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Periprosthetic rice body containing cyst formation in a case of revision total hip replacement in a known RA - Diagnostic Dilemma: A case report

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Abstract
Introduction: Recurring painful mass with no signs of inflammation over the hip in a case of revision total hip replacement in known RA is a diagnostic dilemma and has not been reported when no Implant wear debris, infection, expanding hepatoma, Pseudoaneurysm, synovial cyst, sarcoma were excluded in a multiple operated Revision THR.
Case Presentation: We report a case of a large rice body containing lobulated recurring clinically non-inflammatory painful cystic swelling over the lateral aspect of a multiple operated revision total hip replacement on the right hip in a known RA patient, a 64 years female. She had both hip and knee primary replacement done in 2008 and revision right Hip following periprosthetic fracture in 2016.
Conclusion: This case report helps in a systematic approach to diagnose and manage a case of large recurring painful rice body containing cystic swelling of the hip following multiple operated Revision THR.

Keywords: Periprosthetic, diagnostic dilemma, rheumatoid arthritis, recurring painful

Introduction
Rice body formation is an uncommon phenomenon first described by Riese *et al.* 1895 as an intra-articular proteinaceous mass seen in tuberculosis [1]. Since then there are various reports of rice body formation in inflammatory arthritis, chronic bursitis, post traumatic conditions, tendon sheaths and in peri-implant tissues. The composition of rice bodies vary from compact fibrin and mononuclear cells to collagen fibers, reticulin, and elastin. They vary in shape and size. Their occurrence in RA is well established [2]. Rice body formation following THR has been reported [3]. However, there are no reports of rice body containing cyst following revision THR. We report here the case of a painful multi-lobulated rice body containing cyst in a 64 years known case of Rheumatoid arthritis female patient with right sided Revision THR.

Case Report
A 64 year female, with known Rheumatoid Arthritis, on DMARDs since 2006, had sequential un-cemented total hip replacement of both hips in March 2008 with metal on polyethylene bearings. She had bilateral knee replacement in one sitting in June 2008 and was independently mobile since then. In 2016, she had a fall and sustained a peri prosthetic fracture on her right femur for which she had revision of the femoral stem prosthesis with a long Wagner stem with bone build up using free fibular graft. She was apparently alright subsequently and was on DMARDs with low dose steroids. In March 2020 she presented with a painful swelling on the lateral aspect of the right upper thigh. On examination, there was a large fluctuating mass, extending from 7 cm above the trochanter tip to middle of the thigh over the lateral surface of the hip with no signs of inflammation on examination. Hip range of motion was normal and painless. Active straight leg raising was possible and painless.
X-rays revealed a stable implant with no osteolysis. A large soft tissue shadow was seen all around the femur. Her blood counts were within normal limits but her ESR was raised to 65 mm 1st hour. Anti-CCP and RA factor were highly raised.

On aspiration of the cyst, 1500 ml of brownish turbid fluid was obtained and was sent for analysis. Culture for bacterial and tuberculosis growth were negative. GeneXpert TB was negative. In cytology, total cell counts were 8000/cc with predominance of Neutrophils (80%) with few RBCs. Biochemical analysis showed fluid glucose of 109 mg/dl, protein level of 6.02 gm/dl, fluid lactate dehydrogenase of 1847 with alkaline pH. She was put on anti-inflammatory drugs and short term antibiotics (Amoxicillin and Clavulanic acid). Her pain reduced and swelling size decreased.

However, she presented with similar swelling by the end of May 2020. Repeat aspiration was done and similar brown turbid fluid of 800ml was obtained which was again sent for analysis, which showed almost similar parameters. However, this time her real time PCR was positive for Kochs. She was put on anti-tubercular drugs and was asked to follow-up.

She presented again with similar painful swelling in three months. Repeat aspiration revealed a similar picture and her LFT was deranged with a high APTT range and low haemoglobin. MRI scan films were obtained which revealed a large multi-lobulated cystic swelling extending from the hip down to the middle third of the thigh encircling the femur having thousands of large T2 hypo intense granules of size 6 to 7 mm resembling rice grain granules.

After stabilizing the blood parameters, the patient was taken for surgical excision of the cyst. Surgical exploration identified a well circumscribed cyst overlying and extending into the hip joint containing thousands of highly organized grayish-white irregular mass of size 0.5cm to 1.5 cm bathed in turbid fluid. The contents of the cyst were emptied and capsular lining was traced all around the femur and into the hip joint and comely excised. There was no evidence of prosthetic loosening, poly wear or any metal debris. However, one en-cerclage wire was loose and was removed. The fibular graft was well taken up. Thorough debridement and irrigation was done and the wound was closed in layers over suction drain. All cultures taken from the wound including aerobic, anaerobic fungal, and acid fast bacilli were negative for infection. Histopathology confirmed the diagnosis of rice body containing cyst.

Following excision, the patient had immediate pain relief and was ambulatory. Her inflammatory markers for rheumatology were highly raised and she was put on DMARDs with low dose steroids along with antibiotics including anti-tubercular drugs as per the institute protocol. At one year follow-up, the patient was comfortable, no recurrence of the swelling was seen and she was ambulatory.

Discussion

There are several well described causes of painful mass following THR. Metal and poly debris induced reaction causing cyst formation, pseudotumor, infection, heamatoma following trivial trauma are few of the examples. Rarer causes include synovial sarcoma, osteosarcoma, malignant fibrous histiocytoma have been reported following THR. Synovial cyst from the hip joint capsule or from the iliopsoas bursa following THR, although reported, is extremely rare. In some cases, vascular compression by the cyst was what prompted exploration and surgical excision of the compressive mass [4-6]. In most of these cases, pathologic examination of these synovial cysts demonstrated fluid within the cyst with synovial cells on the inner surface of the cavity. Many of the specimens contained polyethylene debris suggesting that a hypersensitivity reaction to wear particles may be responsible for cyst formation [4-6]. An allergic reaction to the cobalt-chromium molybdenum hip prosthesis was implicated in the development of neuro-muscular and local symptoms characterized by metallic debris and necrosis [7].

