International Journal of Research in Medical Sciences
Saxena A et al. Int J Res Med Sci. 2023 May;11(5):1814-1816
www.msjonline.org
pISSN 2320-6071 | eISSN 2320-6012

DOI: https://dx.doi.org/10.18203/2320-6012.ijrms20231362

Case Report

Gasserian ganglion metastases of oral squamous cell carcinoma in the absence of loco-regional metastases masquerading as trigeminal neuralgia

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Received: 04 January 2023
Accepted: 31 March 2023

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ABSTRACT

We presented an unusual case of a 43-year-old man presenting with symptoms of trigeminal neuralgia with a clinico-radiological diagnosis of maxillary nerve schwannoma. Surgical excision and histopathology revealed a diagnosis of metastases of squamous cell carcinoma to Gasserian ganglion. On further investigation, the primary tumour was detected in the right alveolar region of mandible. Our purpose of presenting this case was to make the clinicians aware that all cases presenting with trigeminal neuralgia may not be benign and may be the sole presentation of brain metastases of oral cancer without the loco-regional spread.

Keywords: Trigeminal neuralgia, Schwannoma, Squamous cell carcinoma metastasis, Gasserian ganglion

INTRODUCTION

Review of the literature showed that brain metastases of oral squamous cell carcinoma in the absence of loco-regional spread are extremely rare accounting only for about 0.4% and 2-8% for those patients who already have distant spread to the lungs or other extracranial sites. The distant spread usually occurs late in the course of the disease and typically in the setting of advanced local-regional disease.[1-8]

We presented an unusual case of a 43-year-old man presenting with symptoms of trigeminal neuralgia with a clinico-radiological diagnosis of maxillary nerve schwannoma. Surgical excision and histopathology revealed a diagnosis of metastases of squamous cell carcinoma to Gasserian ganglion.

CASE REPORT

A 43-year-old man presented to neurology out patient department (OPD) in March 2020 with complaints of headache in right temporal region, loss of sensation in right cheek, V1, V2 pain and hypoesthesia in right side of face with no motor weakness for 3 months. Magnetic resonance imaging (MRI) brain revealed enhancing mass lesion in the right middle cranial fossa or maxillary nerve schwannoma. After a written informed consent and pre-operative evaluation, the patient underwent right fronto-temporal craniotomy with sub temporal extradural approach for tumor excision and tumour specimen was submitted for histopathological examination. Intra-operatively tumor was firm, non suckable, mildly vascular and adherent to trigeminal nerve and right internal carotid artery. Post-operative period was uneventful. Post-operative CT-scan showed post-

operative changes in right parieto-temporal region with mild hemorrhage and edema in right temporal lobe.

Histopathologic examination of the tissue revealed the tissue composed of nerve bundles and ganglion cells that were infiltrated by a malignant tumor in cords and islands (Figure 1A). Tumor cells were moderately pleomorphic, had vesicular nuclei and eosinophilic cytoplasm. Few keratin pearls were seen with foci of calcification. Immunohistochemical stains showed diffuse and strong positivity for Pan cytokeratin (Figure 1B). S100 immunostain highlighted the nerve bundles and synaptophysin immunostain highlighted the ganglion cells and schwann cells in between the tumor tissue (Figure 1C). Ki67 was increased in the epithelial islands of tumor tissue. Based on histology and immunohistochemistry, a diagnosis of metastases of squamous cell carcinoma (SCC) to Gasserian ganglion, likely primary in oral cavity, was rendered.

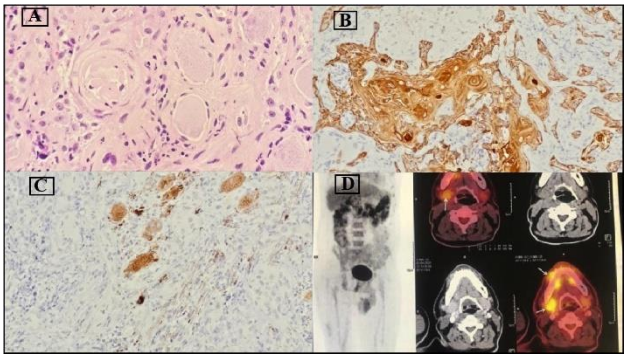

Figure 1: (A) Nerve bundles and ganglion cells infiltrated by SCC cells in cords and islands tumor cells are moderately pleomorphic having vesicular nuclei and eosinophilic cytoplasm and keratin pearls. (Hematoxylin & Eosin stain×20 ×); (B) tumor cells showing diffuse strong positivity on immunohistochemistry for Pan cytokeratin; (C) synaptophysin immunostain highlighting the ganglion cells and schwann cells in between the tumor tissue; (D) PET CT scan shows FDG avidity along the course of mandibular nerve extending from the right foramen ovale, traversing along the right mandibular foramen reaching up to right mental foramina of mandible (along the inferior alveolar nerve) with widening and irregularity of the foramen ovale and right mandibular foramen (perineural spread of disease).

