Malakoplakia of Proximal Tibia- A Case Report

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Abstract

Introduction: Malakoplakia is chronic granulomatous disorder resulting from an abnormal immunological response resulting in accumulation of histiocytes [1]. It normally involves the urinary bladder, colon, skin etc. But bone is rarely involved. Only seven cases have been reported in the literature. Ours is the first case involving the tibia.

Case Report: We report a rare case of malakoplakia of bone of upper end tibia in a 24 year old female with 1 month complaints of pain in the knee.

Conclusion: The importance of this case report lies in the fact that malakoplakia can mimic benign lytic lesion and we need not be over aggressive while treating these patients. These can be treated with course of antibiotics [2].

Keywords: Malakoplakia, lytic lesion, histiocytes.
Introduction

We report a rare case of malakoplakia of upper end of left tibia in a 24 year old female with 1 month complaints of pain in the knee. Malakoplakia is an abnormal immunological response resulting in accumulation of histiocytes. It normally involves the urinary bladder, skin etc. But bone is rarely involved. Only seven cases have been reported in the literature. Ours is the first case involving the upper end tibia.

Case Report

A 24 year old female presented in outdoor patient department with complaints of pain in the left knee of 1 month’s duration. Pain was insidious in onset and increased gradually over a period of 1 month. There was no swelling over the knee joint. There was no history of any significant trauma, fever, cough and night sweats. On clinical examination there was localized tenderness over the tibia just below the joint line. There was no evidence of joint effusion. Passive and active range of motion was within normal limits. There was no evidence of synovial hypertrophy. McMurray’s and Drawer test were negative.

An X-ray [Fig. 1&2] of the knee joints (AP and lateral) showed a lytic lesion in the centre of the right tibia in the epiphysis just below the joint line measuring 2*1.5 cm. It was well defined lesion with well defined border and narrow zone of transition. Other bones being normal.

MR imaging was suggestive of Brodie’s abscess. Differential diagnosis–Brodie’s abscess, tuberculosis, Giant Cell Tumor, Chondroblastoma, Langerhans Cell Histiocytosis, Metastases, Aneurysmal bone cyst, Osseous Lipoma, Osteomyelitis, Subchondral Cyst.

Fine needle aspiration cytology was inconclusive. Total leukocyte count was 9400/cmm with 60% of neutrophils and 35% of lymphocyte. ESR was 25 mm by Wintrobe’s method. Since MRI was suggestive of Brodie’s abscess, so needle biopsy or true cut biopsy was not done.

Treatment: Since aspiration cytology was inconclusive and MR was suggestive of brodie’s abscess so our treatment was to drain the abscess cavity and fill the cavity with bone graft substitutes. Intra-operatively there was thick sclerotic wall but there was no pus formation. There was soft tissue accumulation which had a yellowish tinge and cavity was found. Tissue was sent for histopathology and culture sensitivity. The cavity was filled with bone graft substitutes. Routine antibiotics were given post-operatively with Gram positive and Gram negative coverage.

Histopathology [Fig. 4&5]: Report was suggestive of malakoplakia and there was no growth on culture. Histopathology shows sheets of histiocytes and plasma cells in the bone. CD markers study showed positive for CD 68 and negative for CD 138, CD 38 and light chains (kappa and lambda). Michaelis and Gutman bodies were not seen. Our case was in stage 1 of malakoplakia also called as ‘early prediagnostic’ phase in which there is infiltration with plasma cells and macrophages with absent Michaelis and Gutman bodies.

Discussion

Malakoplakia is rare inflammatory disorder involving an abnormal immunological response to underlying infection. It occurs most commonly in the urinary tract with causative
agent being E. coli [4]. It is rarely found in the bone [5, 6&7]. Only 7 cases have been reported to date. Otherwise it is commonly seen in viscera, vagina, uterus, skin, thyroid, testes and lungs.

Malakoplakia was first described in the form of yellow mucosal plaques in the mucosa of urinary bladder in the early 1900s [1]. The word malakoplakia is derived from malakos (soft) and plakos mean plaque as the lesion was first described in urinary bladder [2].

Malakoplakia can occur at all ages and equally involve both male and female [1]. Peak incidence is in age group of greater than 50 years [4] .It is known to occur in patients who are malnourished or immunocompromised[2]. It has been found in association with tuberculosis, sarcoidosis and certain malignancies like lymphoma and carcinoma [8]. But our patient is not immunocompromised.

Malakoplakia is a spectrum of disease consisting of three stages- 1) early prediagnostic phase 2) classical phase 3) fibrosed phase [13].

Treatment modality for malakoplakia of bone is mainly a course of antibiotics for 4-6 weeks. If the disease process does not subside then it requires the removal of the material in the lytic cavities. Usually it follows a benign course and settles down with antibiotics [9]. Vitamin C has also been found to be beneficial as it lays new collagen fibrils.

Our treatment was clearing the cavity and filling it with bone substitutes. Patient is completely asymptomatic at follow up period of 18 months. There is no recurrence of any cavity or any lytic areas [Fig.4].

**Conclusion**

The importance of this case report lies in the fact that malakoplakia can mimic benign lytic lesion. Treatment of these lytic lesions depends upon the diagnosis. Aspiration cytology must be accurate to get proper diagnosis so that treatment can be given accordingly. But as far as Malakoplakia is concerned, it can be managed with course of antibiotics. We need not be over aggressive while treating these patients.

**Clinical Message**

Malakoplakia must be differentiated from other benign lytic lesions by accurate histopathology and CD markers. Histopathology is important for diagnosis and it is a spectrum of disease. Treatment is 4-6 weeks of antibiotics.

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