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Aneurysmal benign fibrous histiocytoma occurring at injection site: A case report and review of literature

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Abstract

Aneurysmal benign fibrous histiocytoma is a very rare variant of a common skin neoplasm. We present the first reported case of Aneurysmal benign fibrous histiocytoma in a Nigerian female occurring at an injection site. The tumour was populated by benign plump spindle cells with numerous blood-filled spaces containing foamy histiocytes and altered blood, prominent haemosider in deposition and a few multinucleate giant cells. The tumour cells were strongly positive for factor XIII a but completely negative for SMA, S100 and CD34.

Keywords

benign; histiocytoma; aneurysmal; factor XIIIa; nigeria

Introduction

Aneurysmal benign fibrous histiocytoma is a very rare variant of a cutaneous benign fibrous histiocytoma that is characterised by extensive cystic haemorrhage [1]. This intralesionalhaemorrhage often results in rapid growth and a blue-black appearance clinically as well as cavernous blood-filled cavities lacking endothelial lining histologically. These unique features often lead to confusion with a cyst, melanoma or hemangioma clinically [1-3]. Most reported cases have not been associated with trauma [4]. This report describes the first case occurring at an injection site as well as the first described in a Nigerian patient.

Case Presentation

A 44yrs old woman presented with a 20 year history of progressively expanding painless right gluteal mass with darkening of the overlying skin at the site of chloroquine injection for malaria. There was no associated itching, differential warmth or ulceration. It had been managed prior to presentation with topical hydrocortisone at various hospitals with no improvement. Previous biopsy done in a private hospital was reported as chronic non-specific inflammation. There was no family history of similar lesion. Physical examination revealed a tender right gluteal mass with fairly regular margins measuring 4x4cm. It was soft to firm in consistency. There was overlying skin hyper pigmentation but no ulceration. There was no regional lymphadenopathy or neurovascular compromise. The viral markers (HIV and HbsAg) were negative.

The specimen was submitted in 10% neutral buffered formalin as multiple fragments of greyish-

white tissue aggregating to 2.0 x 2.0 x 1.0cm. On histology, an apparently circumscribed lesion was seen with very scanty surrounding fibrous connective tissue containing deep dermal skin adnexal structures. The highly cellular lesion was extremely vascular and formed by uniform plump spindle cells with no significant pleomorphism, atypia or mitotic activity. Besides the blood vessels, numerous spaces without endothelial lining that contained foamy histiocytes and altered blood were also seen. There was extensive haemosiderin pigment deposition in areas as well as scattered relatively few multinucleate giant cells. Immunohistochemical analysis showed strong uniform expression of factor XIIIa and complete negativity with CD34, SMA and S100. A diagnosis of aneurysmal/hemosideroticbenign fibrous histiocytoma was made with a comment about its propensity for recurrence when incompletely excised. The patient is currently being followed up.

Discussion

Aneurysmal benign fibrous histiocytoma (ABFH) is a very rare variant of the very common dermatofibroma accounting for only about 1.7% of all cases [2]. The aetiopathogenesis of this tumour is unknown and there is no proven association of its haemorrhagic features with trauma [4,5]. It has been suggested by some workers that blood extravasating from the capillaries of the tumour form cavernous or cleft-like areas that characterise this lesion [6]. Most case reports show no association with trauma and the haemorrhage most often happens spontaneously [4,5,7-9]. The case in this report was located at an injections site which would suggest that at least a proportion of cases are associated with trauma.

Clinically, ABFH has been reported over a wide age range; however it tends to occur in young to middle-aged adults with a slight predilection for females [2,3,9]. They occur as a solitary nodule in a wide variety of locations though the lowest extremity is the commonest site of involvement . They often range in size from 0.5 to 4cm but are usually larger than the ordinary cutaneous BFH. Unlike cutaneous BFH, they can be characterized by rapid growth in size or sudden change in pigmentation which has been attributed to intralesional haemorrhage [2,3]. This often leads to confusion with a variety of other lesions. Indeed ABFH has been confused with a cyst, melanocytic neoplasm, hamengioma etc. They also have been reported to have more unusual presentations such as resembling a skin tag and a few proportion of cases have been reported with systemic symptoms [9]. The case presented here occurred in the lower extremity of a middle aged woman however there was no rapid growth or sudden change in pigmentation.

