Malignant Fibrous Histiocytoma—Case Report

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Abstract Fibrous histiocytoma is a tumor composed of a mixture of fibroblastic and histiocytic cells. These lesions most often arise on the skin, but may rarely occur in soft deep tissues. The diagnosis of FH may be clinically difficult when the lesion is located in the deep tissues, and is frequently confirmed after local excision. The most important diagnostic necessity is the separation of this tumor from aggressive forms of fibrohistiocytic neoplasms. We present a unique case of malignant fibrous histiocytoma developing in left buccal mucosa very near to the surgical margin from which a benign fibrous histiocytoma of the mandible was excised 3 years earlier in a 31 year old female. Clinical and histopathological evidence were not sufficient to establish link between these two processes. This article aims to describe the case and discuss the findings.

Keywords Benign fibrous histiocytoma · Malignant fibrous histiocytoma · Malignant change

Introduction

Fibrous Histiocytoma is the most common neoplasm affecting the epidermis [1]. It is a non-epithelial lesional lesion exhibiting fibroblastic and histiocytic differentiation. It has a benign and malignant course. This tumor has been associated with a previous trauma, sun exposure, and chronic infection, rather suggesting that it represents a reactive proliferation of benign cells [2]. The Malignant

Fibrous Histiocytoma (MFH) is a high-grade and aggressive sarcoma, delineated by O'Brien and Stout in 1964 and it has been recognized as the most common soft tissue sarcoma of late adult life [3]. The majority of tumors occur in one of the extremities and MFH rarely occurs in the head and neck region [3, 4].

Soft tissue tumors in the head and neck region rarely display borderline pathological features regarding benign or malignant behavior. Despite similar histological patterns the clinical outcome is often different and difficult to predict. Changing of the histological picture during progression of the FH and transformation of borderline-BFH to MFH has been described in literature in other parts and in bones in head and neck [1]. The aim of the article is to present a case of intra-oral recurrent FH with malignant changes and review the literature with immunohistochemical prediction of the behavior of the such lesion.

Case History

A 31 year old woman was referred to our center because of a fast-growing, dark-brownish, indurated nodule along the left buccal mucosa sized 3 cm in diameter, which involved the deeper soft tissue layers. (Fig. 1) A year before, a benign non-cutaneous BFH had been removed from the same location by excision biopsy (Fig. 2a–d), which had been a painless, slow-growing, brownish lump not exceeding 1 cm in diameter clinically. Histopathologically a dense proliferation of spindle-shaped cells with oval nuclei and conspicuous collagenous stroma tumor cells were arranged in bundles forming storiform patterns. (Fig. 3a) Among the lesional cells, larger polyhedral cells with eosinophilic cytoplasm were scattered singly or in aggregates. Ultrastructurally, the tumor consisted of

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Fig. 1 Frontal view of patient

Fig. 2 Lateral view of the patient

elongated, fibroblastic cells and irregular-shaped, histiocytic cells. (Fig. 3b) A diagnosis of benign fibrous histiocytoma (BFH) was made earlier. Surgical margins had been disease-free. She was not given radiation or chemotherapy after the previous surgical resection. Her excisional biopsy revealed a polypoidal lesion made up of spindle and polyhedral cell with vesicular nuclei and arranged in storiform pattern particularly around blood vessels with perivascular hyalinization. The patient currently did not report

of any other medical abnormality. No history of weight loss and on general examination the vital signs were normal without specific lymphadenopathy. Extra-orally, a swelling was observed that extended from the lower border of the zygomatic arch up to the lower border of the mandible superior-inferiorly and medially from the nasolabial fold, involving the entire cheek region in the left side. There was no proptosis or any orbital abnormality. On palpation the swelling was warm. It was soft, not fluctuant and tender, no pulsations could be felt. The margins of the mass were well defined. There was no other associated abnormality detected. Scar from previous excision could be seen on the right buccal mucosa. All teeth were including 3rd molars were present. There was no significant dental history. Routine blood and urine analysis was done and was well within normal limits. Blood analysis revealed elevation in the serum alkaline phosphatase level. Orthopantomogram did not reveal any bony involvement in the cystic mass. Contrast computed tomography and 3D CT revealed well defined enhancing hyper vascular mass of size $27 \times 25 \times 25$ mm in subcutaneous plane of left cheek closely adherent to fibers of zygomaticus major muscle. There was displacement of buccal pad of fat. Masseter and buccinator muscles were normal. Stenson's duct was normal. Evidence of erosion was observed in the maxillary bone underlying the cystic mass. (Figs. 4, 5, 6, 7, 8, 9) The cystic mass was in very close proximity to the dermis. A provisional diagnosis of a recurrent benign histiocytoma was arrived and a wide resection of the tumors with adequate margins of normal surrounding tissues was done and sent for histopathological examination. The biopsy report suggested the lesion as a malignant histiocytoma revealing an infiltrative, well-vascularized mesenchymal neoplasm containing pleomorphic tumour cells, increased mitotic count with some atypical mitoses, and myxoid stroma rich in collagen fibers (Fig. 10). The lesional cells were strongly positive for vimentin, CD34 and CD68 while cytokeratin, CD31 and Desmin were definitively negative.

