ISSN: 2230-9926

Available online at http://www.journalijdr.com

IJDR

International Journal of Development Research
Vol. 5, Issue, 03, pp. 3836-3839 March, 2015

International Journal of
DEVELOPMENT RESEARCH

Full Length Research Article

INTRAORAL FIBROSARCOMA OF TONGUE: AN UNUSUAL CASE REPORT WITH REVIEW

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ARTICLE INFO

Article History:
Received 11th December, 2014
Received in revised form
15th January, 2015
Accepted 16th February, 2015
Published online 31st March, 2015

Key words:
Fibrosarcoma,
Herringbone pattern,
Immunohistochemistry,
Intraoral, Tongue

ABSTRACT

Sarcomas are the rare malignant neoplasms originating from the mesodermal tissues that constitute connective tissues of the body. Among these, fibrosarcoma constitutes 5-15% of all sarcomas present in the head and neck region. It is the neoplastic lesion of malignant fibroblasts that shows no other evidence of cellular differentiation and is capable of recurrence with metastasis. The most common anatomic site of occurrence of this tumor is deep soft tissues of lower extremities and in the head and neck region, intraoral adult fibrosarcoma is very rare with no particular sex predilection. Therefore this article discusses a case report of fibrosarcoma which is arising from the ventral surface of the tongue and the diagnostic difficulties associated with it.

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INTRODUCTION

Fibrosarcoma (FS) has been defined as a malignant neoplasm of mesenchymal cell origin in which histologically the predominant cells are fibroblasts that divide excessively without cellular control; they can invade local tissues and travel to distant body sites (metastasize) (1). FS is an uncommon neoplasm in the head and neck region and constitutes about 1% of all the malignancies affecting the human race (Tupkari *et al*., 2014). This rare tumor can occur anywhere in the body but most common in deep soft tissues of the lower extremities, particularly the thigh and knee followed by the upper extremities and trunk (Weiss and Goldblum, 2008). Only 23% cases of head and neck fibrosarcomas occur within the oral cavity. Therefore a case of primary soft tissue FS arising from the ventral surface of tongue of 57-year-old male patent is documented here, which will be an addition to the cases reported earlier in the literature.

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Case report

A 57-year-old male patient presented with a swelling on the ventral surface of tongue. The swelling was first noticed 2 years back, which slowly increased to the present size. There was no pain associated with it but a discomfort during the mastication. The family history of patient was non-contributory. On physical examination, he was healthy and hematological findings were within the normal limits. Extra-oral examination did not reveal any significant observations. On intraoral examination, a solitary mass (Figure 1) was noticed, measuring around approximately 3 cm X 5cm in dimensions and present on the ventral surface of tongue in relation with 31, 32, 33, 41, 42 and 43. There was no evidence of paresthesia on the affected site and the overlying mucosa appeared slightly ulcerative. Based on the history and clinical findings provisional diagnosis of traumatic fibroma, salivary gland neoplasm and malignant soft tissue tumor neoplasm was given. The incisional biopsy was performed. A macroscopic examination showed multiple bits of soft-tissue specimens, which were irregular in shape and soft in consistency. Microscopic examination of Hematoxylin and Eosin (H and E) stained section showed highly cellular lesional tissue consisting of numerous malignant spindle shape cells with

elongated nuclei, showing fascicles, whorls and a palisaded arrangement and were separated by interwoven collagen fibers arranged in parallel fasion. At many places herringbone pattern (Figure 2) was also evident. These cells were vary in size and shape and showed scanty cytoplasm with indistinct borders.

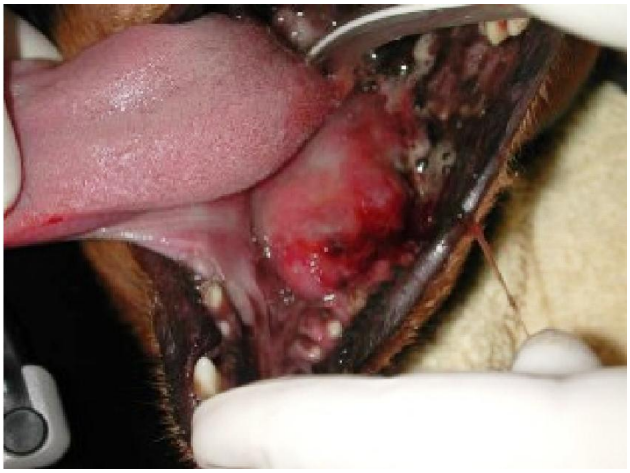

Figure 1. Intraoral photograph shows nodular swelling on the ventral surface of tongue

