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Case Report

A DELAYED DIAGNOSIS OF LEFT TRIGEMINAL NEURALGIA WITH TINITITUS SECONDARY TO TRIGEMINAL SCHWANNOMA AT ORAL MEDICINE DEPARTMENT, KHYBER COLLEGE OF DENTISTRY, PESHAWAR:A CASE REPORT

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A B S T R A C T

Background: Trigeminal neuralgia (TN) greatly affects quality of life (QOL) as slightest stimulation of various trigger zones on the face and inside the mouth may trigger a painful episode, thus preventing the patient from speaking, eating, drinking, touching or washing of the face and brushing teeth. Trigeminal schwannomas are rare benign tumors that may present with TN like pain.

Case Report: 55 year old gentleman presented with severe lanciating episodic pain on the left side of his face along with tinititus in the left ear, loss of corneal reflex and sleep disturbance due to pain. The pain was earlier misdiagnosed as a tooth ache that resulted in some of his teeth being unnecessarily extracted. A multiecho and multiplanar imaging through the brain revealed 1.8 x 1.5 x 1.5cm sized T1W/T2W iso-intense mass along the left antero-lateral aspect of the pons in the region of trigeminal nerve showing intense enhancement of post-gadolinium images suggestive of trigeminal schwannoma.

Conclusion:After meticulous history and methodical clinical examination with a focus on detailed assessment of cranial nerves Magnetic Resonance Imagining(MRI) is mandatory gold standard for timely exclusion of intra-cranial space occupying lesion in patients with long history of TN.

INTRODUCTION

Trigeminal neuralgia (TN) greatly affects quality of life (QOL) as it is a sudden, usually unilateral, severe, short-lasting, stabbing, episodic pain in the distribution of one or more branches of the trigeminal nerve[1]. Since it is triggered by slightest stimulation of various trigger zones on the face and inside the mouth, this prevents the patient from speaking, eating, drinking, touching or washing of the face and brushing teeth[1,2,3]. In very rare cases, trigeminal neuralgia is secondary to other conditions including tumors, multiple sclerosis etc[1]. Patients with histories of TN usually undergo several ablative procedures or inappropriate interventions before accurate diagnosis[2,3]. As the sensory nerve supply of the teeth and jaws is the trigeminal nerve, TN is often misdiagnosed as a dental problem resulting in tooth extraction[3,4]. Magnetic Resonance Imagining is mandatory gold standard for timely diagnosis of TN secondary to intra-cranial pathologies[1,3]. Schwannomas are very uncommon benign tumors originating from the Schwann cells of the nerve sheaths that may involve any peripheral, cranial or autonomic nerve of the body with the exception of the olfactory and optic nerve[5]. Although extremely rare, trigeminal schwannomas represent only 0.07%-0.36% of all intracranial tumors and 0.8% to 8% of intracranial schwannomasneverthelessthey are the second commonest intracranial schwannoma[5,6,7,8]. They may arise from the cisternal segment, trigeminal/ Gasserian ganglion in Meckel's cave or from one of the three branches of the nerve[9]. Clinically, they usually present with facial pain, numbness and paresthesia in the distribution of one or all the divisions of the trigeminal nerve depending on the location of the tumor[10,11].

Case Report

55 year old gentleman presented with severe lanciating pain on the left side of his face since 2007 initiated by chewing, drinking and washing of the face. He also complained of tinititus in the left ear for the last few months. The pain awakened the patient from sleep during the night.On clinical examination there was no neurological deficit apart from the loss of corneal reflex. On intra oral examination he was edentulous partly due to inability to preserve his natural teeth as tooth brushing triggered pain and TN pain misdiagnosed as

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a tooth ache had also resulted in some of his teeth being unnecessarily extracted.

An MRI brain scan was advised to exclude any intra-cranial cause for his pain. A multi-echo and multi-planar imaging through the brain was performed. Post Gadolinium images were also obtained. There was 1.8 x 1.5 x 1.5cm sized T1W/T2W iso-intense massalong the left antero-lateral aspect of the pons in the region of trigeminal nerve showing intense enhancement of post-gadolinium images suggestive of trigeminal schwannoma which was causing mild compression upon the left antero-lateral aspect of the pons as shown in Figure No.1, 2 & 3 respectively.