In view of histological diagnosis of squamous cell carcinoma, an oncology opinion was sought. Clinical examination did not show any visible growth in the oral cavity and PET CT scan was advised. PET CT scan revealed FDG avidity along the course of mandibular nerve extending from the right foramen ovale, traversing along the right mandibular foramen reaching upto right mental foramina of mandible (along the inferior alveolar nerve) with widening and irregularity of the foramen ovale and right mandibular foramen (perineural spread of disease) (Figure 1D). Operated area in the PET CT showed metabolically active ill-defined soft tissue thickening in the right cavernous sinus region involving Meckel's cave with mild thickening of dura covering the right temporal lobe (post surgery residual disease). Asymmetrical FDG avidity was also noted in bilateral submandibular glands along with small soft tissue thickening in right lower alveolar region lateral to second premolar.

All these findings were indicative of a primary carcinoma arising from lower alveolar region of right mandible that was missed earlier.

The patient was advised radiotherapy and after one year of follow up the patient is symptom free.

DISCUSSION

Our patient presented with symptoms of classical trigeminal neuralgia with a diagnosis suggestive of maxillary nerve schwannoma on MRI. Classic trigeminal neuralgia is characterized by sudden, recurrent, brief stabbing pain, involving the distribution of one or more branches of the trigeminal nerve (TN). Atypical painful trigeminal neuropathy implicates secondary structural causes, such as demyelinating plaques, trauma, infection, and skull base lesions, affecting the fifth cranial nerve. Inflammatory diseases, including neurosarcoid, collagen vascular disease and neoplastic infiltration can result in pain, sensory and motor dysfunction of TN.[5,6]

TN is most vulnerable for perineural invasion (PNI) as it gives cutaneous innervation to major region of the head and neck. PNI is considered as a distinct third mode of tumor metastasis together with lymphatic being number one and vascular being second mode of invasion. PNI is a common pathologic finding in many head and neck cancers, including OSCC and adenoid cystic carcinoma.[3,8]

TN is the largest cranial nerve that can be divided into five segments: intra-axial (brain stem), cisternal, Meckel's cave and cavernous sinus, skull base and extra cranial segments. The Gasserian or semilunar ganglion is located in the inferior aspect of Meckel's cave and gives off three branches: ophthalmic (V1), maxillary (V2) and mandibular (V3).[7]

In our patient, considering the age and neuralgic symptoms, initially any possibility of neoplastic infiltration of cranial nerves causing the symptoms of trigeminal neuralgia was not considered by the neurologist. Only after surgery, on histological examination it came as a diagnostic surprise showing metastases from a SCC to Gasserian ganglion. Histology combined with PET CT findings helped us in arriving at a diagnosis of primary in the lower alveolar region of right mandible with perineural spread traversing along the mandibular nerve to the Meckel's cave and to the

Gasserian ganglion. There was conspicuous unusual absence of loco-regional lymphadenopathy.

After a detailed clinical history on asking leading questions, the patient told that he had difficulty in opening the mouth which he thought was due to neuralgic pain. Now thinking in retrospect, this possibly could have been due to a pre-existing sub mucous fibrosis progressing to oral cancer and was missed initially. The patient reported only when he had neuritic pain.

Rao et al 2020 in an exhaustive review of published studies on oral sub mucous fibrosis from the year 1966-2019, have emphasized on the need of early diagnosis of oral sub mucous fibrosis specially in pan masala and gutkha chewers, a habit quite prevalent in Indian subcontinent. According to the review, current estimates showed that areca nut was consumed by 10-20% of the World's population in a wide variety of formulations. The global South Asian diaspora also has a significant problem with cases reported from the United Kingdom, USA, South Africa, and many European countries. Therefore, detailed clinical history is of single most important clue to the correct diagnosis.[9]

The painful trigeminal neuropathy may present with diagnostic difficulty in the absence of a known primary. Changes in neuroimaging may be missed or misinterpreted in absence of a clinical suspicion of a primary or metastasis. Therefore, detailed clinical history, personal habits and clinical assessment of the patient are of utmost importance which can give a clue to the presence of an underlying malignancy.

CONCLUSION

To conclude, the present case with neoplastic perineural involvement of trigeminal nerve by OSCC in the absence of regional lymph nodes, clearly was a case of diagnostic miss. In countries like India, where sub mucous fibrosis is common with tobacco and pan masala chewers, possibility of oral sub mucous fibrosis must always be kept in mind as an important cause in all patients presenting with trigeminal neuralgia with difficulty in opening the mouth and the patient should be investigated accordingly.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Saxena A, Singhal N, Rath A, Singh V, Shankar R, Goel MM. Gasserian ganglion metastases of oral squamous cell carcinoma in the absence of loco-regional metastases masquerading as trigeminal neuralgia. Int J Res Med Sci 2023;11:1814-6.