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Gamma knife radiosurgery to the trigeminal ganglion for treatment of trigeminal neuralgia secondary to vertebrobasilar ectasia

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Abstract

Background: We report the result obtained using Gamma knife stereotactic radiosurgery on the trigeminal ganglion (TG) in a patient with trigeminal neuralgia (TN) secondary to vertebrobasilar ectasia (VBE).

Case Description: Retrospective review of medical records corresponding to one patient with VBE-related trigeminal pain treated with radiosurgery. Because of the impossibility of visualization of the entry zone or the path of trigeminal nerve through the pontine cistern, we proceeded with stereotactic radiosurgery directed to the TG. The maximum radiation dose was 86 Gy with a 8-mm and a 4-mm collimator. The follow-up period was 24 months. The pain disappeared in 15 days, passing from Barrow Neurological Institute (BNI) grade V to BNI grade IIIa in 4 months and then to grade I. The patient did not experience noticeable subjective facial numbness.

Conclusions: This experience showed that Gamma knife radiosurgery was effective in the management of VBE-related trigeminal pain, using the TG as radiosurgical target.

Key Words: Gamma knife radiosurgery, trigeminal neuralgia, trigeminal ganglion, vertebrobasilar ectasia

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INTRODUCTION

Trigeminal neuralgia (TN) consists of brief and severe paroxysms of pain in the facial distribution of the trigeminal nerve, which is often triggered by facial movements or stimulation of sensory endings in the trigeminal area. TN can be caused by tumors involving the trigeminal nerve, ganglion, or divisions, as well as by demyelinating plaques in the dorsal nerve root entry zone, descending trigeminal tracts, or brainstem nuclei due to multiple sclerosis. It is often associated with neurovascular compression of the trigeminal nerve root entry zone. In most cases, the vascular compression is related to arterial compression from the superior cerebellar artery or anterior inferior cerebellar artery, or less commonly, from prominent draining veins of the brainstem or cerebellum.[16,17]

Vascular compression of the trigeminal nerve secondary to basilar artery ectasia is an unusual cause of TN.[19] Only 2% of patients have TN due to compression by the complex vertebrobasilar.[16]

Several treatment options for TN are available including microvascular decompression, radiofrequency rhizotomy, glycerol rhizolysis, microballoon compression, and alcohol block. All of these procedures, except for microvascular decompression, are ablative procedures, which are associated with a risk of facial numbness. Patients with decompression of a tortuous VEB have a higher risk of trigeminal dysfunction, diplopia and hearing loss than standard microvascular decompression.[16] Gamma Knife Surgery (GKS) is a less invasive ablative procedure, which uses target-directed radiation. Most patients are treated using a single isocenter through a 4-mm collimator helmet targeting the root entry zone of the trigeminal nerve.[12,36,37]

We report a case of severe TN secondary to vertebrobasilar ectasia (VBE), where the trigeminal nerve could not be properly visualized on neuroimaging studies. It was decided to treat Gasser ganglion to relieve his painful condition.

CASE REPORT

A 60-year-old male nondiabetic, nonsmoker, hypertensive, obese, presented with intermittent right-sided, severe, sharp, and lancinating facial pain for the past 4 years, with an identifiable trigger area located at upper right canine teeth. The patient had difficulties for chewing, eating, drinking, shaving, washing his face, altering his daily activities. The pain used to come in sudden bursts lasting 1-5 min and recurs more than 15 times a day. On physical examination, arterial hypertension and obesity were found. Neurologic examination showed facial trigger points in the right maxillary region and nasogenian fold without other neurological findings. Patient had longstanding pain resistant to medical management with agents such as carbamazepine (1200 mg/day), alone or in combination with gabapentin or pregabaline. The patient underwent multiple punctures and local infiltration with alcohol and steroids without improvement. He did not have previous surgical procedures. No sensory loss over a particular nerve division was reported by the patient at any time. Facial sensory exam with pin or light touch was normal before the radiosurgical procedure. Corneal blink reflex was normal.

