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Percutaneous Pulsed Radiofrequency Treatment in a Patient with Chronic Bilateral Painful Glossopharyngeal Neuropathy

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Conflict of interest: None declared

Patient: Female, 41-year-old
Final Diagnosis: Chronic bilateral painful glossopharyngeal neuropathy
Symptoms: Chronic pain
Medication: —
Clinical Procedure: Percutaneous pulsed radiofrequency treatment
Specialty: Anesthesiology

Objective: Rare disease

Background: Due to its rareness, we present a case of chronic, bilateral, painful glossopharyngeal neuropathy, which developed after nasal septum and inferior concha surgery, and was non-surgically treated with percutaneous pulsed radiofrequency at the glossopharyngeal nerve, using an extra-oral approach.

Case Report: A 41-year-old Caucasian female patient (60 kg, 1.57 m, body mass index 24.8 kg/m^2) was referred to the Pain Center by her general practitioner because of ongoing pressing pain in her throat 4 months after nasal septum and inferior concha surgery. Based upon medical history, physical examination and the results of additional questionnaires, a probable diagnosis of atypical neck pain was made, based on ongoing glossopharyngeal stimulation, involvement of the pterygopalatine ganglion or/and superior cervical ganglion, with secondary involvement of the muscles of the neck. We changed the analgesic regimen and performed a pulsed radiofrequency treatment of the glossopharyngeal nerve on both sides. The patient had made progress and reported that she actually felt better but she asked for repeat treatment because of residual complaints. We performed the procedure for a second time on both sides. The results of the questionnaires before (T0) treatment, 3 months after the first (T1) and 3 months after the second (T2) treatment are provided. After the second procedure, the patient reported that her swallowing complaints had further diminished, as well as the pain behind her ears. She stopped using pregabalin. Residual complaints were manageable.

Conclusions: In patients with painful glossopharyngeal neuropathy, a non-surgically treatment with percutaneous pulsed radiofrequency at the glossopharyngeal nerve, using an extra-oral approach, seems to be an effective and safe method to use.

MeSH Keywords: Facial Neuralgia • Glossopharyngeal Nerve • Glossopharyngeal Nerve Diseases • Pulsed Radiofrequency Treatment • Surgical Procedures, Minimally Invasive

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Background

Glossopharyngeal neuralgia presents itself with severe pain episodes at the tongue, external ear, tonsil, and beneath the angle of the jaw [1]. Overlap amongst the nerves supplying the face may occur. Besides a varied location of the pain, unusual presentations can occur, i.e., cardiac arrhythmias and syncope [2,3]. In a short review article published in 2011, the occurrence of syncope is about 20%, and that of convulsive syncope about 5% [4]. The nerve of Hering, a branch of the glossopharyngeal nerve to the carotid sinus, may be accountable for the arrhythmogenicity [5].

The annual incidence rate per 100 000 population in a 1991 published report was 0.7 for both sexes combined, with no significant differences between the sexes [6]. At that time, glossopharyngeal neuralgia was suggested to be a mild disease and bilateral occurrence was not uncommon but observed in one-fourth of the patients. In an article from 1981, Rushton et al. reviewed 217 cases of glossopharyngeal neuralgia and reported that bilateral involvement was 12% [7]. Diagnostic difficulties and unawareness of the disease make reporting a specific incidence difficult.

Pain attributed to a lesion or disease of the glossopharyngeal nerve may be classified as glossopharyngeal neuralgia and painful glossopharyngeal neuropathy [8]. The primary pain in glossopharyngeal neuralgia is usually continuous or near-continuous. The pain is experienced as burning and squeezing, and the gag reflex may be weak or missing. This distinguishes painful glossopharyngeal neuropathy from glossopharyngeal neuralgia. However, despite the literature on bilateral occurrence of the glossopharyngeal nerve pain, the International Headache Society (HIS) classification still speaks of unilateral pain.

The cause of painful glossopharyngeal neuropathy and glossopharyngeal neuralgia may be idiopathic and without any obvious lesion, or occurring after trauma (e.g., fracture, penetrating injury, intubation, direct carotid puncture), infection, radiation, neoplasm, surgery, vascular malformations, demyelination, and Eagle's syndrome [5]. As in the case report by Saccomanno et al., Eagle's syndrome is suspected mostly in adult women when the pain is unilateral and not responsive to pain medication [9]. While they are a rare condition, pediatric glossopharyngeal neuralgia has also been reported [10].

For most of the patients, nonsurgical therapy is the treatment of choice [5]. First-line treatment is the use of anticonvulsant medications such as carbamazepine, gabapentin, or pregabalin [11]. Due to its resistance to analgesics, early interventional pain treatment can be the preferred method [12]. Intra- and extra-oral approaches have been described, both with its advantages and disadvantages [1,5].

