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Case Report

Maxillofacial fibrosarcoma arising from dermatofibrosarcoma protuberance: A case report and review



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ABSTRACT

Fibrosarcoma is an infrequent malignant fibroblastic tumor of maxillofacial region, and it can be a rare event in dermatofibrosarcoma protuberance (DFSP). It is locally aggressive but has low incidence of locoregional lymph node and/or distant hematogenous metastases. We report here a case of a 52-year-old Somali woman with a massive tumor on her right maxilla, with repeated recurrences after surgical excision. Histological differentiation of this spindle-shaped tumor cells provide some diagnostic challenges as most of these fibroblastic lesions are lacking specific immunohistochemical markers and typical histomorphological pattern may be absent.

The findings of the present case are being discussed with existing literature.

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1. Introduction

Fibrosarcoma is a rare tumor in the maxillofacial region. Only 10% of this malignant tumor occurs within this area [1]. In this region, the neck area is the most common site for fibrosarcoma (25%), next is the face (20%), followed by the scalp (16%), maxillary sinuses (12%) and intraoral region (12%) [2]. Intraorally, fibrosarcomas may have affected the pharynx, palate, lips or periosteum of the maxilla and mandible [3]. Although fibrosarcomas is a locally aggressive tumor with an invasive growth, it has rare locoregional lymph nodes and/or distant hematogenous metastases. An inadequate surgical margin is one of the important contributing factors of the high recurrence rate of fibrosarcoma in the head and neck region [7]. Histological differentiation of this spindle-shaped tumor cells provide some diagnostic challenges as most of these fibroblastic lesions are lacking specific immunohistochemical markers and typical histomorphological pattern may be absent.

The main objective of reporting this paper is to describe a rare case of maxillofacial fibrosarcoma arising from dermatofibrosarcoma protuberance (DFSP) with a discussion on making the definitive diagnosis. We will also review some reported cases of fibrosarcoma in the maxillofacial region.

2. A case report

A 52-year-old Somali woman was seen at the Otolaryngology Out-Patient Clinic, Universiti Kebangsaan Malaysia Medical Centre (UKMMC), complaining of 6 months history of rapidly enlarging right cheek mass associated with gross facial deformity and oral dysphagia. The swelling was painless and recurred despite three previous attempts of excisions done elsewhere. She had no significant constitutional symptoms or any other symptoms to suggest an involvement of neighboring cranial nerves nor airway compromise. Examination revealed a bulky, nodular, firm mass was seen arising from the right maxillary region, tethered to the overlying skin. It measured 17 cm × 12 cm × 18 cm in its largest dimensions. Superiorly, it has displaced the right eyeball, however, her right vision is still intact and laterally it has pressed the lateral wall of the nose. While inferiorly the tumor extended to the angle of the mandible as well as eroding the entire upper lip (Fig. 1A). Intraorally, the lesion occupied the entire right buccal mucosa and space. Otherwise, no other masses were seen in the upper aerodigestive tract.

[☆] AsianAOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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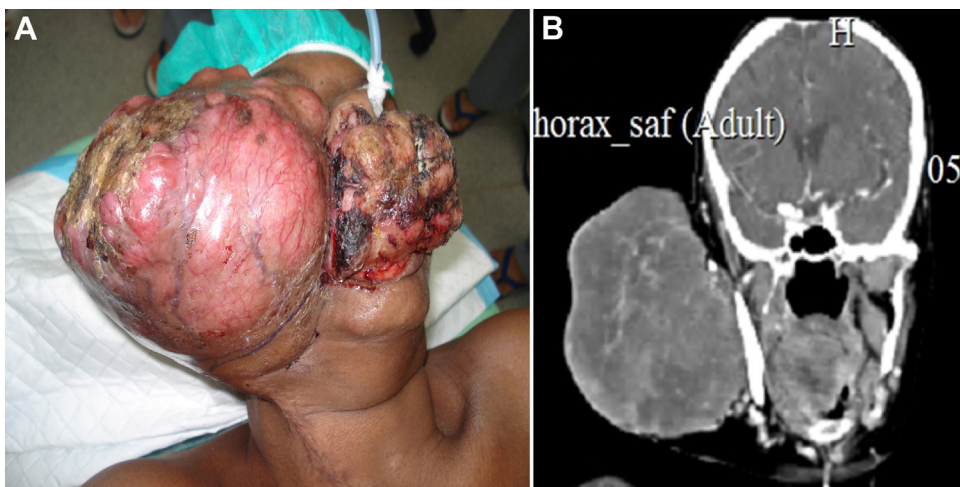


Fig. 1. Fibrosarcoma arising from fibrosarcoma protuberance in the maxilla of a 52-year-old woman. (A) Facial view and (B) coronal computed tomography (CT) image. The lesion was a bulky, nodular, and firm mass arising from the right maxillary region, where the tumor extended to the mandibular angle and eroded the entire upper lip. CT image showed a large fungating tumor attached to the right side of the facial region.

