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Case Report

Plasma Cell Granuloma of the Clavicle- A Rare Case Presentation

Akhtar K*, Kamal M , Alam S and Sherwani RK

Department of Pathology, Aligarh Muslim University, India

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2. Keywords

Plasma cell granuloma; Clavicle; Histopathology

1. Abstract

Plasma cell granuloma (PCG), also known as inflammatory pseudotumor, is a rare non neoplastic lesion. Although the etiology of the lesion is unclear, an altered antigen antibodyreaction have been implicated to play a role. Some authors consider PCG to be a result of inflammation following minor trauma or surgery. Others have reported cases of PCG associated with drug (amlodipine and cyclosporine) intake. Thus the etiology seems to be multifactorial. The lesion shows no sex predilection and may occur at any age. PCG is usually asymptomatic but can become symptomatic secondary to its size and location. Here we present a rare case of plasma cell granuloma of the clavicle. To the best of our knowledge plasma cell granuloma of the clavicle is the first case of PCG at this foci to be reported in literature till date.

3. Introduction

Plasma cell granuloma (PCG), also known as inflammatory pseudotumor, is a rare non neoplastic lesion comprised of a predominant polyclonal plasma cell infiltrate admixed with other inflammatory cells in a fibrovascular background [1]. PCG primarily occurs in lungs, but can be seen in other extrapulmonary locations as well which includes oral cavity, brain, kidney, stomach, heart and temporal bone [2-5].

In 1968, Bhaskar, Levin and Firch first reported the cases of gingival plasma cell granuloma [2]. Due to the rarity of plasma cell granuloma each new case should be reported in order to better understand its etiopathogenesis, treatment and prognosis.

Here we present a rare case of plasma cell granuloma of the clavicle. To the best of our knowledge plasma cell granuloma of the clavicle is the first case of PCG at this foci to be reported in literature till date.

4. Case Presentation

A 35-year old male presented with swelling on the medial end of right clavicle with history of difficulty in elevating right arm for last 3 months. On examination, the swelling was non tender with a smooth surface and well defined margins. It was firmin consistency and measured about 2 cm in diameter. The overlying skin had normal temperature as compared to surrounding area. There was restriction of movement of arm during abduction along with pain. X-ray showed an osteolytic lesion on medial end of right clavicle (**Figure 1**).

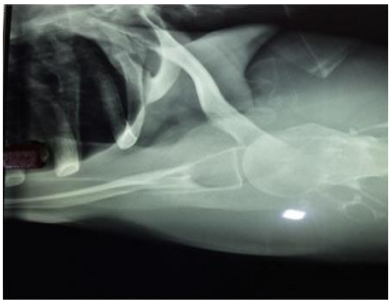

Figure 1: X-ray showed an osteolytic lesion on medial end of right clavicle.

Fine needle aspiration cytology was done from the lesion and the smears showed plenty of small plasmacytoid cells in dispersed fashion. Wide excision biopsy from anterior approach was performed and the respected specimen was sent for histopathological examination. Grossly, the mass was globular and measured 2x3cm with irregular surface and presence of bony tissue. Microscopy revealed sheets and clusters of plasmacytoid cells in sheets having eccentrically placed nuclei, coarse condensed “clock-face” chromatin pattern and abundant cytoplasmadmixed with lymphocytes and histiocytes on the background of loose myxoid and collagenized stroma showing scattered fibroblasts and myofibroblastswith foci of Russell bodies (**Figures 2,3**). Mitotic figures or nuclear atypia were not seen. Immunostaining

***Corresponding Author (s):** *Kafil Akhtar, Department of Pathology, Jawaharlal Nehru Medical College, Aligarh Muslim University, Aligarh, India, E-mail: drkafilakhtar@gmail.com*

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for kappa and lambda light chains revealed a polyclonal plasma cell population.

Other tests for systemic myeloma were performed to rule out systemic myeloma. There was absence of any evidence of anemia, hypocalcaemia, hyperuricemia or nephropathy. The patient refused subsequent radiotherapy. Serum "M" protein was 4 g/L but on follow up of 6 month it was raised to 35 g/L. The bone marrow aspiration was normal initially but at 12 month follow-up, the patient developed systemic myeloma.

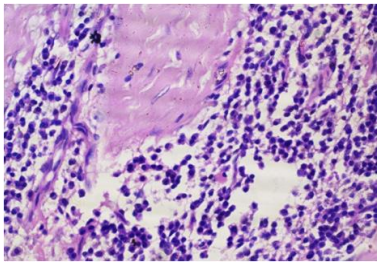

Figure 2: Microscopically tissue section shows predominance of plasma cells along with Russell bodies. Hematoxylin and Eosin ×40X.

