



Glossopharyngeal Nerve Injury Following Tonsillectomy

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Abstract

Tonsillectomy is one of the most frequently performed ambulatory operations in the United States, with over 500,000 performed yearly in children less than 15 years of age. While hemorrhage, dehydration, and pain are commonly encountered complications, glossopharyngeal nerve injury is less frequently sited. Glossopharyngeal Neuralgia (GPN) is characterized by episodes of severe pain in the sensory regions innervated by the auricular and pharyngeal branches of the glossopharyngeal nerve. In this report we present a brief review of the literature regarding GPN and complications of tonsillectomy along with two case reports of GPN which developed post-tonsillectomy.

Keywords: Tonsillectomy; Glossopharyngeal; Neuralgia; Glossopharyngeal nerve; Pain; Neurology

Introduction

Tonsillectomy is one of the most frequently performed ambulatory operations in the United States, with over 500,000 performed yearly in children less than 15 years of age [1]. While hemorrhage, dehydration, and pain are commonly encountered complications, glossopharyngeal nerve injury is less frequently sited. In this report, we present our experience with two cases of glossopharyngeal neuralgia which developed following tonsillectomy. Glossopharyngeal Neuralgia (GPN) is a relatively rare condition characterized by episodes of severe pain in the sensory regions innervated by the auricular and pharyngeal branches of the glossopharyngeal nerve. The 3rd edition of the International Classification of Headache Disorders describes it as “a severe, transient, stabbing, unilateral pain experienced in the ear, base of the tongue, tonsillar fossa and/or beneath the angle of the jaw” which is often “provoked by swallowing, talking and/or coughing”. The pain is often paroxysmal and intermittent similar to that of classical trigeminal neuralgia [2]. Up to 10% of patients may experience vagal nerve involvement causing syncope, seizures, bradycardia, hypotension, and even asystole [3,4].

Data is lacking on the incidence of various facial pain syndromes in the pediatric population, so estimates are often based on data from adult studies. In the general population an incidence of 1 in 25,000 has been reported for trigeminal neuralgia, with only 1% of those cases involving patients under the age of 20 years [5]. While trigeminal neuralgia seems rare, estimates suggest it is between 77 and 5,000 times more common than glossopharyngeal neuralgia [4,6]. Data is lacking in pediatrics, but estimates for the incidence of glossopharyngeal neuralgia in the general population are between 0.2 to 0.8 per 100,000 individuals per year [3,7].

The presentation of glossopharyngeal neuralgia is typically more severe in the pediatric population compared to adults, as they have less pain-free time and their response to pain is frequently exaggerated [8]. In contrast to adults, odynophagia in pediatric patients may discourage children from eating leading to weight loss, malnutrition and potential developmental delays. Facial pain syndromes in adolescents are more likely to lead to anxiety, depression and suicidal tendencies while affecting social functioning and school attendance and performance [5,8,9]. Therefore, accurate diagnosis and treatment is even more pressing in children.

The diagnosis of glossopharyngeal neuralgia can be difficult to make, with a mean latency of 7.3 years from symptom onset to eventual diagnosis [4]. Glossopharyngeal neuralgia is a clinical diagnosis based on a consistent history and typical triggers. The evaluation should include a thorough physical exam including neurologic, oropharyngeal and dental exams. Glossopharyngeal neuralgia may be either idiopathic or symptomatic, so an evaluation for secondary causes is a necessary

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part of the diagnostic work-up [3]. Imaging studies, especially MRI, can be valuable to visualize the nerves and surrounding structures, and to rule out secondary causes [5]. Serum markers of inflammation and autoimmune disease are useful screening tools for secondary causes as well [7]. Potential secondary causes include neurovascular compression and vascular malformations, trauma, tumors, infection, Chiari I malformation, infarction, vertebral artery dissection, demyelinating disease, and post-radiation effects [5,7,8]. The majority of cases are secondary to nerve compression by a vessel at the root entry zone of the brainstem, with the Posterior Inferior Communicating Artery (PICA) being the most common source of vascular compression [7]. Treatment options include medications and surgical procedures, but evidence-based recommendations, especially in pediatrics, are lacking [5,8].

While the vast majority of GPN are idiopathic, there have been reported cases of GPN in post tonsillectomy patients thought to be caused by scar formation resulting in tethering of the mucosa and underlying nerve. Fisher et al. [6] reported a case of a patient experiencing GPN, which resolved after infiltration of procaine into the tonsillectomy scar [6]. We discuss our experience with two cases of GPN in post-tonsillectomy patients to add to the medical literature.