No palpable cervical lymphadenopathy was present nor any signs suggesting distant metastasis to the lungs, liver or vertebrae. Computed tomography (CT) scan revealed a fungating mass arising from the right side of the face destructing all walls of the right maxillary sinus, superiorly eroding the floor of the orbit and posteromedially involving the infra-temporal fossa (Fig. 1B). Excision of the tumor with anterolateral thigh free vascularized flap reconstruction was performed and the tumor weighing 1.7 kg. Unfortunately, patient's post-operative recovery was complicated with flap failure. Two further attempts of pedicled flap were

required to cover the huge defect created by the initial excision. The patient was discharged to Somalia with no known further recurrences.

Upon sectioning, the mass exhibited white whorled appearance. Histomorphologically, there were two distinct patterns within the tumor mass, one of which showed a hypercellular area composed of spindle cells fascicles arranged in herring-bone pattern (Fig. 2C). This hypercellular area was present throughout the mass. The tumor cells were generally monomorphic and displayed spindle-shaped hyperchromatic to vesicular nuclei, inconspicuous nucleoli

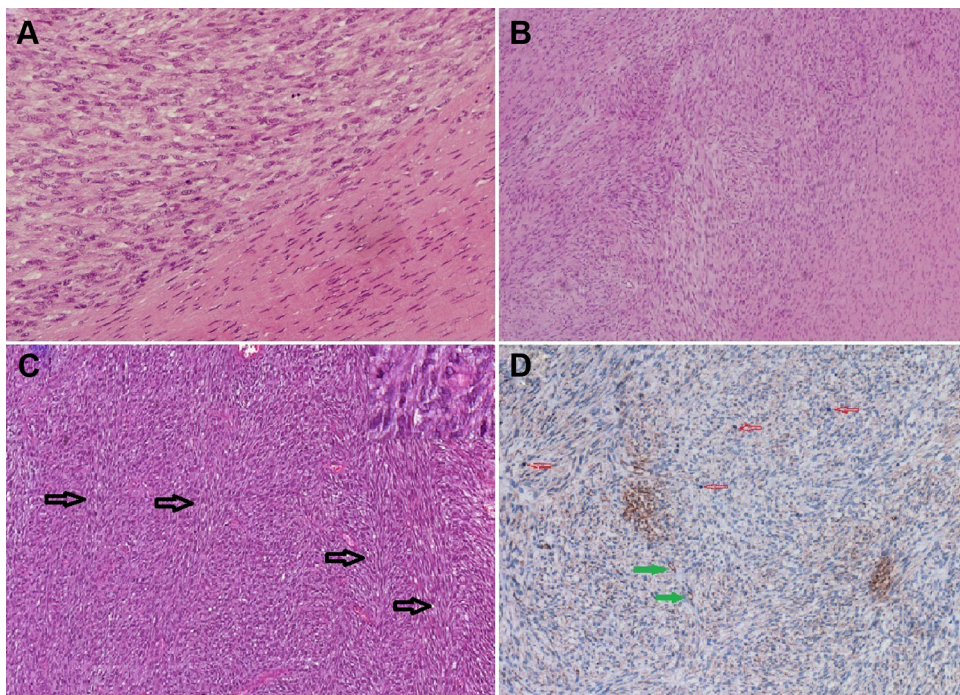


Fig. 2. Micropictures of the tumor in H&E and immunohistochemistry stainings. (A) Transition zone; (B) feature of dermatofibrosarcoma protuberance (DFSP) within the tumor; (C) feature of fibrosarcoma; (D) immunohistochemistry staining of CD34. The transition zone in (A) exhibited the less cellular DFSP area in fascicular pattern with the very cellular fibrosarcoma (magnification 200 \times). While the DFSP area (B) showed a more typical storiform pattern as seen on the left-hand side area (magnification 100 \times). An H&E microscopic view of the fibrosarcoma (C) showed fascicles of spindle-shaped cells arranged in herringbone pattern (arrows). These spindle cells are rather uniform and have ovoid, basophilic nuclei, coarse chromatin and pale cytoplasm (magnification 100 \times). There were many mitotic figures as depicted in the higher power picture (magnification 400 \times) inserted on top right. CD34 (D) showed weak positivity with some focal strong immunostain. High mitotic activity can be observed in the upper half of this section (red arrows) while internal positive stains can be seen at the endothelium-lined channels (green arrows) (magnification 100 \times). (For interpretation of the references to color in this figure legend, the reader is referred to the web version of the article.)

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