Fig. 3 a (on left) Shows high power view of benign fibrous histiocytoma. Note the spindle and polyhedral cell with vesicular nuclei and arranged in storiform pattern particularly around blood vessels with perivascular hyalinization
(b right)

Fig. 4 The multi sliced image of the hypervascular mass

Fig. 5 The multi sliced image of the hypervascular mass

Fig. 6 3D CT showing the cystic mass on the left maxillary bone below the zygomatic arch. No bony erosion exposed

Few isolated areas of S100 positivity were observed. With these findings a diagnosis of Myxoid variant of malignant FH was arrived.

Fig. 7 Hypertrophied facial artery with intact peri vascular fat plane in bucco—massetric space. There were few arterial twigs from facial artery major feeders for the mass

Fig. 8 Surgical resection of the tumor

Fig. 9 Specimen after resection

Clinical staging investigations of the current malignancy (chest X-ray, MRI-scan of the neck and skull, abdominal and neck ultrasound) showed superficial infiltration of the

Fig. 10 A well-vascularized mesenchymal neoplasm containing pleomorphic tumour cells (a), increased mitotic count with some atypical mitoses (b, c), and myxoid stroma rich in collagen fibers (d)

cutis and subcutis. Neither infiltration of the Masseter nor metastases in the regional lymph nodes or elsewhere were found. No immediate postoperative radio- or chemotherapy was applied. Patient was referred to an oncology team for further management.

Discussion

Intraosseous BFH followed by MFH in the adjacent soft tissue is extremely rare. To the best of my knowledge, this is the first such case ever to be reported, though occurrence in association with bone is in literature [5]. In the present case, BFH of the cheek was originally diagnosed. BFH has been reported to possess a locally aggressive behavior and has been accepted to be not one of diagnostic exclusion under similar condition [5]. However, no information has been given on individual cases affecting jaw bones since Cale et al. [5] reviewed previous reports diagnosed as fibrohistiocytic lesions, although a few cases were included

in review series [5]. The differential diagnosis of BFH is broad and includes other intraosseous benign lesions such as metaphyseal fibrous defect, nonossifying fibroma, giant cell lesions, eosinophilic granuloma, aneurysmal bone cyst, desmoplastic fibroma, neurofibroma and myofibroma. Malignant tumors such as MFH and fibrosarcoma are also to be considered [5]. In the present case, the initial lesion occurred in an middle aged female adult, and had a slow-growing mass with tenderness showing a well-delineated margin. Those clinical features were common to those cases of BFH arising in the soft tissue, and excluded some benign lesions likely occurring in childhood [5]. Histologically, the lesion demonstrated the typical features of BFH without any characteristics that suggested malignancy, and microscopic features showing high cellular lesion predominantly consisting of spindle-shaped cells arranged in fascicular or storiform patterns identical to those involving oral soft tissues. In addition, recent studies reported CD 68 should be of value as a definitive diagnostic marker of BFH combined with conventional examination [1, 3, 5]. Other immunomarkers used could also separate BFH from other possibilities except for MFH [1, 5]. Thus, the diagnosis of BFH was not difficult to make in our case although it has been challenging and might be predicated on eliminating other possibilities.

MFH appeared in the cheek adjacent to the surgical margin from which BFH had been initially excised a year ago. The relatively early development of MFH and the limited ability of immunohistochemical analysis to differentiate between BFH and MFH [5] raise the question as to whether MFH was a second primary tumor or had existed focally from the outset. The fact that meticulous examination of the widely excised specimen did not show any focal clues for the features of the previously diagnosed BFH as opposed to the impression that MFH might have been overlooked in the sampling at the original diagnosis. However such a possibility cannot be ruled out.

In addition, since MFH is known to be one ends of continuation of fibrous histiocytoma [5]. It might be possible that recurrent BFH underwent transformation to MFH although this phenomenon has not been reported earlier.

The development of MFH following radiation therapy has been reported in the oral and maxillofacial regions [5]. However, in our case no radiation treatment was given after the BFH of the mandible was excised. There have been reports of MFH occurring in previous surgical sites or in sites involved with chronic repair processes [5]. In our patient, no other major or minor surgeries were performed in the buccal mucosa before the development of BFH or MFH. These factors indicate that this case provides evidence that a recurrent BFH could turn malignant and present as a MFH.

Conflict of interest None.

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