A 1.8 x 1.5 x 1.5cm sized T1W/T2W iso-intense mass along the left antero-lateral aspect of the pons in the region of trigeminal nerve showing intense enhancement of post-gadolinium images suggestive of trigeminal schwannoma which was causing mild compression upon the left antero-lateral aspect of the pons

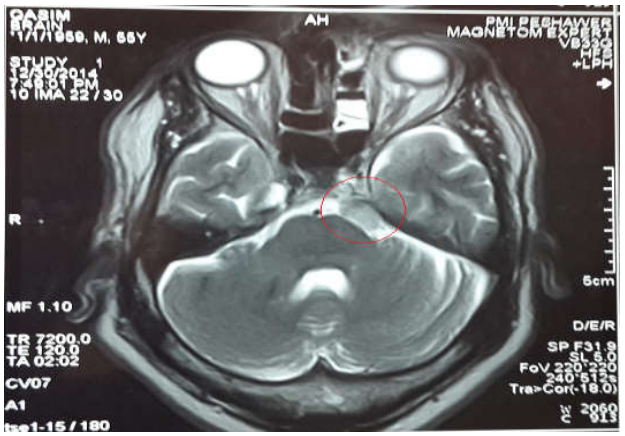

Figure No 1

Figure No 2

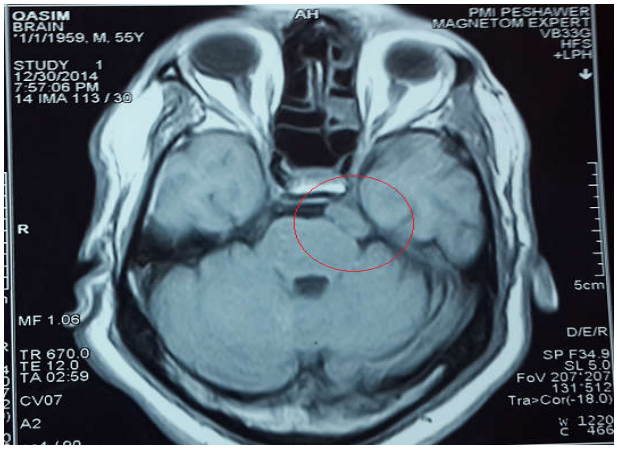

Figure No 3

Table No 1 The Dose Plan for the target

Target	Location	Prescription	Volume
A	Left Trigeminal Schwannoma	16.0 Gy @ 50%	1.9 cm^3

Table No 2 Gamma knife Data

Calibration dose	3.461 Gy/min at 2008-04-26
Days since calibration at treatment date	2477
Treatment dose rate (2015-02-06)	1.416 Gy/min
Collimator factors (4, 8, 14, 18)	0.870 , 0.956, 0.984, 1.000

Table No 3 Treatment Data

Target	Shots	Prescription	100% [Gy]	Max [Gy]	X [mm]	Y [mm]	Z [mm]	Grid [mm]
A:Lt. Tri. Sch	8	16.0 Gy @ 50%	32.0	32.0	111.7	108.9	117.6	1.0
Total numbers of shots: 8								
Verify that all shots can be achieved with Gamma Knife.								
Treatment plan name: Plan1								
Total beam-on time: 94.2 min								

Table No 4 Shot Summary

Run-Step	shot	X [mm]	Y [mm]	Z [mm]	G [deg]	Coll	Plug	Weight	Time [min]	Notes
1-1	A3	107.6	110.8	111.9	90	8	None	1.00	12.29	
1-2	A1	106.5	109.2	119.8	90	8	None	1.30	16.12	
1-3	A2	115.4	104.0	119.4	90	8	None	0.70	8.61	
1-4	A5	113.5	116.1	121.9	90	8	None	1.00	12.26	
2-1	A4	113.5	101.4	114.4	110	8	None	0.70	8.61	
3-1	A8	112.9	109.4	109.8	90	4	None	0.70	9.38	
3-2	A6	110.6	116.0	111.5	90	4	None	1.20	16.08	
3-3	A7	111.4	104.1	122.4	90	4	None	0.80	10.89	

