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Case report

Malignant peripheral nerve sheath tumor of the tongue with an unusual pattern of recurrence

Soumyajit Roy MD^a, Ajeet Kumar Gandhi MD^{b,*}, Bharti Devnani DNB^a, Lavleen Singh MD^c, Bidhu Kalyan Mohanti MD^a^a Department of Radiation Oncology, All India Institute of Medical Sciences, New Delhi 110029, India^b Department of Radiation Oncology, Rajendra Institute of Medical Sciences, Ranchi 834009, India^c Department of Pathology, All India Institute of Medical Sciences, New Delhi 110029, India

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ABSTRACT

Malignant peripheral nerve sheath tumor (MPNST) of oral cavity is an extremely uncommon malignancy. Less than 15 cases have been reported since 1973 though none of them describes a distant metastasis. We present a rare case of MPNST of the tongue who presented with features of hypoglossal nerve palsy. Incisional biopsy showed a malignant spindle cell tumor in the sub-epithelial connective tissue. The tumor cells were immune-positive for S-100. He underwent surgery followed by adjuvant chemotherapy. Later the disease recurred in the form of isolated pelvic bone metastasis. Palliative chemotherapy was offered to him. With this case report we intend to refer to such unusual presentation and pattern of recurrence in a MPNST of tongue.

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Introduction

Malignant peripheral nerve sheath tumors (MPNSTs) account for 5–10% of soft tissue sarcomas and 6–16% of head and neck sarcomas [1]. Almost 50% of MPNSTs are associated with type 1 neurofibromatosis (NF1) and this shows the tendency of this tumor to arise from pre-existing neurofibroma. It has been suggested that the molecular pathways for development of NF1 and MPNSTs are not the same. NF1 does not independently cause MPNSTs but increases the predisposition of the patients for such an event [2]. Higher incidence has also been reported in patients with history of radiation exposure [3]. Median age for occurrence of MPNST is between 30 and 50 years for sporadic cases and 20–40 years for NF1 associated cases. No sex predilection has been reported for this disease [4]. Patients usually present with a progressive swelling which may be painful and associated with neurological symptoms like paresthesia or weakness. Sudden enlargement or new, worsening and persistent pain in a pre-existing neurofibroma is also a pointer towards possible development of MPNSTs. Surgical resection is the primary treatment modality. Currently, radiotherapy is

also recommended as an adjuvant treatment modality [5]. We highlight a rare case of MPNST of the tongue with an unusual pattern of recurrence.

Case report

A 30 year old man presented to our multi-disciplinary head and neck cancer clinic with a painful, slowly progressive exophytic growth over the right lateral border of tongue for two years. He also had associated history of severe odynophagia for 16 months. On examination, the tongue was deviated to right side and fasciculation was noted over the right half of the tongue. These features were suggestive of right hypoglossal nerve palsy. There was a 10 × 9 cm growth over the posterior third of right lateral border of tongue involving its full thickness. The growth was infiltrating into the floor of mouth and ipsilateral tonsillo-lingual sulcus. Neck examination and rest of the systemic examination did not reveal any abnormality.

A contrast enhanced magnetic resonance imaging (CEMRI) of the face and neck revealed evidence of an asymmetric altered signal intensity lesion at the posterior third of right side of the tongue appearing hyper intense on T2 weighted images. However, no specific nerve involvement could be identified (Fig. 1).

Radiograph of chest did not show any abnormality. Incisional biopsy from the growth was done. Microscopic examination showed a malignant spindle cell tumor. The tumor cells were

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* Corresponding author at: Department of Radiation Oncology, 305, Academic Block, Dr Ram Manohar Lohia Institute of Medical Sciences, Vibhuti Khand, Gomti Nagar, Lucknow 226010, India.

E-mail address: ajeetgandhi23@gmail.com (A.K. Gandhi).

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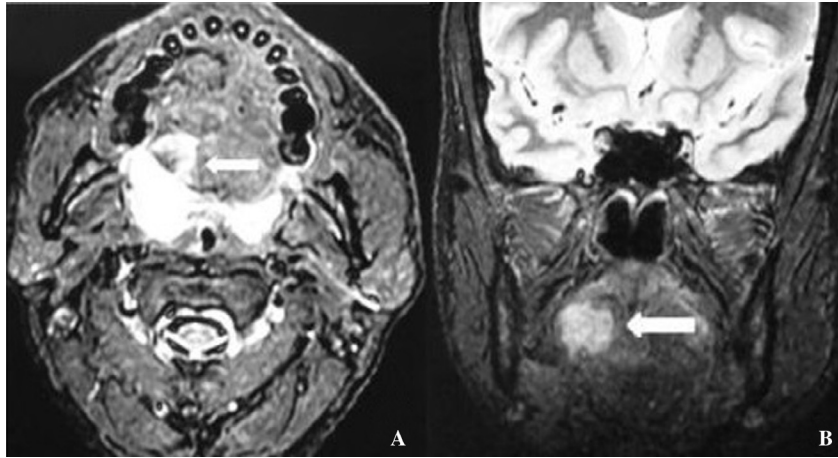


Fig. 1. Axial T2W TSE fat suppressed image reveals a hyper-intense mass in the right half of the posterior one third of tongue (arrow). The mass is not extending till the midline septum and the neurovascular bundles are intact (1A). Coronal T2W TSE fat suppressed image shows the mass in the right half of the oral tongue (arrow). The mass is infiltrating into the floor of mouth (1B).