METHODS

Radiosurgical technique

Leksell Gamma Knife model 4C was used (Elekta Instruments, Inc.). Under mild sedation and local anesthesia, the Leksell Model G stereotactic frame (Elekta Instruments) was applied. The patient underwent stereotactic magnetic resonance (MR) imaging to identify the trigeminal nerve. The MR imaging (MRI) was performed using contrast-enhanced, short repetition time sequences and axial phase volume acquisitions of 256 × 256 matrices divided into 1-mm slices. A long relaxation time MR imaging study was obtained. A treatment plan was implemented using the Leksell Gamma Plan treatment planning system (Elekta AB).

MRI findings

A three-dimensional time-of-flight (3D-TOF) sequence was performed using a 1.5-Tesla MRI scanner (General Electric Signa HD) to visualize the trigeminal nerve and its relationship with the VBE. A fast imaging employing steady state acquisition sequence (FIESTA) provides images included in the MR protocol [Figure 1].

The affected trigeminal root was not recognized in the MRI scan at the entry zone neither at the pontine cistern. A large, elongated, and tortuous vertebrobasilar artery was causing mechanical compression at the right trigeminal nerve root with displacement and marked deformity of brainstem by compression of pons [Figure 2].

Dose selection

Based on the successful experience in treating our patients with typical TN, focusing the radiation at the entry zone, we decided to apply the same dose of 43 Gy prescribed to the 50% isodose. Two isocenters were used, one 8-mm isocenter placed on the Meckel's Cavum and a 4-mm at the exit zone of the trigeminal nerve from the trigeminal ganglion (TG).

Clinical follow-up

Patient's records are updated every 4 months. The patient continued medical treatment for 4 months, when the dose was gradually tappered. We evaluated the degree of pain relief, latency interval to pain relief, drugs used, development of new symptoms or signs, and the need and response to additional surgical procedures. The follow-up period was 24 months after radiosurgery procedure. Outcome was assessed using the scoring criteria BNI pain intensity.[31]

RESULTS

Clinical response

Treatment outcomes were obtained during follow up visits or telephone contact. The BNI pain score at baseline was V (severe pain despite medication). Fifteen days after the procedure, the patient was pain-free (BNI grade IIIb), and 24 months after the procedure, the patients is pain-free without medication (BNI grade I). No paresthesias have been reported.

DISCUSSION

Vertebrobasilar ectasia

Secondary TN is mainly related to tumors of the middle or posterior fossa. Meningiomas and neurinomas are the most common tumors in this area.[29] Intracranial arterial dolichoectasia is a rare condition of secondary TN characterized by enlargement, tortuosity, or elongation of

