



## Case report

Rice body formation due to *Haemophilus parainfluenza*-associated chronic arthropathyPavan Bhat<sup>a,\*</sup>, Sahiba Khurana<sup>b</sup>, Rachel Fanaroff<sup>c</sup>, Scott M. Adams<sup>d</sup>, Ronald P. Rabinowitz<sup>e</sup><sup>a</sup> Department of Medicine, Division of Infectious Diseases, University of Maryland School of Medicine, Baltimore, MD, USA<sup>b</sup> Department of Internal Medicine, MedStar Franklin Square Medical Center, Baltimore, MD, USA<sup>c</sup> Department of Pathology, University of Maryland Medical Center, Baltimore, MD, USA<sup>d</sup> Orthopaedic Associates of Central Maryland, Catonsville, MD, USA<sup>e</sup> Department of Medicine, Division of Infectious Diseases, University of Maryland School of Medicine, The R. Adams Cowley Shock Trauma Center, Section of Infectious Diseases, Baltimore, MD, USA

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## ABSTRACT

A 68-year-old woman with a medical history significant for psoriatic arthritis was found to have an enlarged, painful lump on her left hip 15 months after intramedullary rod placement for a left subtrochanteric femur fracture sustained in a fall. Histopathological findings showed rice body formation (RBF) with concurrent *H. parainfluenza*. RBF is a relatively rare arthropathy of a subset of chronic inflammatory disease such as rheumatoid arthritis or tuberculous arthropathy. RBF associated with psoriatic arthritis or orthopedic hardware placement has been reported in a handful of cases in the literature but there has not been any definitive evidence for RBF as a result of *Haemophilus parainfluenza* infections and is a rather unusual characteristic of this case.

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## Introduction

Rice body formation (RBF) is an infrequent sequela of chronic, inflammatory arthropathy associated with rheumatoid arthritis (RA) or tubercular arthropathy [1,2]. This inflammatory abnormality of fibrinous deposits can be debilitating and limit musculoskeletal motion [2,3]. To our knowledge, non-tubercular or RA-related RBF is rare and has not been reported in association with *Haemophilus parainfluenza* infection. In this case, we describe clinical, radiological, and histopathological findings on a 68-year-old woman with psoriatic arthritis who developed rice-body disease secondary to chronic inflammatory arthropathy associated with post-fracture orthopedic hardware placement and *Haemophilus parainfluenza* infection.