Conclusion

In the present case, the absence of clinical, serological and radiological findings made the early diagnosis difficult. MRI raised a strong suspicion of rice body containing cyst in view of known RA in the patient. Surgery and subsequent histopathology confirmed the diagnosis. Rice body containing cyst has been well documented in Rheumatoid arthritis as well as in tuberculosis. There are few reports of rice body containing cyst in patients having neither of these diseases and hip arthroplasty. Hence, as a result of our case report and of the ones stated previously, surgeons should be mindful of this rather uncommon diagnosis when dealing with patients of similar signs and symptoms.

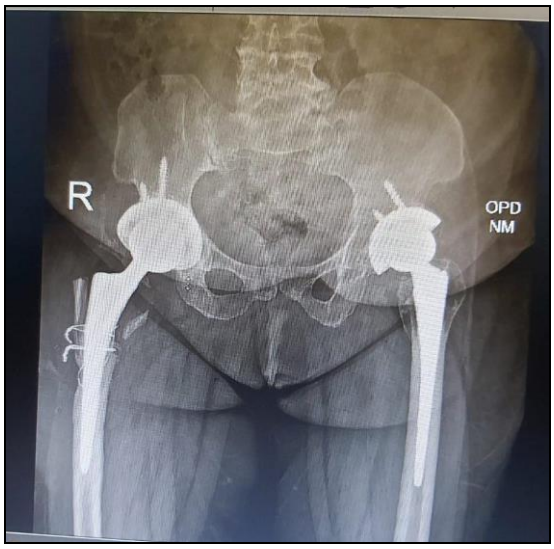

Fig 1: Digital Xray film pre-operative

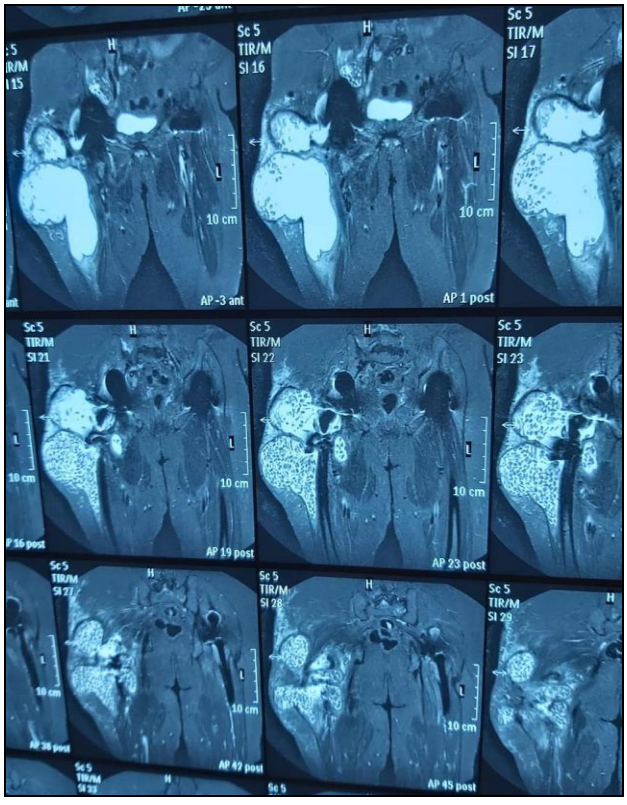

Fig 2: MRI films of pre-operative pictures

Fig 3: Clinical picture of the swelling

Fig 4: Intra-operative picture of the mass

Fig 5: Excised rice bodies with sac in-toot bathed in turbid fluid

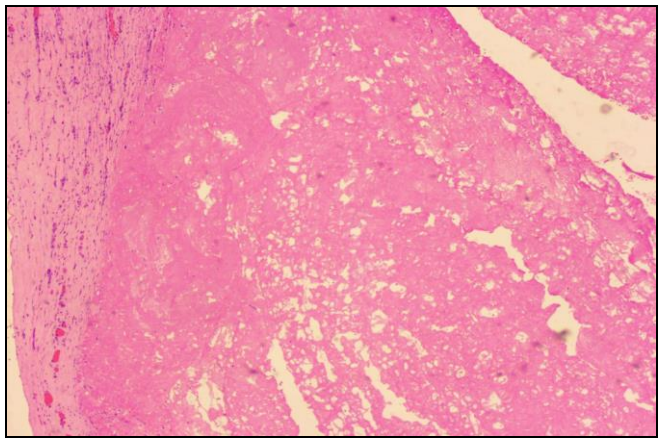

Fig 6: Histo-pathological slide of the rice bodies (Gross description - Grayish-brown to grayish-white, polypoid projections varying in size from 0.3cm to 1.2cm. Microscopic description - acute on chronic synovitis, thickened granulation tissue and large area of fibrin deposition)

Consent

The patient has given her informed consent for this case report to be published.

Conflicts of interest

The authors declare that they have no financial, or personal relationships to disclose.

Abbreviations

THR - Total Hip Replacement.
DMARDs - Disease Modifying antiRheumatic Drugs.
ESR - Erythrocyte Sedimentation Rate.
CCP - Cyclic Citrullinated Peptide.
RA - Rheumatoid Arthritis.
TB – Tuberculosis.
PCR - Polymerase Chain Reaction.
LFT - Liver function test.
APTT - Activated Partial Thromboplastin Time.
MRI - Magnetic Resonance Imaging.

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