary sinusitis. The prevalence of this condition is unknown, however approximately 100 cases have been reported in the medical literature. Although typically dominated by unilateral ophthalmic complaints, such as painless and progressive enophthalmos and hypoglobus, our patient presented with facial pain of unknown origin. The patient was previously seen by multiple subspecialties including Ophthalmology, Oculoplastics and Otolaryngology before presenting to a multidisciplinary tertiary care pain clinic. In this case report, we present a treatment option that has not been previously described in the literature for persistent, atypical facial pain in the setting of silent sinus syndrome.

Case Presentation

A 71-year-old male veteran with the diagnosis of Silent Sinus Syndrome s/p orbital wall reconstruction presented to a Veterans' Hospital Pain Clinic for chronic, persistent pain along the right lateral aspect of the face. The pain started one-year

prior to evaluation without inciting trauma [1]. He had diagnostic workup performed by Ophthalmology and Otolaryngology including MRI imaging of the orbit and CT scan of the sinuses, both of which were unremarkable. Prior to presentation, the patient had undergone burst dose oral steroids and four sequential nerve blocks to the zygomatico-facial and zygomatico-temporal nerves by Ophthalmology for presumed diagnosis of neuroma. Two injections were performed with 0.25 ml of Triamcinolone (Kenalog 10 mg) and 0.25 ml of 0.25% Marcaine, followed by two injections of 0.4 ml of Lidocaine 2% and 0.4 ml Methylprednisolone. All injections were performed 1 month apart [2,3]. Each injection provided approximately 2 weeks of relief before pain returned to baseline. The pain was described as sharp and stabbing with constant symptoms present throughout the day. Pain intensity was rated 7/10 on the Visual Analog Scale (VAS). His pain increased with certain facial activities such as chewing and talking. No relieving factors or modalities were identified. Physical exam was pertinent for mildly depressed right inferior orbital wall and reduced sensation to light touch in the right V1-V3 dermatomal distributions. In addition, tenderness to palpation was found along and superior to the zygomatic arch [4].

Results

Given limited relief with previous trials of oral acetaminophen, non-steroidal anti-inflammatory agents, topical

analgesics, oral steroids and repeated nerve blocks, a trial of incobotulinumtoxinA (Xeomin) was performed to the right facial muscles surrounding the area of pain. During the first visit, the patient received 55 units of incobotulinumtoxinA with injections to Procerus (5 units), Corrugator (5 units each side), Frontalis 20 units) divided into 4 areas) and right temporalis (20 units divided into 4 areas). One-week post injection, he reported 50% reduction in his daily pain. After four weeks, the facial pain reduced by 80%. At 12 weeks, the patient's pain was rated 2/10 on VAS. At his 12-week follow up visit, he received 65 units of incobotulinumtoxinA, in the same locations as before with an additional 10 units at the right lateral orbicularis oculi muscle [5,6]. At his 36-week follow up, the patient received the same 65 unit protocol with continued overall relief, with average pain intensity rated 2/10 on the VAS prior to incobotulinum injections and complete resolution of pain 4 weeks after every series of injections. He continues to receive 65 units of incobotulinumtoxinA for his atypical facial pain every 3 months. Of note, no adverse outcomes were identified to date (Figure 1) [7].



Figure 1: Left: CT Sinus Pre-Op (Coronal View). Complete opacification of a hypoplastic right maxillary sinus with chronic left deviation of nasal septum. **Right:** CT Sinus Post-Op (Coronal View). Status post right maxillary antrostomy; now with resolution of previous opacification of right maxillary sinus.

Discussion

Botulinum toxin has been used for close to thirty years to treat various neurological disorders including focal dystonia, spasticity, trigeminal neuralgia, chronic migraines and pain syndromes associated with muscle spasm. This medication blocks neuromuscular transmission through decreased acetylcholine release, resulting in muscle relaxation [8]. Gobel, et al. proposed

non-neuromuscular effects of botulinum toxin, including increased muscle spindle activity, reverse intake of botulinum toxin in the peripheral and central nervous systems, modulation of central peptide function, and normalization of endplate dysfunction. It has also been proposed that botulinum toxin may induce neuronal processes of central reorganization. It is unclear which mechanism(s) was responsible for this patient's pain relief; however, the improvement raises the possibility of future roles of toxin therapy in otolaryngologic conditions that trigger atypical facial pain symptoms, including spasm. IncobotulinumtoxinA was used off-label for treatment of neuropathic, facial pain.

Conclusion

More research is needed to explore other mechanisms of incobotulinumtoxinA, including its possible role in affecting the trigeminal nucleus or blocking autonomic pathways. In this case report, we discuss a rare clinical presentation and suggest an effective treatment option for pain post surgical correction of Silent Sinus Syndrome.

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