Table No 5 Skull Geomery

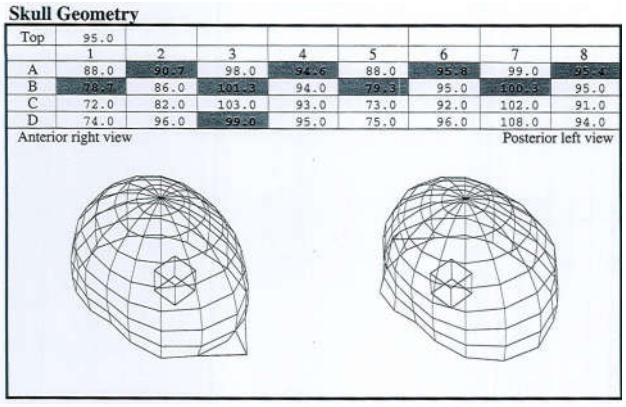

Table No. 6 Shot Dose Data

Shot	Weight	Dose rate at focus [Gy/min]	Extrapolated Skull Radii
A1	1.30	1.25	0
A2	0.70	1.26	0
A3	1.00	1.26	0
A4	0.70	1.26	7
A5	1.00	1.26	0
A6	1.20	1.16	0
A7	0.80	1.14	0
A8	0.70	1.16	0

Table No 7 Volumes

Name	Volume	Min	Max	Mean	Int. Dose	Comment
Lt. Tr. Sch	1.9 cm^3	11.0 Gy	32.3 Gy	21.4± 3.5 Gy	40.5 mJ	Plan=Plan1
Br. stem	10.8 cm^3	0.9 Gy	14.0 Gy	3.2± 1.7 Gy	34.2 mJ	Plan=Plan1

The patient was subsequently referred to Pakistan Gamma Knife & Stereotactic Radiosurgery Center, Neurospinal& Medical Institute Karachi. Gamma knife surgery procedure

was performed with Laksell Gamma Unit 4-C. Patient remained asymptomatic after two years follow-up. The Dose Plan for the target, Gamma knife Data, Treatment Data, Shot summary, Skull Geometry, Shot Dose Data,Volumes are given in Table No.1, Table No.2, Table No.3, Table no.4, Table No.5, Table No.6 and in Table No.7 respectively

DISCUSSION

Schwannomas are tumors arising from the Schwann cells in the axon myelin sheaths.Trigeminal schwannomas can arise and localize along the neurolemmalglial junction, the sensory Gasserion ganglion or along the distal branches of the nerve.These are classified into three main types depending on their location[12,13].

1. Preganglionic schwannmomas: These are confined anterior to thepons.
2. Ganglionic schwannomas: These arise from the Gasserion ganglion and are thus centred at the Meckel's cave.
3. Postganglionic schwannomas: These involve one or more divisions of the Trigeminal nerve.

Trigeminal schwannomas may present with dysesthesia, facial pain, headaches; or with symptoms caused by mass effect and compression of adjacent structures namely conductive hearing loss from Eustachian tube blockage, diplopia or hemianopsia from cavernous sinus involvement, or facial soft tissue asymmetry[14].

Jusué-Torres *et al*. in 2015 reported a case of trigeminal schwannoma presenting with dizziness, loss of balance, decreased sleep, diplopia and cognitive impairment with psychosocial problems. This was the first case of trigeminal schwannoma with obstructive hydrocephalus. Their case had mildly decreased sensation on the left face in all three branches of the trigeminal nerve with an intact corneal reflex[8]. In contrast our patient complained of trigeminal neuralgia like pain on left face and tinititus in the left ear. The pain awakened the patient from sleep during the night but there was no neurological deficit apart from the loss of corneal reflex.

Mandour*et al* in 2016 presented a trigeminal schwannoma with a progressive hearing loss, worsening right-sided facial spasms and facial numbness in the region of the right trigeminal nerve[5]. In our case the pain was left sided which is uncommon for TN and there was numbness on the affected side of the face. Although there was tinititus in the left ear, the hearing ability was still intact.

Ali M *et al* in 2009 reported 75.45% of the patients with a very long history of TN had one or multiple teeth extractions for their pain complaint without any improvement[3]. In our case the patient had most if not all of his teeth extracted in a hope of getting a relief from his pain.

In a survey of patients with TN, Tolle *et al* in 2009 reported that 90% had experienced pain for more than 1 year before receiving an accurate diagnosis, whereas 13% went 10 years without a diagnosis[15]. This observation was congruent to our case where the patient experienced pain since 2007 till he was diagnosed properly in late 2014. He was relieved of his painful condition in February 2015 when he underwent Gamma knife surgery procedure and remained pain free at two years follow-up.

CONCLUSION

For General Dentists and Maxillofacial surgeons, meticulous history and methodical clinical examination with a focus on detailed assessment of cranial nerves are most essential to avoid wrong diagnosis and inappropriate interventions in patients presenting with facial pain. MRI is the imaging modality of choice to exclude intra-cranial space occupying lesion in patients with long history of TN.

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