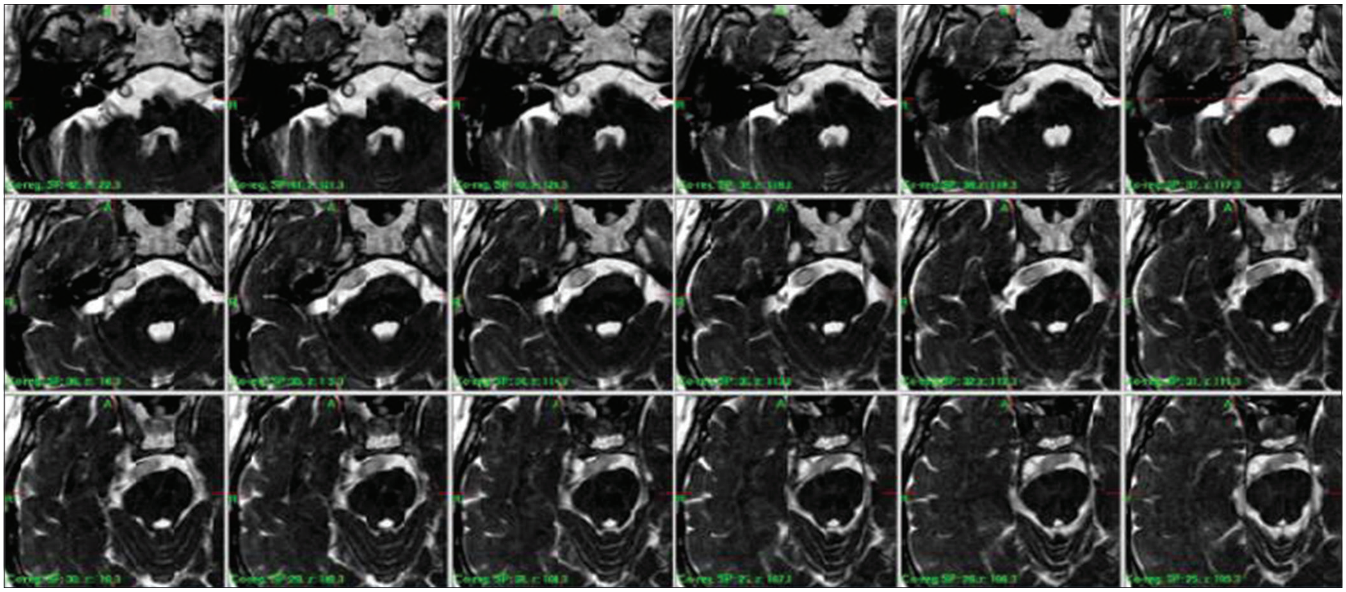

Figure 1: A sequence of FIESTA axial views shows a neurovascular conflict with a dolichoectatic basilar artery crossing and displacing pons; causing compression of the right trigeminal nerve. The nerve is not properly visualized in its entire trayectory

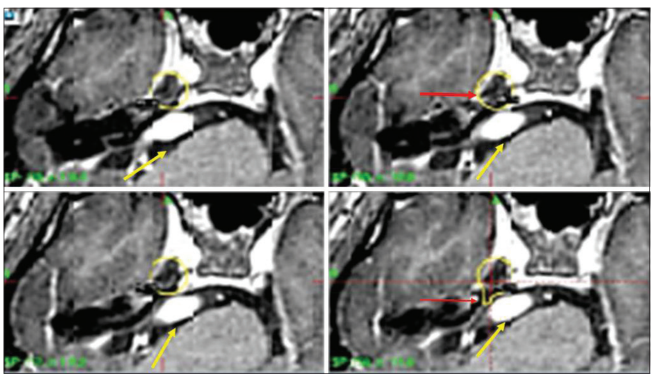

Figure 2: Tortuous vascular loops are compressing and distorting both the right trigeminal nerve and the brainstem (yellow arrows). Trigeminal ganglion (thick red arrow) and exit zone of trigeminal nerve from the ganglion (thin red arrow)

major arteries at the base of the brain. The most common localization of dolichoectasia is the vertebrobasilar system.[10]

VBE is defined when this arterial system is elongated and the basilar artery lies lateral to the margin of the clivus or dorsum sellae or if it bifurcates above the plane of the suprasellar cistern.[33] Ectasia is considered to be present if the basilar artery has a diameter greater than 4.5 mm[18] and may be related to a structural arterial wall defect of the internal elastic lamina.[38] The (VBE) compresses and displaces the trigeminal nerve and the brainstem, not allowing the visualization of the entry zone.[18]

Direct compression by Vertebro Basilal Dolico Ectasia (VBD) is an uncommon cause of TN with an estimated incidence of 1%.[6] Linskey published a large series where only 31 patients in a total of 1404 (2%) had TN caused by vertebrobasilar compression.[16]

In patients with VBD, the compression has a slowly progression, which let the brainstem can tolerate severe distortion without functional disturbances, which explain most patients with VBD are asymptomatic as in our case.[18,25] Some authors have observed that TN caused by VBE is more frequent in male elderly patients, with tendency to suffer arterial hypertension. In general, the left side is the most affected.[8]

Many patients with VBE are poor candidates for a major posterior fossa surgical procedure because of advanced age or the presence of other medical comorbidities. In such patients, other less invasive options are necessary.

