CASE REPORT

Risks and Complications of Glycerol Rhizotomy in a Patient of Trigeminal Neuralgia Due to Cavernous Sinus Meningioma

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ABSTRACT

We present a case of trigeminal neuralgia caused by a cavernous sinus meningioma. The pain was difficult to manage with medical treatment. Due to the presence of background medical conditions and risk of ophthalmoplegia, a surgery was considered too risky. After consideration of available options, glycerol rhizotomy was offered. Post-procedure, the patient suffered from chemical meningitis, treated successfully with steroids and lignocaine infusion. The patient's symptoms recurred soon, suggesting ineffective procedure. We have considered the potential reasons underlying occurrence of chemical meningitis and its failure to relieve symptoms, in the context of meningioma resulting in reduced capacity of Meckel's cave, anatomical distortion, and propensity for glycerol to extend into posterior fossa in such patients and whether it may be possible to predict these from imaging and avoid this procedure in such patients.

Key words: Glycerol rhizotomy, meningioma, trigeminal neuralgia

INTRODUCTION

Since its inception by Häkanson, glycerol rhizotomy continues to prove a useful method for the treatment of trigeminal neuralgia.[1] Häkanson's earlier trials in stereotactic gamma radiation for treatment of trigeminal neuralgia involved using a glycerol carrier to inject tantalum dust into the trigeminal cistern.[1] The breakthrough discovery was revealed when relief from pain was established using a glycerol medium alone; this was thought to be due to demyelination and axonal fragmentation upon further analysis.[2,3] Following on from Häkanson, the procedural technique has generally had a satisfactory therapeutic effect and has undergone slight modification. Intraoperative imaging helps accurately guide a needle toward the trigeminal cistern through the foramen ovale. A contrast medium is injected into the cistern, outlining Meckel's cave. Once the position is confirmed, anhydrous glycerol is injected into the same area filling the cistern.[4]

In a study that retrospectively analyzed 260 consecutive retrogasserian glycerol rhizotomies for trigeminal neuralgia treatment, it was noted that complications (either transient or persistent) occurred in 67.3% of cases.[5] Most complications were related to mild sensory defects and were non-disabling for the patient. More serious post-operative complications included labial herpes, anesthesia dolorosa, dysesthesia, chemical meningitis, and infectious meningitis. Most patients experienced relief from trigeminal neuralgia pain within a few hours of rhizotomy and were discharged on the day of the procedure without post-operative imaging.[6] Although trigeminal neuralgia is not a static disorder, one characterized by a relapsing-remitting course, long-term pain relief (11 years post-procedure) has been reported by Kondziolka and Lunsford in 77% of patients (n = 1174) who underwent glycerol rhizotomy for trigeminal neuralgia.[7]

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Special consideration must be taken in the treatment of trigeminal neuralgia secondary to tumors. Cheng *et al*. conducted a comprehensive study of 296 patients diagnosed with trigeminal with neuralgia cases secondary to tumor compression. Of all available surgical treatment options, resection of the tumor through craniotomy and debulking was deemed to be the most effective method in achieving pain control. However, in patients with high anesthetic risk, temporarily or permanently blocking afferent impulses through stereotactic radiosurgery and/or glycerol rhizotomy were deemed satisfactory alternatives to a craniotomy and debulking procedure.[8]

CASE REPORT

A 51-year-old female with the left-sided trigeminal neuralgia, secondary to a known left cavernous sinus meningioma, was admitted for elective glycerol rhizotomy following the failure of medical management. The meningioma was diagnosed incidentally 3 years earlier during investigations for thunderclap headache. At the time of diagnosis, the patient was known to have fibromyalgia, chronic obstructive pulmonary disease, a history of recurrent bronchial sepsis requiring several ITU admissions, and Crohn's disease. Given the prolonged pain history, the patient was regularly reviewed by the pain team and prescribed multiple pain medications, including tramadol, pregabalin, amitriptyline, and duloxetine. The patient was on these medications for 3 years, before glycerol injection was considered. Her trigeminal neuralgia was of sharp lancinating pain to her left face, affecting both maxillary and mandibular distributions of the trigeminal nerve. The cavernous meningioma was managed conservatively with annual magnetic resonance imaging (MRI) scans, and these showed stable appearances with no change in the size of the tumor [Figures 1 and 2]. Given the significant facial pain and comorbidities precluding general anesthesia, the patient was offered percutaneous glycerol injection.