## Case report

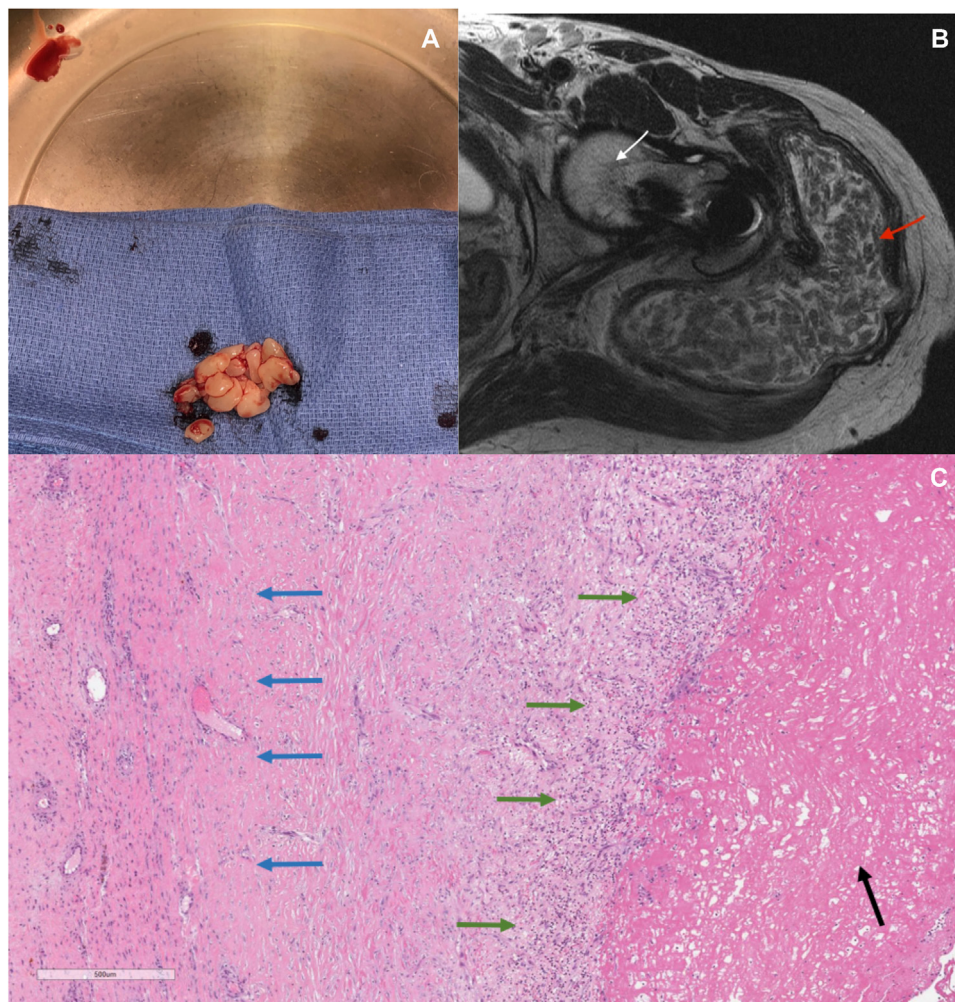
A 68-year-old woman with a medical history significant for well-controlled psoriatic arthritis on monoclonal antibody therapy (most recently adalimumab), HTN and COPD presented with an enlarged, painful mass on her left hip. She had suffered a left subtrochanteric femur fracture as a result of a fall in January 2018

and underwent intramedullary (IM) rod placement. Her postoperative course was complicated by poor wound healing and she had a bone stimulator for about 3 months. Approximately 15 months post-operatively, the patient noted the development of a large, fluctuant, nonerythematous mass over her upper left thigh that was painful and warm to touch. She denied any open wounds or drainage and systemic symptoms such as fevers or chills. She briefly lived in Israel in her early 20s but otherwise denied any other travel history. She had not been incarcerated nor exposed to any known sources of tuberculosis (TB). She was a former smoker with a 30 pack-year cigarette smoking history and drank 1–2 standard alcoholic drinks daily. She was divorced and worked as a real estate agent and denied any illicit substance use.

An outpatient magnetic resonance imaging (MRI) study revealed a large thick walled collection with innumerable debris and filling defects along the femur above the IM rod with extension to posterior thigh concerning for complex bursitis with superimposed infection (Fig. 1B). Culture from initial aspiration in August 2019 grew *Haemophilus parainfluenza* and methicillin-sensitive *Staphylococcus aureus* (MSSA) in broth only. She underwent another aspirate in September 2019 that was culture negative. No antimicrobials were prescribed in between these aspirates. Other than discomfort from the mass over the left hip, the patient remained clinically stable. Because of the continued pain and swelling, she electively underwent complete evacuation of the large complex cyst in October 2019. Intraoperatively, she was

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**Fig. 1.** A) Gross specimen illustrating smooth, polished-appearing clumps of RB; B) Axial, T2-weighted MRI demonstrating large thick-walled collection with innumerable debris and filling defects (red arrow) along the femur (white arrow) above the IM rod with extension to posterior thigh; C) Routine-stained sections of the specimen reveal fibrinous debris (black arrow), an acute/chronic inflammatory infiltrate (green arrows), and an organizing inflammatory response (blue arrows). No organisms or pigment deposition is present (Hematoxylin and eosin; 50× magnification).