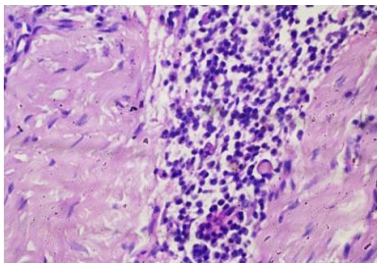

Figure 3: Microscopically tissue section shows predominance of plasma cells along with Russell bodies. Hematoxylin and Eosin ×40X.

5. Discussion

The plasma cell granuloma is a rare non-neoplastic lesion reported for the first time in the literature by Bahadori and Lie bow in 1973 [5]. The etiology of PCG/Inflammatory Pseudo Tumor (IPT) is unknown. The histologic diversity has led to conflicting opinions regarding the inflammatory or neoplastic nature of this lesion. The finding of human herpesvirus-8 DNA sequences and over expression of human interleukin [6] and cyclin D1 has been recently reported in seven cases [6]. Although the etiology of the lesion is unclear, an altered antigen antibody reaction have been implicated to play a role [7]. A number of studies have shown an association of the Epstein-Barr virus with PCG, suggesting its possible role in the etiopathogenes is of the lesion [8]. Some authors consider PCG to be a result of inflammationfollowing minor trauma or surgery [9,10]. Hence postinflammatory reactive process could be another possible etiology. There have been reported cases of PCG associated with drug (amlodipine and cyclosporine) intake [11,12]. Thus the etiology seems to be multifactorial. The lesion shows no sex predilection and may occur at any age [4]. WHO (World Health Organization) Classification of Tumours of Soft Tissue and Bone still places PCG under IMT or inflammatory pseudotumor due to the rarity of the lesion and lack of extensive research; however, PCG in the lung should not be used as a synonym for IMT as per WHO [5-8].

PCG is usually asymptomatic but can become symptomatic secondary to its size and location. Diagnostic of PCG can be a challenge as these lesions may mimic malignancy and may even destroy the surrounding tissue [13,14]. It is crucial to perform histopathologic and immunochemical evaluation of the lesion to distinguish PCG from other pathological entities like plasmacytomas or lymphomas that have a poor prognosis. Plasma cell granuloma is a rare

tumor like lesion characterized histologically by fascicles of spindle mesenchymal cells admixed withchronic inflammatory cells predominantly plasma cells. It has various components like fibroblasts, myofibroblasts, inflammatory cells (plasma cells, lymphocytes, histiocytes, mast cells and eosinophils). The stroma is collagenous and/or myxoid. All these components are arranged in varying proportions and thus create a marked histological diversity. Depending upon the predominant components, it has various nomenclatures like plasma cell granuloma, plasma cell pseudotumor, inflammatory pseudotumor, inflammatory my fibroblastic tumor, and myofibrohistiocytic proliferation [8,9].

Polyclonality of plasma cells in plasma cell granuloma is the most important factor distinguishing it from plasmacytoma [11]. Inplasmacytoma, there are diffuse sheets of neoplastic, variably differentiated, monoclonal plasma cells. Mitotic activity and amyloid deposition may be present and the inflammatory cells are very sparse [13,14]. PCG/IPT may be misinterpreted by the pathologists as nodular fasciitis, fibromatosis, fibrosarcoma or plasmacytoma. Nodular fasciitis is characterized histologically by the presence of loose myxoid matrix containing short linear curved fascicles of spindle cells. Fibromatosis usually occurs in young adults and it is characterized histologically by broad interlacing fascicles of mature fibroblasts with a variable degree of collagenisation. An inflammatory component is absent. PCG needs to be distinguished from the recently described follicular dendritic cell tumor, which runs an indolent course with a tendency of local recurrence. It can closely mimic inflammatory pseudotumor with whorls or fascicles of plump spindle cells in an inflammatory background of lymphocytes and histiocytes. In contrast, plasma cells constitute a significant proportion of the chronic inflammatory cells in inflammatory pseudotumor. The distinction can be established by the positive staining for CD21, Ber-MAC-DRC, and Ki-M4 in follicular dendritic cell tumor [8,9].

Treatment of PCG consists of complete surgical resection, although, radiotherapy and/or steroid have been tried with success in some patients where the lesion was unrespectable [15]. PCG is considered to be a benign, nonrecurring condition, however,

local aggressiveness, and recurrences may affect the prognosis of the disease [9,10].

6. Conclusion

Plasma cell granuloma is a rare non-neoplastic lesion, the exact etiopathogenesis and prognosis of which is yet to be known. The gross and microscopic similarities to other spindle cell tumors can also be misinterpreted as those of a more aggressive lesion. So awareness of PCG and its distinctive morphologic features is important in avoiding the misdiagnosis. It is essential to distinguish PCG from other poor prognostic mimikers to avoid unnecessarily aggressive treatment.

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