Sluijter et al. described the use of isothermal radiofrequency treatment (pulsed radiofrequency or PRF) in 1996 [13,14]. Applying PRF leads towards a disruption of the pain signaling, but with a limited heating of the tissue. The radiofrequency wave is broken up into short bursts of signal output with no signal in between. This form of treatment delivers the radiofrequency signal without producing destructive levels of heat. In contrast to continuous radiofrequency does PRF produce less pronounced tissue destruction. As a consequence of PRF treatment, mitochondria, microtubules and microfilaments undergo damage in a variable sense, which progressively worsens from A-delta towards A-delta and C fibers. The effect of PRF on A-delta and C fibers shows that this treatment alters the sensory nociceptors and relatedly saves the tactile sensory input [15,16].

Due to its rareness, we present a case of chronic, bilateral, painful glossopharyngeal neuropathy, which developed after nasal septum and inferior concha surgery, and was non-surgically treated with percutaneous PRF at the glossopharyngeal nerve, using an extra-oral approach.

Case Report

A 41-year-old Caucasian female patient (60 kg, 1.57 m, body mass index 24.8 kg/m^2) was referred to the Pain Center by her general practitioner because of ongoing pressing pain in her throat 4 months after nasal septum and inferior concha surgery. The surgery itself had not diminished her complaints. There were no records of a traumatic intubation, nor was there any relevant past medical history. Her pain radiated from her throat towards both ears, neck, and the anterior upper part of her thorax. The tongue and part of the mouth felt swollen. She did not have any fever. Repeat consultations with her otorhinolaryngologist did not reveal any new insights.

During the next few months, her complaints increased, and swallowing became increasingly troublesome. She had used naproxen, codeine, and tramadol without success. A trial with antibiotics (amoxicillin/clavulanic acid) failed to reverse the situation, nor did physiotherapy. She became increasingly dependent regarding domestic responsibilities and daily care. A diagnosis of "somatic unexplained physical complaints" was made and the patient was told that referral towards a pain center could be a next option, as well as towards a psychiatric hospital.

During physical examination it was noticed that she experienced pain when pressure was applied on the greater cornua of the hyoid bone and, to a lesser extent, on the muscles of the lateral sides of the neck. No other particularities were observed. Based upon medical history, physical examination, and the results of additional questionnaires (Patient Reported

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Table 1. Results of the patient reported outcome measurements (PROMS) questionnaires before (T0), 3 months after the first (T1), and 3 months after the second (T2) percutaneous pulsed radiofrequency treatment of the glossopharyngeal nerve.

		T0	T1	T2
NRS		10	6	5
BPI	General activity	10	7	7
	Mood	10	6	6
	Walking ability	7	7	5
	Normal work	10	8	7
	Relations with other people	10	5	7
	Sleep	10	4	4
	Enjoyment of life	10	7	6
HADS	Anxiety	20	14	12
	Depression	18	9	13
NDI		44	23	20

NRS – Numerical Rating Scale for pain; BPI – Brief Pain Inventory (pain interference items); HADS – Hospital Anxiety and Depression Scale; NDI – Neck Disability Index.

Outcome Measurements or PROMS, Table 1), a probable diagnosis of atypical neck pain was made, based on ongoing glossopharyngeal stimulation, involvement of the pterygopalatine (sphenopalatine) ganglion or/and superior cervical ganglion, with secondary involvement of the muscles of the neck. The whole situation had significant impact on her quality of life, as well as that of her family.

Several treatment possibilities were offered, consisting of supportive care of the specialist pain nurse, multidisciplinary consultation, anti-neuropathic medication, pulsed radiofrequency (PRF)/infiltration of the greater cornua of the hyoid bone, PRF of the glossopharyngeal nerve, PRF of the pterygopalatine (sphenopalatine) ganglion and PRF of the superior cervical ganglion.

The results of the questionnaires are presented in Table 1. The analgesic regimen was changed from Tramadol, Codeine and Paracetamol into Carbamazepine 100 mg oral twice daily and increased after 2 weeks to 200 mg oral twice daily. This decreased her pain considerably, but unfortunately had to be discontinued because of side effects (generalized skin rash). The analgesic regimen was then changed to pregabalin 75 mg oral twice daily, increasing the dose to 150 mg oral twice daily after 2 weeks. This again had a positive effect, now without any side effects.

In the meantime, the patient went for a second opinion to an otorhinolaryngologist of the nearby university hospital but again no causes could be found. After a few weeks she asked us for an alternative to the medication and so it was decided to perform a minimally invasive treatment with PRF.