Criteria for choosing the gasser ganglion

In April 20, 1953, Leksell performed the first "stereotaxic radiogangliotomy."[14] A second procedure was performed on June 11, 1953 in another patient. Both treatments were successful at 2 weeks and 1 day, respectively. Although Leksell obtained very good results with these two patients, he emphasized that no definite conclusions concerning the optimal dose of radiation or the exact mechanism and site of action in the root or ganglion should be drawn. Lindquist *et al.* reported[15] reported in 1991 the results obtained in the management of TN in 46 patients, using the gasserian ganglion as radiosurgical target. Thirteen patients were pain free after 6 months but only four remained so after 2.5 years. The target localization was performed by stereotactic cisternography and no dose options were recommended. The authors established that the imperfections in the target localization and fixation techniques may contribute to inaccuracies and unsatisfactory results.

Rand *et al.*[28] presented their experience many years after Leksell's report. Twelve patients had radiosurgery. Seven were treated with an 8-mm collimator and five with a 4-mm collimator. Target localization was determined by computed tomography (CT) or MRI. Doses varied between 57 and 75 Gy. The retrogasserian region was the target in eight cases. In four patients, the entry zone was the choice. In seven patients complete relief or improvement was observed. No complications were reported. Based on the fact that some patients did not respond, the authors concluded that the gasserian ganglion was probably not appropriate as a primary target for radiosurgical treatment. The entry zone appeared in the scene as the new target for TN radiosurgery treatment.[12,28] The TG was forgotten.

Recently, Chen *et al.*[7] presented their experience in treating 40 patients with typical idiopathic TN. The TG was the preferred target. The rational of this choice is that the TG has been the target of radiofrequency for long time and it is easy to identify by MRI. The radiosurgical treatment was performed with a Linac and the target dose used was 70 Gy with a 4-mm collimator. The isodose was not specified. They categorized the results as excellent (BNI I), good (BNI III), and poor (BNI IV-V). A total success rate of 82.8% was reported. The mean time of initial relief was 12.5 days. The pain recurrence was observed in 1 patient (3%) of the 33 patients who obtained excellent and good results.

In our case, the patient had a very intense and acute pain. He did not desire a surgical solution for the VBE and he had severe arterial hypertension and obesity. The entry zone was not accurately visualized by T1-, T2-weighted images, or FIESTA. The only well-defined and visible target was the TG and a small portion of the exit zone.

Park *et al.*[24] in their study reported 20 patients treated with GKS for TN secondary to VBE, medically refractory. The entry zone was the target of choice. Two 4-mm isocenters were used in four patients. They created an ovoid plan and covered a longer nerve segment more anteriorly, possibly due to difficulties visualizing adequately the entry zone region as in the case we presented. Eleven patients had undergone one or more surgical procedures prior to GKS. Five patients had an atypical TN. It has been well established that the best radiosurgical results for TN are obtained in patients without any kind of previous surgical interventions.[12,37] It is possible that this fact, in conjunction with the atypical cases, explains the conclusions obtained by Park *et al.*,[24] about the inferior pain control rates of GKS in their report. The patients were less likely to achieve initial adequate pain relief and were less likely to maintain pain relief. Despite initial pain improvement in most patients, additional medical or surgical pain management was required in the majority within 3 years after GKS. Another important factor is the identification on images of the trigeminal nerve, which is difficult in most cases with VBE.

Alpert *et al.*[1] evaluated the response with two isocenters and increasing dose. The radiation dose was escalated using less than or equal 80 Gy in 20 patients, 85 Gy in 21 patients, and greater or equal to 90 Gy in 22 patients. In 35 patients, two isocenters were used, yielded an oval dose distribution and included greater volume of trigeminal nerve within the high-dose area. The first 4-mm isocenter was placed adjacent to the trigeminal nerve entry zone and the second 4-mm isocenter was located 3-4 mm more distally along the trigeminal nerve and away from the brainstem and from the first isocenter. Patients had a mean maximum dose of 88.3 Gy. The majority of the patients experienced improvement in pain with low morbidity during the median follow-up period of 10 months. There were no severe complications. Five patients developed mild facial numbness. Flickinger *et al.*[5] did not detect a difference in pain relief with one shot compared with two shots in a prospective randomized study. There was no significant increase in treatment-related complication between the two groups. However, the maximum dose used was 75 Gy, which probably explained their results. The use of two isocenters in our case was determined to cover the TG and a very small and narrow exit zone of the nerve, due to the vessel compression.