The procedure was performed with conscious sedation using intravenous propofol and fentanyl. The patient was positioned supine with a slight head extension to approximately 15 degrees. A 20-gauge spinal needle was guided from 2 cm lateral to the corner of the mouth towards the foramen ovale with X-ray guidance [Figure 3]. Cerebrospinal fluid (CSF) release was noted on the passage of the needle through foramen ovale. 0.5 ml of radio-opaque contrast (Niopam 300®) was instilled under direct lateral fluoroscopy and a submental vertex check X-ray confirmed the correct cannulation of Meckel's cave and delineation of the trigeminal ganglion, with no contrast escaping into any blood vessels. Following this, 0.4 ml of 99.9% non-radiopaque anhydrous glycerol solution was instilled. Sedation was weaned, and the patient was transferred to theater recovery.

On waking in recovery, the patient became agitated and complained of severe, intractable headache, nausea, and photophobia, with no associated focal neurological deficit elicited on examination. Glasgow Coma Scale remained

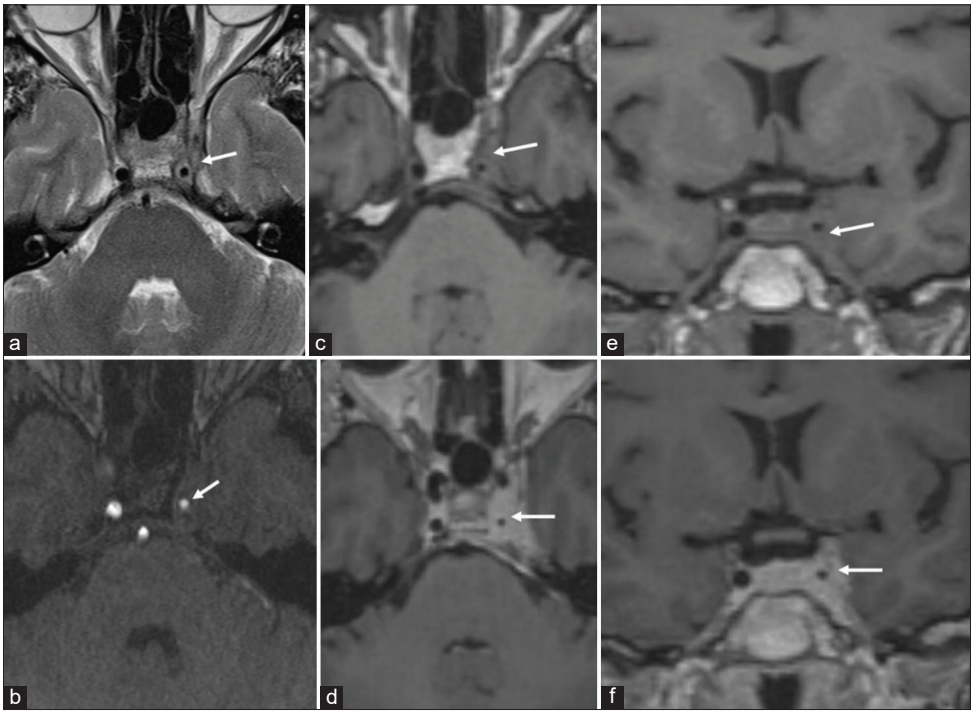

Figure 1: Magnetic resonance imaging brain. T2 axials (a), time of flight angiogram (b), T1 pre- and post-contrast axials (c and d), and T1 pre- and post-contrast coronals (e and f). White arrow shows the enhancing mass in the left cavernous sinus, encasing and narrowing left cavernous internal carotid artery

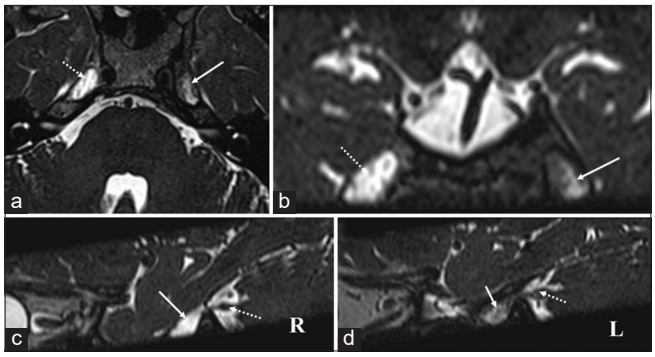

Figure 2: Magnetic resonance imaging Brain. T2 SPACE sequence. Axial (a) and coronal (b) reconstructions show a narrowed left Meckel's cave (white arrow) and normal right Meckel's cave (broken white arrow). (c) shows sagittal reconstruction on the right side showing normal Meckel's cave (white arrow) and trigeminal nerve (broken white arrow). (d) shows sagittal reconstruction on the left side showing narrowed Meckel's cave (white arrow) and trigeminal nerve (broken white arrow)

