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Case Report

Spindle cell squamous carcinoma of the tongue in a child

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ABSTRACT

Background: Spindle cell carcinoma (SpCC) is an infrequent and aggressive type of squamous cell carcinoma (SCC) characterized by the proliferation of epithelial and mesenchymal components. Oral SCC in children is an extremely rare entity and the SpCC variant has been reported in one case in the paediatric patient literature.

Methods: In this paper, we report a case of SpCC of the tongue in an 11-year-old boy treated by on-block surgical resection and microvascular tissue reconstruction.

Results: After 14 months the patient is free of disease with easily intelligible speech and normal swallowing.

Conclusions: Diagnosis and treatment of this rare tumour in this age group is a challenge because of the overlapping of histopathological features and the complex reconstruction required to achieve adequate aesthetic and functional results.

1. Introduction

Spindle cell carcinoma (SpCC) is a rare, aggressive, and poorly differentiated variant of squamous cell carcinoma (SCC) representing 3% of SCC in the head and neck region.

A far as we know, there is only one case reported in the literature of spindle cell carcinoma in a paediatric patient (younger than 20 years). This would be the second case described.

We present a case report of this unusual tumour in an 11-year-old boy, treated by on-block surgical resection and microvascular tissue reconstruction in order to contribute in part to a better understanding and management of this rare malignancy in this age group.

1.1. Case report

An 11-year-old boy was referred to our Department of Paediatric Oral and Maxillofacial Surgery because of a lesion on the right posterior border of the tongue that had lasted over a period of three months with some interruptions.

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The patient reported mild dysphagia and difficulty in lingual extrusion over the last 90 days. No weight loss or other general symptoms were reported.

Physical examination revealed an ulcerated lesion in the right posterior margin of the tongue, a slightly painful lesion on palpation and limitation of oral opening movements (see Fig. 1). There was no palpable cervical lymphadenopathy.

Magnetic resonance imaging (MRI) identified a lesion with the following approximate dimensions: 2× 1.4 × 1.6 cm (AP x T x L) on the right hemitongue, which was moderately hyperintense on T2-weighted images and discretely hypointense on T1, with indistinct edges and strong contrast. It affected mainly the intrinsic muscles of the right hemitongue, with the hyoglossus muscle acting as an anatomic barrier. Extrinsic muscles (genioglossus and mylohyoid muscles) remained unaltered. The MRI scan showed a probably reactive bilateral submandibular lymphadenopathy.

The patient provided a pathological report of a biopsy performed previously at a different hospital. The diagnosis was a poorly differentiated carcinoma with squamous features. A second biopsy showed on microscopic examination malignant spindle cell neoplasm suggestive of sarcoma but unable to rule out other options such as spindle cell carcinoma.

Because of the biopsy diagnosis, we decided to perform an extension study with PET-CT. It was reported as a pathological metabolic focal increase SUVmax 6.18 in the posterior third of the right border of the tongue. Bilateral cervical lymphadenopathy was described as lymph nodes less than 1 cm in diameter at levels II A, II B and submandibular. None of them presented any increase associated with metabolic disease (see Fig. 2).

The case was assigned to the Head and Neck Tumours Multidisciplinary Unit. We opted for a surgical approach to try en bloc resection with clear margins. The surgery was performed through a right cervicotomy for an ipsilateral limited supraomohyoid neck dissection, extending to middle labiomandibulotomy for a mandibular swing approach, in order to perform a right hemiglossectomy preserving the lingual tip and the base of the tongue with wide surgical margins. A temporary tracheostomy was performed to ensure the airway in the immediate postoperative period (see Fig. 3).

Reconstruction was completed with a right lateral thigh fasciocutaneous flap.

During follow-up the patient was decannulated on the fifth day postoperatively. Tolerance began on the sixth day with liquids and the tenth day with solids at the start of rehabilitation for swallowing and speech.

The surgical specimen showed an ulcerated plaque on its dorsal surface of 5 × 4.5 × 2.5 cm. The microscopic examination revealed a tumour mass connected to the epithelial surface of the tongue. The more superficial layers of cells showed epithelioid differentiation whereas the more profound area, resembled sarcomatoid cells. The cells were spindled and formed fascicles in a somewhat storiform pattern. It infiltrated the muscle, with a maximum tumour depth of 0.5 cm. It did not affect the resection margins. The immunohistochemistry techniques showed positivity for cytokeratin AE1-AE3, Vimentin and EMA. Desmin, Smooth Muscle Actin (SMA), Calponin and CD34 were only partially positive. The tumoral cells were negative for S100, p63, Muscle Specific Actin (HHF35), Caldesmon, CD31, BCL2 and CD99. The proliferation rate was 15% (see Fig. 4).

The histological and immunohistochemical profile was consistent with the diagnosis of stage I Spindle-cell carcinoma (SpCC), according to AJCC guidelines.

The case was presented again in the multidisciplinary unit with no other adjuvant criteria.

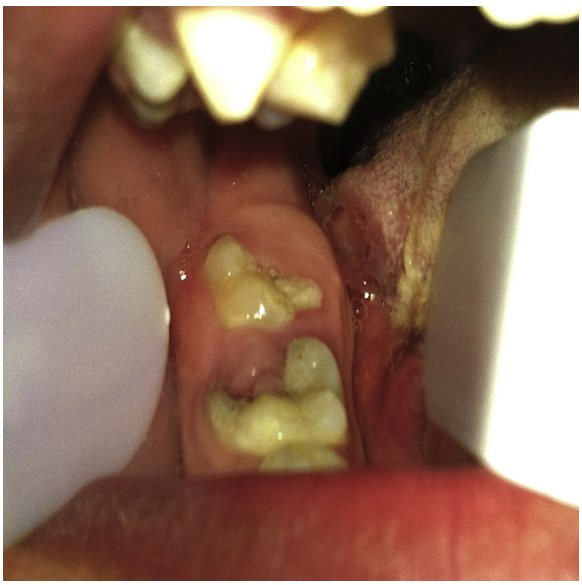

Fig. 1. Primary lesion: A ulcerated lesion on the right posterior mobile edge of the tongue slightly painful on palpation without bleeding.