Histologically, ABFH is characterised by blood-filled spaces lined by discohesive tumour cells rather than endothelial cells [1,2,3]. The background is that of a conventional fibrous histiocytoma with prominent haemosiderin deposition [3]. Storiform architecture if present is usually focal and confined to the periphery of the lesion where the secondary haemorrhage changes are usually absent [2]. In addition to the blood filled spaces, abundant small vessels are seen in the surrounding fibrous stroma. Numerous siderophages, as confirmed by the Prussian blue perls stain were identified. Additionally giant cells are often seen in addition to lymphocytes, and occasionally few plasma cells and eosinophils. Cytologicatypia is usually absent or mild at best. There is also often moderate mitotic activity in the vicinity of the haemorrhage and some degree of hyperplasia of the overlying epidermis [1-3]. Abnormal mitoses are not seen .Though these tumour are very rare and therefore unfamiliar to the practicing pathologist, they tend to appear remarkably similar and conform to the above description as was seen in the index case [2].

Immunohistochemistry plays a limited role in the diagnosis of ABFH. In the study by Calonje et al diffuse positivity for vimentin was the only consistent staining. A few cases showed focal positivity for SMA and none showed positivity for factor XIIIa, the diagnostic marker for conventional BFH [2]. In another report the tumour cells in ABFH showed a transition in factor XIIIa positivity to negativity adjacent to the blood filled spaces [10]. In the index case, there was diffuse strong positivity for factor XIIIa. This shows the variability of expression of ABFH for factor XIIIa expression.

The histologic differentials for ABFH include angiomatoid malignant fibrous histiocytoma, spindle cell hemangioma, nodular kaposi sarcoma, angiosarcoma and spindle cell melanoma [2,3,7]. Angiomatoid MFH shares the presence of blood-filled spaces with ABFH. However angiomatoid MFH has a prominent lymphohistiocytic infiltrate amidst a uniform population of rounded eosinophilic cells that show desmin positivity [2]. KS, unlike ABFH, is composed of CD34 positive spindle cells lining slit like spaces containing red blood cells [2,7]. Cutaneous angiosarcoma occurs almost exclusively in the face and scalp and has proliferating true vascular spaces lined by atypical endothelial cells dissectingthrough the stromal collagen. Spindle cell hemangioendothelioma has true vascular spaces with papillary intraluminal structures as well as solid spindle cell areas. Inflammatory cells are absent or scanty [2,3].

The treatment for ABFH is surgical excision with clear margins. Twenty percent of cases are associated with recurrence. This is in contrast to 2% for conventional dermatofibroma. It is thought that this is due to a tendency for incomplete resection of these tumours which are more often larger than the conventional dermatofibroma rather than an intrinsic biological property of the tumour [2]. All the recurrent cases reported by Calonje et al had been incompletely excised.

Conclusion

Aneurysmal benign fibrous histiocytoma is a very rare variant of dermatofibroma. This is the first reported case in a Nigerian patient as well as the first reported to occur at the site of injection. Due to the higher recurrence rate than conventional dermatofibroma, excision with clear margins is essential.

Figure

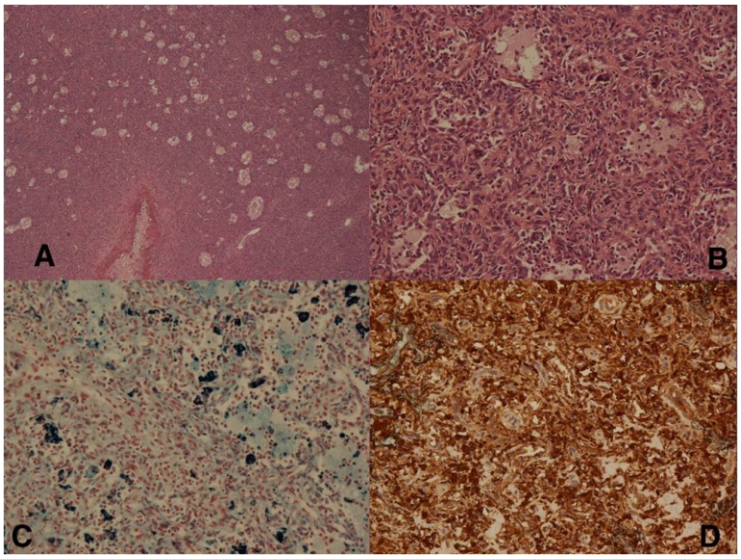

Figure 1: A. Photomicrograph showing the H&E x 4.
B. Photomicrograph showing the H&E x 20
C. Photomicrograph showing positivity of Perlsprussian blue stain x 20 Objective
D. Photomicrograph showing positive expression of Factor XIII x 20 Objective

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