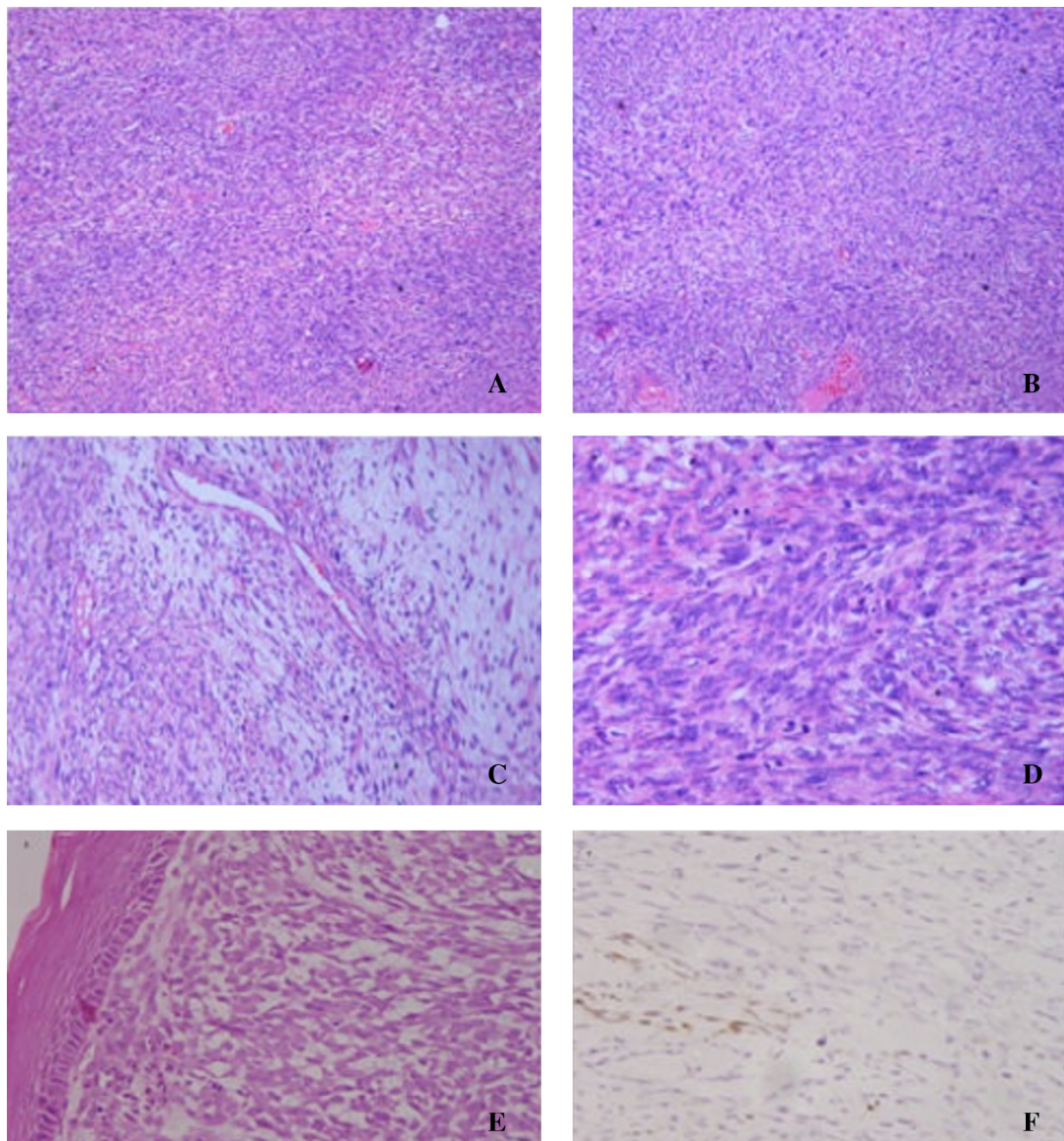


Fig. 2. A malignant spindle cell tumor of tongue with mitotic activity in the sub epithelial connective tissue (HE, X4) (2A) (HE, X10) (2B) (HE, X 20) (2C) (HE, X40) (2D) (HE, X200) (2E). Neoplastic cells are arranged in cellular fascicles and showing nuclear palisading. On IHC they are focally Immuno-positive for S100 (2F).

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