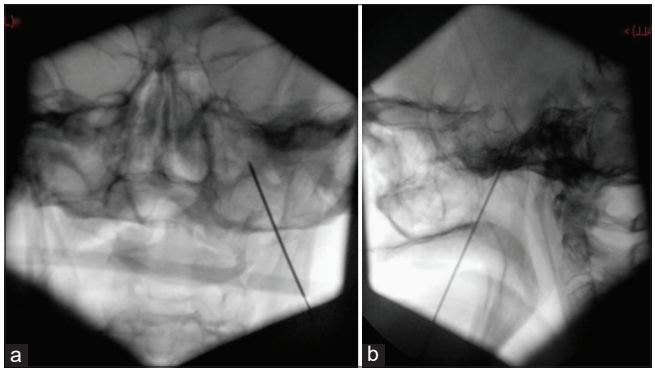

Figure 3: Intraoperative radiographs (a) frontal and (b) lateral views show the needle in position

15; pupils were equal and reactive bilaterally. Analgesia was administered. Urgent computed tomography (CT) head revealed effacement of sulcal spaces and basal cisterns, due to the presence of subarachnoid contrast and cerebral edema [Figure 4] with clearly seen leaked contrast. The patient's headache proved difficult to treat, though her facial pain significantly improved initially. The severe headaches suggested that not only contrast but also glycerol that had leaked out to the subarachnoid space. The diagnosis of aseptic chemical meningitis was made, and treatment in the form of high-dose steroid commenced alongside an intravenous Lidocaine infusion. Her symptoms gradually improved over the next 5 days. Follow-up CT scan after 7 days revealed the resolution of abnormalities with the return of normal appearances [Figure 5]. The patient was later discharged home on the second post-operative day. The patient reported relative stability of facial pain for the first 6 weeks postoperatively. However, she, unfortunately, suffered from intermittent exacerbations thereafter. Considering ongoing symptoms, she continued pregabalin and duloxetine under

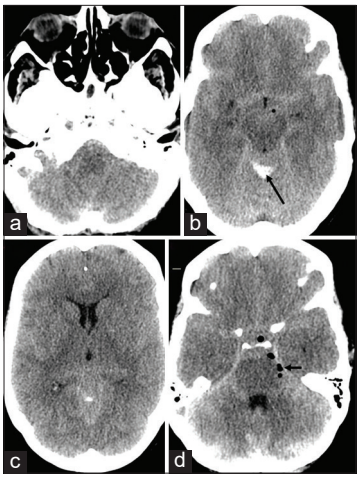

Figure 4: (a-d) anImmediate post-operative non-contrast computed tomography. Axial reconstructions. Black arrow in b shows hyperdense material (contrast). There is further diffuse effacement of sulcal spaces, sylvian fissures, and basal cisterns, partly due to contrast and partly due to brain swelling due to chemical meningitis. Black arrow in d shows intracranial gas, post-procedure

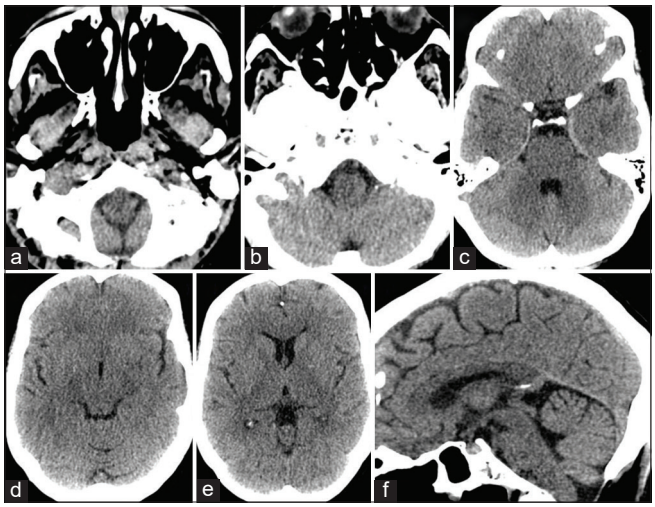

Figure 5: (a-f) Follow-up non-contrast computed tomography (CT) shows the resolution of changes in pre-operative CT [Figure 4] and normal appearances

the care of the pain team, and her meningioma was referred for treatment with stereotactic radiotherapy (SRT) 6 months later as second-line treatment of trigeminal neuralgia. In the interim, the tumor was noted to have increased, and she was deemed not suitable for SRT and underwent conventional radiotherapy. Unfortunately, the patient's symptoms have not improved in the first 6 weeks following radiation, and she continues to be on follow-up.