Case Presentation

Case 1

The first case is an 8-year-old girl who presented to the neurology clinic with a chief complaint of pain behind the ears. Approximately 3 months prior to presentation to the neurology clinic she started having intermittent, (5-6) second episodes of “exploding pain” in the bilateral ear canal, anterior to the ear, behind the ears, and less often occipitally. The pain is (8-10) of 10 in severity and described as exploding with a crescendo like resolution. It occurs 50 to 100 times per day, often clustered in the morning or afternoon. Once an episode resolves, a residual lower intensity pain still remains. The pain may be associated with nausea but no vomiting. She denied phonophobia, photophobia, weakness, vision change, tinnitus or vertigo. The pain was first noted in the throat and ear and developed in association with recurrent episodes of Streptococcal pharyngitis. About a month after the pain started, she had a tonsillectomy and adenoidectomy performed and her symptoms have worsened since that time. There is no clear trigger for the pain, and it was not associated with swallowing, coughing, yawning, or chewing. There were no sores on the mouth, face, or in the ears to suggest herpes simplex virus or varicella zoster virus infections. Her headaches are not associated with unilateral symptoms of eye redness, tearing, runny nose, ptosis, or dilated pupil to suggest a trigeminal autonomic cephalgia. The headaches do not wake her from sleep and are not associated with substantial periods of confusion or excessive vomiting. The headaches are not progressive and are not clearly positional. She has had one episode associated with a change in her balance. A review of systems was otherwise negative. She had tried Motrin and Tylenol for relief in the past which were not helpful.

She was the product of a 37-week twin gestation born *via* CS without complications. She has had typical development and no significant past medical history. Both mom and paternal aunt have a history of headaches but no history of brain tumor, clotting disorders, or brain aneurysms in the family. She is in 3rd grade. Her neurologic exam was normal without focal findings or asymmetries. There was no tenderness or pain with palpation of the ears, neck, or

temporomandibular joint. She had an MRI and MRA of the brain without contrast completed which showed a normal brain and focal narrowing of the right ICA at the level of the distal siphon. She continues to follow up and has had mild improvement in her pain since starting Gabapentin, Oxcarbazepine, and Riboflavin. She has also previously tried loratadine, metasone furonate, and carbamazepine which did not help, and Magnesium which had intolerable side effects.

Case 2

The second case is a 3-year-old girl who presented to the neurology clinic with a complaint of throat and face pain after tonsillectomy. The patient had been in her normal state of health until about 4 months prior to her presentation to clinic. At that time, she had a tonsillectomy performed due to difficulty swallowing and breathing difficulties at night. After the procedure she developed throat pain that was so severe that she would not sip water resulting in her becoming hospitalized for dehydration. She continued to have sharp pain lasting 1 min to 2 min affecting the throat, mouth, angle of the jaw and left jaw to the ear. The pain is triggered by coughing, vomiting, eating or drinking. She has been intermittently treated with pain medications including ibuprofen, acetaminophen, tramadol, and gabapentin. Her pain had been well controlled on gabapentin and tramadol but rebounded when attempts were made to discontinue them. When her pain is well controlled, she has no problems with swallowing including coughing, choking, or moving her face. Her headaches are not associated with unilateral symptoms of eye redness, tearing, runny nose, ptosis, or dilated pupil to suggest a trigeminal autonomic cephalgia. Her review of systems was otherwise negative.

She has an unremarkable past medical history with the exception of recurrent ear and sinus infections with surgical history of tonsillectomy, tympanostomy tubes with multiple replacements, and adenoidectomy twice. She has had delayed speech, thought to be secondary to poor hearing in the setting of recurrent ear infections. She is otherwise typically developed. There is a family history of recurrent pharyngeal infections leading to tonsillectomy and adenoidectomy, and grandmother had breast cancer. There is no family history of headaches noted. On exam she is normocephalic with an otherwise normal general exam. Neurologic exam limited as patient would not open her mouth to check cranial nerve function. Otherwise, she had an intact CN exam, normal strength, sensation, DTRs, coordination and gait. An MRI brain was performed which showed a well-marginated ovoid enhancing lesion along the expected course of the left glossopharyngeal nerve at the C2 level concerning for a normal lymph node, although schwannoma or neurofibroma could not be excluded. She is now undergoing surgical evaluation.

Discussion

The implications of glossopharyngeal nerve injury can include persistent neuralgia, dysphagia, and taste dysfunction. As the glossopharyngeal nerve exits through the jugular foramen at the skull base, it travels toward the tongue base and gives off a lingual and tonsillar branch near the posterior inferior aspect of the tonsil. These branches usually lie deep to the pharyngeal constrictor muscles, thereby protecting them from injury during the subcapsular dissection of the tonsils [10]. However, Ohtsuka et al. [11] found that in 83 adult cadavers the lingual branch of the glossopharyngeal nerve location was quite variable and was adherent to the tonsillar capsule in nearly 25% of the cadavers [11]. Ford et al. [12] studied 20 cadaveric heads

and found that the mean distance from the posteroinferior tonsillar fossa to the lingual branch of the glossopharyngeal nerve was 6.5 mm [12]. Hill and colleagues evaluated 138 pediatric patients undergoing a tonsillectomy and found that approximately 20% of those children had at least one exposed nerve and 6.5% had bilateral nerve exposure during the operation [10].