Determination of treatment dose

The doses used by Leksell,[14] were 1650 and 2200 Rads. In many subsequent reports, all the doses used at the entry zone have increased and the best results were obtained with doses above 80 Gy. The trend over time has been to increase the maximum dose between 70 and 90 Gy in an attempt to achieve better pain control. Higher doses have been reported to increase the efficacy of the procedure, but with an increased risk of facial numbness.[21,26]

GKS is associated with high rates of pain control. Patients report excellent or good pain relief in more than 70% of the cases.[20,23,27,30,36]

Kim *et al.*[11] found in the analysis of 44 patients that 85 Gy brought more rapid clinical improvement without causing more complications. They concluded that 85 Gy seems to be preferable to 80 Gy, but prospective randomized trials are mandatory to get a more definite conclusion on the optimal dose for GKS of TN.

There is no dose established to be used over the TG. Chen *et al.*[7] reported good results with dose of 70 Gy in the management of their patients taking the Gasserian ganglion as a preferred target. On the other hand, we have seen good results in our patients with TN treated at the entry zone at doses of 86 Gy, with an acceptable rate of complications. Based on these results, we decided to use the same dose in this case with very good results.

Follow-up

A stretch follow-up of the patient is being accomplished with clinical examinations routinely each 4 months and with a continuous communication by phone each month. So far, the clinical status is normal after 2 years of the radiosurgical treatment. Moreover, this period is still short and we will continue observing the case on time. In addition, concern has been present in relation to the appearance of complications as anesthesia dolorosa. In a review of results and complications of percutaneous ablative techniques made by Taha and Tew,[34] they analyzed a total number of 6205 patients for radiofrequency, 1217 for glycerol rhizotomy, and 759 for balloon compression. Anesthesia dolorosa occurred in 1.5%, 1.8%, and 0.1%, respectively. Other series show rates between 0.3% and 4%, 0% and 2%, and 3 and 5%, respectively.[2-4,9,32,35] Facial numbness occurred in 98% of the patients after radiofrequency rhizotomy, in 72% after balloon compression, and in 60% after glycerol injection.[34] Thus, when anesthesia dolorosa occurs, it commonly develops weeks to months after the therapeutic procedure. One might suspect that this complication is likely to appear if the patient, at an early stage after the procedure develops numbness that is particularly disagreeable, as reported by Nashold and Rossith.[22]

Kondziolka *et al*.[13] presented a retrospectively review of 503 patients treated with Gamma knife radiosurgery. A single 4-mm collimator was used, and a maximum dose was 80 Gy to the entry zone. Sensory dysfunction was found in 11% and only 1 patient (0.19%) had a deafferentation pain. Young *et al*.[36] followed up 315 consecutive patients treated with Gamma knife using a 4-mm isocenter. All patients were treated with 90 Gy maximum dose to the trigeminal nerve. The incidence of numbness was 17.3%, higher than the incidence in most reports. No anesthesia dolorosa was diagnosed.

In this case, the patient did not have any kind of alteration in facial sensation until this moment.

CONCLUSION

Our case describes a male patient, diagnosed with TN 4 years prior to treatment. After GKS, there was resolution of symptoms within 15 days, without relapse for at least 2 years. This reports show that stereotactic radiosurgery for treatment of TN by targeting the Gasser's ganglion is a safe and effective treatment for TN secondary to compression by vertebrobasilar artery ectasia where neither the entry zone nor its path through the pontine cistern were truthfully seen in MRI studies. We will continue following-up this case to examine any possible complications later on. The developments of new case studies are necessary to support the results given in this case.

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