The patient was positioned in the supine position. Vital parameters were recorded before, during and after the procedure. Intravenous access was obtained,and mild sedation was given with intravenous (IV) propofol 1–3 mg/kg/hour and alfentanil if needed (0.25–0.5 mg IV bolus). Oxygen at 3 L/minute was continuously provided nasally. Anteroposterior (AP) and lateral images were obtained, showing the angle of the mandible, mastoid process and styloid process. The area was aseptically prepared and draped. A 22-gauge 6 cm radiofrequency needle (Equip-Medikey, Gouda, The Netherlands) was advanced until bony contact with the styloid process was made. The needle was then walked off posteriorly and advanced 1 cm (Figures 1, 2). With the patient awake, sensory stimulation at 50 Hz up to 1 V was used to reproduce concordant pain. The needle was adjusted to ensure correct position and PRF treatment (using a Sluijter-Teixera poisson (STP) pulse, random mode, 8 minutes, 45 V) was performed (TOP TLG-10 lesion generator, Equip-Medikey, Gouda, The Netherlands). After the intervention, the patient was monitored for two hours for any adverse effects.

The results of the PROMS questionnaires at T1 (3 months after the invasive procedure) are presented in Table 1. The patient had made progress and she reported that she actually felt better but asked for repeat treatment because of residual complaints. We performed the procedure as explained above for a second time on both sides; the patient was again monitored for 2 hours for any adverse effects. The results of the PROMS questionnaires at T2 (3 months after the second invasive procedure) are presented in Table 1. After the second procedure, patient reported that her swallowing complaints had further diminished, as well as the pain behind her ears. She stopped using pregabalin. Residual complaints were manageable.

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Figure 1. Lateral view of the percutaneous pulsed radiofrequency (PRF) treatment of the glossopharyngeal nerve. The needle is directed towards the styloid process and moved posteriorly.

Discussion

This case report describes the use of percutaneous PRF at the glossopharyngeal nerve in a patient with rarely seen chronic, bilateral painful glossopharyngeal neuropathy. In the patient presented in this case report, trauma (intubation) and infection might have triggered the glossopharyngeal neuropathy. However, mainly based upon the results of the questionnaires, a psychologic/psychiatric condition must be included in the differential diagnosis.

Efficacy of PRF treatment to the glossopharyngeal nerve in the case of chronic, non-cancer pain has been shown in few case reports [17,18]. In a retrospective study of 80 patients (71 for follow-up) treated between 2003 and 2014, percutaneous glossopharyngeal nerve PRF treatment proved to be a safe and effective method [11]. This was also the case in another retrospective trial from the same year, reporting the results in 103 patients [19]. In patients with oropharyngeal cancer pain, PRF treatment of the glossopharyngeal nerve can be used effectively and safely for the treatment of glossopharyngeal neuralgia [12]. The use of radio frequency lesioning of the glossopharyngeal nerve in the case of malignancies has been reported in several articles [20]. When comparing nerve sectioning, microvascular decompression, or stereotactic radiosurgery, nerve section provided the most favorable treatment response [21]. Sectioning of the cranial nerves IX and X, microvascular decompression or a combination of the 2 has been suggested as a safe and effective surgical therapy [22].

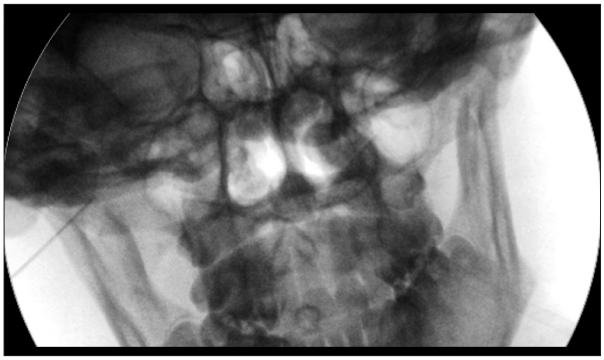

Figure 2. Anteroposterior view of the percutaneous pulsed radiofrequency (PRF) treatment of the glossopharyngeal nerve. The needle is directed towards the styloid process and moved posteriorly in order to glide off the styloid process.

This report obviously has several limitations. First of all, this is a case representation and therefore no control group nor case series or blinding are available. Possibly due to language barriers, before answering the questions asked by the pain team, the patient discussed everything with her husband. In addition, the oral answers she provided at T1 and T2 seemed to be pointing towards an even more effective treatment than the selected answers on the questionnaires. However, this might just be our opinion.

Conclusions

A non-surgically treatment with percutaneous PRF at the glossopharyngeal nerve, using an extra-oral approach, seems to be an effective and safe method to use in patients with painful glossopharyngeal neuropathy

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