DISCUSSION

Despite being a widely described technique, we could not find any reports primarily studying the efficacy of injecting

glycerol into Meckel's cave for patients with cavernous meningiomas. Unlike other skull base meningiomas, in which excision ± microvascular decompression carries an 80% 10-year relief of trigeminal pain,[9] radical cavernous meningioma surgery carries a significant risk of ophthalmoplegia,[10] which would be unacceptable to our patient, as her tumor was stable in size with no diplopia or vision loss. Moreover, our patient had chronic obstructive pulmonary disease and bronchiolitis obliterans organizing pneumonia which would pose a significant anesthetic risk. Therefore, a less invasive procedure was opted for, in the form of glycerol rhizotomy.

The frequency of aseptic chemical meningitis after Gasserian ganglion glycerol injection has been estimated at 1.5%. The mechanism remains unclear, but the overflow of irritating contrast or glycerol into the posterior fossa from the trigeminal cistern within Meckel's cave has been hypothesized.[5] This theory especially applies to our patient, given the abnormal middle fossa floor anatomy secondary to her meningioma.

Meckel's cave, being composed of the dura, is affected in approximately 1% of meningiomas.[11] The trigeminal cistern is formed by the upward extension of subarachnoid space from the prepontine cistern of the posterior fossa.[12,13] The previous cadaver studies undertaken to determine the cubic capacity of the trigeminal cave have radiographically noted Myodil contrast overflowing into the posterior cranial fossa after injection through the foramen ovale. Myodil contrast has been shown to produce similar radiographic appearances *in vivo* when injected presumably into the substance of the trigeminal ganglion itself or into the extra-arachnoid plane within Meckel's cave.[14] Furthermore, escape of contrast medium through the porus trigeminus to the posterior fossa has also been noted to occur when a patient's head is not adequately tilted forward during intraoperative glycerol injection.[15]

The release of CSF on needle entry into Meckel's cave suggests that we injected contrast and then glycerol into the correct area. However, pre-operative high-resolution MRI SPACE imaging [Figure 2] demonstrated that the volume of Meckel's cave has been reduced by tumor. We, therefore, postulate that the standard volume of 0.4 – 0.5 ml glycerol injected may have been too great in this case and caused spillage through the porus trigeminus into the posterior fossa due to distortion of normal anatomy and reduced capacity of the Meckel's cave. It is also possible that this alteration of anatomy may have resulted in rendering the head tilt ineffective and made more likely for glycerol to leak posteriorly. It is difficult to guess how much-reduced glycerol volume would have been safe, this factor, when considered in isolation, would suggest that there could be a potential for offering treatment with reduced/adjusted volume while avoiding complications.

It is, however, more pertinent to note that the patient had only transient relief of trigeminal neuralgia that may well have been due to medical management. Since the anatomy of Meckel's cave was distorted, it is difficult to comprehend if glycerol would have come in contact with the ganglion/nerve in the same way as usually expected as it may have been shielded by the tumor or distorted anatomy, thereby causing suboptimal relief of neuralgia. This may have been an additional factor in the leakage of glycerol, contributing to suboptimal relief of neuralgia. We, therefore, suggest that in tumors extending into Meckel's cave or when the capacity of Meckel's cave is seen to be reduced on high-resolution sequences such as T2 SPACE, while there may be a temptation to perform the procedure with a reduced volume of glycerol, the option for glycerol rhizotomy should not be considered for risk of being ineffective anyway.

CONCLUSION

We have presented a case of difficult to treat trigeminal neuralgia caused by a cavernous sinus meningioma, treated by glycerol rhizotomy. We have discussed the risks and complications and the potential explanations for these and have highlighted the risk of failure of this treatment in such cases, particularly when the anatomy is distorted and the volume of Meckel's cave is reduced. While there may be a temptation to offer this treatment by reducing the volume of glycerol, it may still be ineffective and not serve any purpose. It, therefore, remains a challenge, how to effective treat such cases.

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