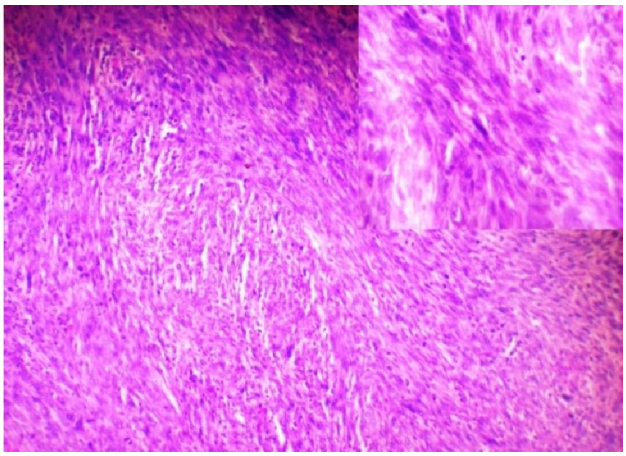

Figure 2. Herringbone pattern of malignant spindle shaped cells separated by interwoven collagen fibers (H and E, original magnification 10 X) (Cells in fibrosarcoma having irregular, spindle shape with elongated nuclei under higher magnification [inset])

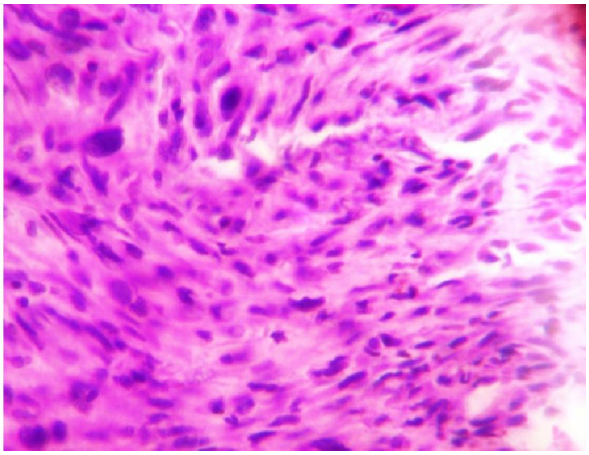

Figure 3. Malignant fibroblasts showing large pleomorphism with enlarged hyperchromatic nuclei and mitotic figures (H and E, original magnification 40X)

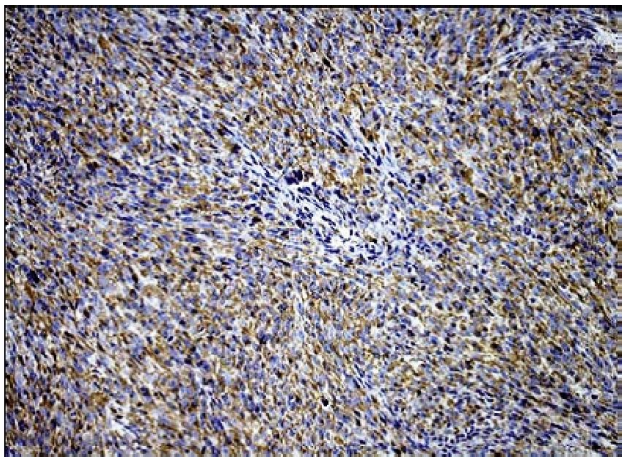

Figure 4. Mesenchymal malignant spindle cells showing diffuse and intense positivity with vimentin (Immunohistochemical stain, original magnification 40X)

The nuclei were hyperchromatic, pleomorphic and showed varied mitotic figures (Figure 3). The histopathological findings were suggestive of Low- grade fibrosarcoma. Immunohistochemical stain helped to confirm the diagnosis as the mesenchymal component stained strongly and diffusely with vimentin (Figure 4). Tumor cells were negative for cytokeratin, smooth muscle actin (SMA), S100. Correlating the clinical, histopathological and immunohistochemical investigation, a final diagnosis of Low-grade fibrosarcoma was given. Patient was referred to the "Tata Memorial Hospital and Cancer Research Center" for further evaluation, management and advised post-operative short term with long-term follow-up visits.

DISCUSSION

Fibrosarcoma subjectively termed as a sarcoma which is showing spindle cells, collagen forming cells within lesional tissue. FS was defined by Rokitansky (1842) as a tumor with varieties depending on 'the form and arrangement of fibers'. Various attempts have been made to classify fibrosarcoma under differential categories. Wilks (1849) considered it as fibroblastic whereas Borst (1902) labeled it as fibroma sarcomatosum. Later on Ewing (1940) described fibrosarcoma, spindle cell sarcoma or fascial sarcoma under the term nerogenic sarcoma; as he believed that the majority of them are of neurogenic origin (Kumar *et al*., 1990). The etiology and pathogenesis of FS was not clear. The repeatedly trauma was considered as one of the factor because the predominant cells seen within the lesional tissue are fibroblasts (Delpla *et al*., 1998). Reported a non random chromosomal change involving t (2; 19) with involvement of 2q21-qter (Limon *et al*., 1998). Factors other than trauma have also been implicated as prosthetic vascular graft (Eckstein *et al*., 1992). Clinically most of the patient showed deep seated mass that is painful upto one third of cases. The most common location is lower extremities (45%) followed by upper extrimities (28%), trunk (17%) and head neck region (10%) (Meis- Kindblom *et al*., 1995; Antonescu and Baren, 2004 and Battiata and Casler, 2005). Soft tissue FS are very rarely seen within the oral cavity. The age range is wide seen in 3rd to 5th decades of life and most studies have reported a male gender predilection (Meis- Kindblom *et al*., 1995 and Antonescu and