found to have a subfascial, thick-walled cystic structure originating near, but not involving, the IM hardware filled with fluid and white/tan colored particulate debris. There appeared to be no bone or intramuscular involvement. Specimens were sent for pathologic evaluation and for bacterial, fungal and AFB culture and 16S genetic sequencing. Pathologic sections revealed acellular fibrinous debris and granulation admixed with neutrophilic inflammatory cells. Grocott's methenamine silver (GMS) and modified acid-fast staining were both negative. Fungal and AFB cultures, in addition to the 16S sequencing, were all negative. Aerobic bacterial culture of the abnormal fluid revealed *Haemophilus parainfluenzae*. She was eventually discharged from hospital with amoxicillin-clavulanate for two-week course with a diagnosis of infectious bursitis.

## Discussion

Rice body formation is a relatively rare entity that results in fibrinoid deposits in joints, tendons and bursa sheaths. Although it is most commonly seen in association with rheumatoid arthritis or tubercular arthritis, it can occasionally occur in the absence of any underlying systemic disorder [1,2]. On gross evaluation, rice body formations have the appearance of polished grains or clumps of rice. They generally form as a nonspecific response to chronic inflammatory arthropathy and tend to occur in synovial fluid,

particularly of the knee, bursae and, less frequently, tendon sheaths [2,3]. Although there is no clear correlation, RBF associated with psoriatic arthritis or orthopedic hardware placement has been reported in a handful of cases in the literature [4]. After a thorough literature review, we did not find any cases of RBF attributable to *H. parainfluenzae* infection and is what sets this case apart from the previous cases of this disease.

The pathogenesis of RBF is largely speculative. Researchers have suggested they may form due to cumulative sloughing of micro-infarcted synovium into the joint space which then get encased by fibrin [1,2]. Another hypothesis suggests that rice bodies form de novo independently of other synovial factors and eventually enlarge due to fibronectin and fibrin aggregate deposition [1,2]. Macroscopically, they develop clumped, loose bodies that can resemble polished grains of rice. Microscopically, they contain an inflammatory, nebulous acidophilic core surrounded by fibrin and collagen [1].

Magnetic resonance imaging (MRI) imaging is now the modality of choice for evaluating rice bodies. They exhibit hypointense signals on both T1 and T2-weighted sequences [1–4]. Differential diagnosis includes synovial osteochondromatosis and pigmented villonodular synovitis (PVNS). However, synovial chondromatosis exhibits synovial soft tissue ossification and hyperintensity on T2-weighted sequences. Furthermore, synovial

chondromatosis is primarily of hyaline cartilage as opposed to fibrinous tissue seen in RBF (Fig. 1 C) which would be readily seen in pathologic specimens to allow for differentiation of these 2 entities [1,2]. RBF can be differentiated from PVNS by the presence of hemosiderin deposits microscopically in the latter.

Treatment is largely surgical with excision and drainage as evacuation of these outgrowths reduces compressive changes and provides symptomatic relief. Surgical drainage would also allow a full workup that would help exclude any other pathologic diagnosis. Accordingly, the finding of RBF should alert clinicians to the presence of a chronic inflammatory arthropathy, infectious or non-infectious, and warrants further investigation.

In conclusion, we present a rare case of rice body formation associated with *Haemophilus parainfluenzae* infection in a patient with psoriatic arthritis after repair of a femur fracture who presented with a chronic mass and pain. She was treated with surgical debridement and oral antimicrobials and recovered

#### Author's contribution

Drs. Bhat and Khurana completed the background research, drafted and edited the manuscript. Dr. Adams provided all surgical and MRI images and reviewed manuscript. Dr. Fanaroff provided pathohistologic images and descriptions of tissue findings and

reviewed manuscript. Dr. Rabinowitz supervised and edited final manuscript.

#### Informed consent

Informed consent was obtained from the patient for educational use of the below mentioned data and no personal patient information has been disclosed.

#### Declaration of Competing Interest

None of the authors have any financial disclosures or personal bias to declare.

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