Case Report

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Inflammatory pseudotumor of the skin mimicking non-melanoma skin cancer

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ABSTRACT

Inflammatory pseudo tumor (IPT) also known as inflammatory myofibroblastic tumor (IMT) or plasma cell granuloma (PCG) is mostly reported in viscera, with very rare primary cutaneous involvement. Its clinical presentation mimics non-melanoma skin cancers. This neoplasm, though considered benign, actually has intermediate biologic potential. We report a case of 52-year-old female who presented with a nodular lesion with central scab on face and hard sub mental lymph node. As patient refused for biopsy, FNAC was done, which showed highly atypical cells suggestive of squamous cell carcinoma from nodular lesion and reactive hyperplasia from lymph node. Patient underwent excision biopsy and supra-omohyoid lymph node dissection. Biopsy report showed it as inflammatory pseudo tumor of the skin and cervical lymph nodes showed non-caseating granuloma. We present review of this entity.

Keywords: IPT, Non-melanoma, PCG, Skin cancer

INTRODUCTION

In 1939 Brunn described first IPT in the lung. Umiker et al in 1954 named it inflammatory pseudotumor, because of its propensity to clinically and radiologically mimic a malignant process. Alternative terminology used in literature is IMT, Plasma cell granuloma, pseudosarcomatous myofibrobroblastic proliferation, inflammatory fibrosarcoma, post-operative spindle cell lesion of the genitourinary tract. These lesions are characterized morphologically by a mixture of myofibroblasts, fibroblasts, lymphocytes, and plasma cells. IPTs have been reported to occur mainly in children and young adults.

IPT occurs in various organs and among them, the lung is the most frequently affected organ followed by the liver, lymph node and spleen.⁵ So far only 17 cases of cutaneous IMT have been reported in the literature.⁶

CASE REPORT



Figure 1: Excised lesion with approximate 1 cm margin.

A 52-year female presented to surgical OPD with complaints of swelling in the left cheek for one and half years associated with swelling in the upper neck for 1 month. On physical examination, left cheek swelling was a nodular lesion with central scab of size 1 x 1 cms along with a palpable hard sub mental lymph node of size approximately 1.5 x 1 cms. As patient refused biopsy from the lesion, FNAC was done. FNAC from the cheek swelling revealed highly atypical cells suggestive of squamous cell carcinoma and reactive hyperplasia from the sub mental lymph node. Hematological and kidney function tests were within normal limits. Metastatic workup was done; X-ray chest and ultrasound whole abdomen were normal. Patient underwent wide local excision with modified neck dissection and the defect was closed with a rotation flap. Per-operatively there was a 1.5 x 1.5 cms firm mass infiltrating superficial fibers of buccinators muscle and multiple neck nodes IA, IB, IIA, IIB, III, IV were dissected. The histopathologically, specimen had skin with a localized, circumscribed lesion in the sub epithelial region, the epidermis is flattened and thinned out. Marked proliferation of fibrous tissue with few large atypical cells having large vesicular nucleus and prominent nucleoli, interspersed in between are aggregates of mature plasma cells and the periphery of the lesion shows many lymphoid follicles with germinal centers surrounded by aggregates of plasma cells. Immunohistochemistry revealed specimen to be weakly ALK positive. Postoperatively, patient had no complaints. Patient is in a follow up for last 3 years and there are no signs of any recurrence so far.



Figure 2: Reconstruction of face with neck dissection.

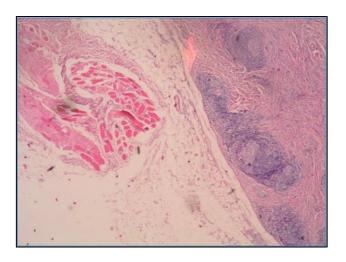


Figure 3: H and E stained 4X microphotograph of the circumscribed lesion in the sub epithelial region.