Given that the glossopharyngeal nerve passes closely to the tonsillar fossa, it seems plausible that the nerve could be vulnerable to injury, especially if there is a poorly delineated surgical plane. This can often be seen in cases of recurrent tonsillitis or peritonsillar abscess, where the loose connective tissue between the tonsillar capsule and muscular wall is scarred. In addition, the thermal injury from electrocautery may be transmitted to the nerve, and oral retractors can result in excessive stretching of the nerve. The glossopharyngeal nerve has motor, sensory and autonomic functions. It supplies motor fibers to the stylopharyngeus muscle and indirectly to the pharyngeal constrictors through pharyngeal plexus branches. It also sends sensory branches to the pharynx, posterior third of the tongue, soft palate, tonsillar pillars, and palatine tonsils. Another important function includes taste to the posterior one third of the tongue [7,12]. Given the innate function of the glossopharyngeal nerve, its preservation may prevent dysphagia, dysgeusia, and pain following tonsillectomy [13].

Post-tonsillectomy dysphagia is commonly encountered after surgery and not necessarily considered a complication unless it persists beyond 10 to 14 days. Severe dysphagia from bilateral glossopharyngeal nerve paralysis has been reported in a few case reports. Ford et al. [12] reported a case of bilateral glossopharyngeal paralysis evident on fluoroscopic studies resulting in severe dysphagia, decreased taste sensation, nasal regurgitation of liquids, nasal voice, and weight loss persisting months after tonsillectomy in an adult patient [12]. Stathas et al. [14] studied the effect of tonsillectomy on taste function on 60 patients including both pediatric and adult subjects and found that all had taste dysfunction in the immediate post-operative period, but this resolved within a month [14]. Goins et al. [15] reported a case of an adult patient who had experienced persistent taste dysfunction, particularly with impairment of bitter taste detection, and chronic foreign body sensation for several months after undergoing a tonsillectomy. Although dysphagia and taste dysfunction are rare and usually transient complications, they have the potential to cause significant distress, food choice alterations, weight loss, malnutrition, and immune deficiency [15]. While pain in the immediate post-operative period is a known complication of tonsillectomy, less commonly reported is glossopharyngeal neuralgia.

Both of our patients developed glossopharyngeal neuralgia and were treated medically with combination therapy that included Gabapentin. Although there are many commonly prescribed medications for GPN, no treatment guidelines exist and there are few studies to refer to on the treatment of children and adolescents with GPN [8,9]. Data is lacking on medication management of glossopharyngeal neuralgia with most existing studies on the treatment of facial pain syndromes focused on trigeminal neuralgia. Shereen R, et al. [8] reported in their review that pharmacotherapy is effective in about one-third of reported cases of GPN [8]. Carbamazepine, Gabapentin, and Pregabalin have all been cited as first line treatments [3,7] although Carbamazepine is the most widely used [15]. In a case series of adult patients from 1990-2009, Carbamazepine was used by 70% of physicians, followed by

Gabapentin and Amitriptyline [4]. There are reports of Phenytoin, Lamotrigine, Valproic Acid, Oxcarbazepine, Baclofen, Sumatriptan, Acyclovir, Methylprednisolone, dextromethorphan, and SSRIs being used for medical treatment [3,5,7,9]. Nerve blocks using Lidocaine and bupivacaine with or without steroids have been tried as well using both intra- and extra-oral approaches [7].

While our second patient has had good results with medical management, our first patient has been somewhat refractory. For cases of GPN that are refractory to medications, there are many available surgical options including percutaneous methods, open surgical methods, and radiosurgery [3]. The list of cited procedures used for GPN includes radiofrequency gangliolysis, glycerol gangliolysis, balloon compression, stereotactic radiosurgery, pulsed radiofrequency neurolysis, peripheral neurectomy, nerve root rhizotomy, cryotherapy, and Microvascular Decompression (MVD) [3,5,7]. Percutaneous radiofrequency thermocoagulation has fallen out of favor due to a high incidence of unfavorable complications and is no longer typically recommended [3,5]. Direct Nerve Sectioning (NS), Stereotactic Radiosurgery (SRS) and MVD are the 3 most commonly used procedures [16], with MVD being the most widely used surgical technique due to its high success rate and low complication rate [3,7]. MVD success rates of 76% to 97% have been shown in various case series [4,7] although studies suggest that MVD has decreased efficacy in childhood compared to adults [5,8]. Stereotactic gamma knife radiosurgery appears to have a higher recurrence rate than MVD, but one benefit is that evidence of neurovascular decompression is not mandatory [3]. There are no studies of gamma knife radiosurgery being effective for glossopharyngeal neuralgia in the pediatric population [8]. In a meta-analysis by Lu VM, et al. [16] comparing Nerve Section (NS), MVD and Stereotactic Radiosurgery (SRS) as treatment options for glossopharyngeal neuralgia, they found that NS and MVD were equivalent for both short- and long-term pain relief. Both were significantly better than SRS for those measures; however, SRS and NS were significantly superior to MVD for operative complications [16]. More data in the pediatric population is needed to guide medical and surgical treatment strategies in pediatric glossopharyngeal neuralgia cases.

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