Baren, 2004) which is in accordance with our case report. Many patients presented with a solitary palpable growth ranging from 3-8cm in greatest dimensions. It is slowly growing and painless mass, sometimes showing ulcerations with fungating overgrowth in the area of ulceration. The present case was of 57 year old male patient which showed a slightly ulcerative growth on the ventral surface of tongue which is very unusual. Grossly, tumor can be misdiagnosed as benign neoplasm as most of them are well encapsulated and circumscribed. Histopatholgically, FS has been classified as 1. Adult Fibrosarcoma (variants such as classic type, mixoid type, fibromixoid type, sclerosing epitheloid type), 2. Juvenile/ Infantile fibrosarcoma (Weiss *et al*., 2008). Classic adult fibrosarcoma showed neoplastic cells predominantly epitheloid in appearance and arranged in variety of patterns like nests, chords, strands, sometimes acini or alveolar. The histologic grading of FS is based on the cellularity, differentiation, mitotic activity and necrosis as low-grade, intermediate grade and high grade. The present case showed herring bone pattern of neoplastic cells with minimal mitotic figures suggestive of low grade FS.

Virtually all FS stained positive for vimentin, only 50% have epithelial membrane antigen positivity that creates confusion with carcinoma, epitheloid sarcoma and synovial sarcoma. This tumor cells are strongly negative for cytokeratins, neural markers {like S100 and neuron specific enolase}, desmin, SMA, HMB45, CD68, CD 34 (Weiss *et al*., 2008). The present case report showed strong positivity for vimentin and negativity for rest of the markers. Differential diagnosis includes wide range of both benign and malignant lesions composed of atleast epitheloid cells like synovial sarcoma, fibromatososis, Malignant peripheral nerve sheath tumor. Monophasic fibrous synovial sarcoma consists of irregularly arranged fascicles of ovoid appearing cells. Lesional tissue usually shows prominent branching (staghorn) vasculature, distinct focal calcification, pseudo-osteiod or gland like spaces. The individual cell usually showed eosinophilic cytoplasm and immunohistochemically (IHC) expressed atleast one epithelial marker like cytokeratin (Tupkari *et al*., 2014 and Weiss *et al*., 2008) which is not found in fibrosarcoma.

Aggressive fibromatosis a non neoplastic spindle cell proliferation of childhood which is locally highly aggressive but no metastatic potential. Fibromatosis occurring in head and neck region is considered as extra-abdominal fibromatosis (Adamicova *et al*., 2011) which looks very similar to low grade fibrosarcoma histologically. The grade 1 FS is usually discernible from fibromatosis by the presence of occasional larger nuclei with ominous chromatin clumping, nuclear overlapping, nuclear hyperchromasia, greater cellularity, greater mitotic activity, more number of nucleoli and thin rather than thick collagen bundles (Adamicova *et al*., 2011 and Scott *et al*., 1989). IHC can be used but it may be little help in diagnosis. MPNST showed a nerve associated clinical symptoms of parasthesis with irregular arrangements of fascicles with neural invasion, necrosis which was not seen in FS. Based on all clinico- histological parameters and IHC findings, we confirmed the current case as a low grade fibrosarcoma which was originating from tongue. The treatment of choice for FS is surgical excision with wide margin. The need of adjuvant radiotherapy and/ or chemotherapy is still unclear and it is normally indicated in high grade tumors because these tumors may present subclinical or microscopic metastases at the time of diagnosis (Orhan *et al*., 2007). It is an uncommon tumor of oral cavity which may show recurrence after excision and spreads by both the local invasion and hematogenous dissemination the commonest site for metastases being lungs and bones and rarely lymph nodes. Metastases occur in 9-63% of patients and showed 5 year survival in 39-54% (Lukinmmaa *et al*., 1988; Dehne and Askin, 1976; shetty *et al*., 2010 and Pritchard *et al*., 1974). In our case, the patient underwent surgical excision of tumor with wide margin and he is under continuous follow up with no recurrence has been reported so far.

Conclusion

Fibrosarcoma arising within the head and neck region is quite rare, thus it should be included in the differential diagnosis, especial in case of soft tissue intraoral lesions arising from ventral surface of tongue. The role of routine and immunohistochemical staining pattern were thought to indicate its origin; however the histogenesis of the tumor remains controversial. Despite the fact that the incidence of fibrosarcoma has markedly decreased in recent years, there have been renewed efforts to identify the wide spreading nature of this tumor has a strong hold in determining the prognosis. The effects of environmental carcinogens are still unclear. Early detection of this aggressive tumor may help reduce morbidity.

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