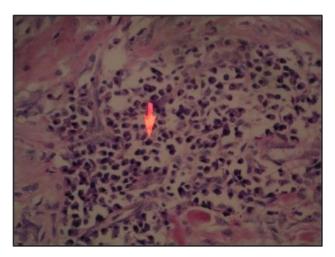


Figure 4: H and E stain, 40X microphotograph marked proliferation of the fibrous tissues with large atypical cells and periphery of lesion shows many lymphoid.

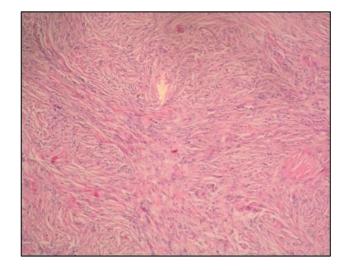


Figure 5: H and E stain, microphotograph with marked proliferation of fibrous tissue.

DISCUSSION

IMT's have a predilection for children and adolescents, although they may arise as late as eighth decade of life.⁷ They are found most commonly in the lungs, but many other sites of visceral or soft tissue involvement have been described mainly as single case report or as small series.⁵ When it presents as a cutaneous lesion, it may be present over face, neck, upper and lower extremities. Morphologically it presents as a well circumscribed nodule which may closely mimic non melanoma skin cell cancer or cutaneous malignant lymphoma.⁸ There is still controversy regarding the pathogenesis, that is, whether it is a reactive process due to some stimulus or it is a true neoplasm.⁹ Due to propensity of inflammatory pseudotumors to be locally aggressive, to frequently be multifocal, and to progress occasionally to a true malignant tumor, some authors believe this tumor is a low-grade fibrosarcoma with inflammatory (lymphomatous) cells. 10 It has been associated with infections caused by organisms mycobacterium aviumintracellulare complex, Corynebacterium Escherichia coli, Klebsiella, Bacillus sphaericus, Pseudomonas, Helicobacter pylori, Coxiella burnetti, Epstein-Barr virus, Actinomycetes and nocardiae. Some authors view it as a result of inflammation following minor trauma or surgery or to be associated with other malignancy.11 Grossly, IMTs may be firm, fleshy, or gelatinous, with a white or tan cut surface. Calcification, haemorrhage and necrosis are identified in a minority of cases. Histologically, IMTs are characterized by a variably cellular spindle cell proliferation in a myxoid to collagenous stroma with a prominent inflammatory infiltrate composed primarily of plasma cells and lymphocytes, with occasional admixed eosinophils and neutrophils.¹² By immunohistochemistry, approximately 50% of IMTs are positive for ALK (Anaplastic Lymphoma kinase), with reactivity in several large series ranging from 36% to 71%.13

Most other myofibroblastic and fibroblastic tumours, including desmoid fibromatosis, nodular fasciitis, calcifying fibrous tumour, myofibromatosis, and infantile fibrosarcoma, are negative for ALK.14 IMTs are classified as tumours of intermediate biological potential by the most recent WHO classification, due to a tendency for local recurrence and a small risk of distant metastasis.¹⁵ Recurrence rate of up to 25% have been reported for extra pulmonarytumors.³ While <2% for tumours confined to the lung.¹⁶ Rare cases of distant metastases and spontaneous remissions have been described.¹⁷ No correlation has been found between tumour size, cellularity, mitotic activity and the presence of necrosis for recurrence.¹⁸ Recently studies have reported that recurrence is very infrequent following complete excision of a solitary lesion.¹⁹ Main stay of therapy usually includes surgical excision as most cutaneous IPT can be completely resected, so other more aggressive treatments are not necessary.20

CONCLUSION

Inflammatory pseudotumors can both radiologically and clinically mimic a malignant process. There is still controversy regarding the pathogenesis, that is, whether it is a reactive process due to some stimulus or it is a true neoplasm. Complete surgical excision seems to be the best approach to management of primary cutaneous IPT.

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