

Lumbar Facet Joint Cyst Caused by Calcium Pyrophosphate Arthropathy of the Spine: Case Report and Review of Literature

Mahmoud R Alkharsawi, MD.^{1,2}, Heinrich Boehm, MD.¹, Mootaz Shousha, MD.^{1,3}, Mohamed Alhashash, MD.^{1,3}

Zentralklinik Bad Berka, Germany¹, Orthopaedic Department, Faculty of Medicine, Tanta University, Tanta, Egypt², Orthopaedic Department, Faculty of Medicine, Alexandria University, Alexandria, Egypt³.

ABSTRACT

Background Data: Calcium pyrophosphate dihydrate (CPPD) deposition disease is a metabolic disorder caused by the deposition of CPPD crystals in the articular or periarticular structures that leads to inflammation of the joints. Spine affection is rare.

Purpose: To present a rare case of lumbar facet joint cyst caused by calcium pyrophosphate arthropathy.

Study Design: A case report with a brief review of the literature.

Patient and Methods: Seventy-three-year-old man, who underwent PLIF L5/S1 ten years ago, presented with right L4 nerve root compression symptoms that have started two months ago. Clinical examination revealed no motor function disturbance but hypoesthesia in the L4 dermatome. He has no history of crystal deposition disorder. The patient underwent decompression and fusion at this level.

Results: The symptoms were completely resolved postoperatively and after one-year follow-up. Histopathological examination of the cyst revealed fibrous granulation tissue. Examination of the tissue under polarized light showed positively birefringent, short blunt crystals of calcium pyrophosphate dihydrate.

Conclusion: Calcium pyrophosphate arthropathy, although rare, should be incorporated into the differential diagnosis of cystic lesions of the facet joints. (2020ESJ221)

Keywords: calcium pyrophosphate disease, facet joint cyst, pseudogout, lumbar spine.

Address correspondence and reprint requests: Mahmoud Alkharsawi, MD
Orthopaedic Department, Faculty of Medicine, Tanta University, Tanta, Egypt.
E-mail: m.alkharsawi@gmail.com

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INTRODUCTION

Calcium pyrophosphate dihydrate (CPPD) deposition disease is a metabolic disorder caused by the deposition of CPPD crystals in the articular or periarticular structures that leads to inflammation of the joints.⁶ The exact cause is not well-established.

The clinical picture ranges widely from asymptomatic incidental discovery to acute arthritis ending with chronic destructive arthropathy. Moreover, chronic arthritis with acute exacerbation (pain, swelling, and erythema) has been reported. Acute presentation or exacerbation may be associated with fever and high inflammatory markers.⁹ Clinical manifestations of the disease are similar to those of gout; hence, it is termed “pseudogout.”⁶

Chondrocalcinosis is a radiological hallmark of the disease. Chondrocalcinosis is characterized by radiodensities in punctate or linear forms that occur in both hyaline and fibrocartilage of the joint and periarticular structures. Examination of the synovial fluid under polarized light microscopy reveals characteristic crystals of CPPD, which are rhomboid and positively birefringent.² Spinal involvement is rare⁶ and usually an incidental finding of the aging spine. Symptomatic involvement is even rarer. Sites affected are ligamentum flavum, most commonly, followed by facet joints, and finally, the disc.⁶

We present a rare case of CPP facet cyst with symptomatic involvement of the lumbar spine.

CASE REPORT

A 73-year-old man was presented with an eight-month history of low back pain that started to radiate to the right leg six months after the onset, with typical distribution to the L4 dermatome. The pain was constant and did not resolve with position changes. He underwent posterior lumbar interbody fusion (PLIF) L5/S1 ten years ago

because of degenerative spondylolisthesis and C5-C7 fusion due to degenerative disc disease. He had no history of crystal deposition disorder (gout or pseudogout). The patient was on anticoagulative treatment due to a history of DVT in the right leg. On examination, the straight leg raising test was positive on the right side. Neurological examination revealed normal muscle tone, power, sensation, and reflexes in both lower limbs. The patient had a normal rectal tone and perianal sensation. No signs of peripheral joint affection could be detected. His inflammatory markers were slightly raised, with a C-reactive protein level of 15 mg/l (reference range, 0–4 mg/l) and erythrocyte sedimentation rate of 46 mm/h (reference range, 0–20 mm/h).

Conventional and functional X-ray views of the lumbar spine showed low L4/5 disc space height with osteophyte formation. The initial thinking was directed towards adjacent segment disease. Computerized tomography showed well-fused L5/S1 level and erosion of the adjacent posterior surface of L4, which may represent pressure atrophy. No calcification of the cyst was noted (Figure 1). Magnetic resonance imaging revealed a cyst (12 mm in diameter) attached to the medial aspect of the right L4-L5 facet joint and exerting pressure on the posterior aspect of the L4 vertebral body and then extends into the spinal canal and intervertebral foramen. MRI has also shown L4/5 disc protrusion (Figure 2).

The patient underwent decompression and fusion at this level and his symptoms were completely resolved postoperatively (Figure 3). Histopathological examination of the cyst revealed hyaline and fibrous cartilage tissues with degenerative changes. Examination of the tissue under polarized light showed positively birefringent, short blunt crystals of calcium pyrophosphate dihydrate (Figure 4). No medical treatment was initiated. The patient went through an uneventful course with complete union. Last follow-up was one year postoperatively. The patient was informed that blind data concerning his case would be submitted for publication and agreed.

Table 1. Review of previously reported cases.

Study	Age	Sex	Level affected	Side	Presentation	Peripheral joint affection	Surgical Treatment	Medical treatment	Outcome
Mahmud ⁷ 2005	70	F	L4/L5	Right	Calf pain	?	Decompression & fenestration	No	Recovered
	81	M	L5-S1	Bilateral	Claudication	?	Decompression & Gill L5 laminotomy	No	Recovered
	79	M	L4/L5	Right	Radicular Pain	?	Decompression & fusion	No	Recovered
Gadgil ⁴ 2002	67	F	L4/L5	Bilateral	Radicular pain	Yes	Decompression	No	Recovered
Fujishiro ³ 2002	71	F	L4/L5	Left	Back & radicular pain	Yes	Fine-needle aspiration	No	Recovered
Namazie ⁹ 2012	69	F	L4/L5 L5/S1	Left Left	Back & radicular pain	No	Decompression	No	Recovered
Current study 2021	73	M	L4/L5	Right	Back & radicular pain	No	Decompression & fusion	No	Recovered



Figure 1. Preoperative X-ray lateral (A) and AP (B) views showing well-fused L5/S1 level. Preoperative CT scan: sagittal reformat (C) and axial image (D) showing erosion of the adjacent posterior surface of L4, which may represent pressure atrophy. No abnormal calcification is noted.

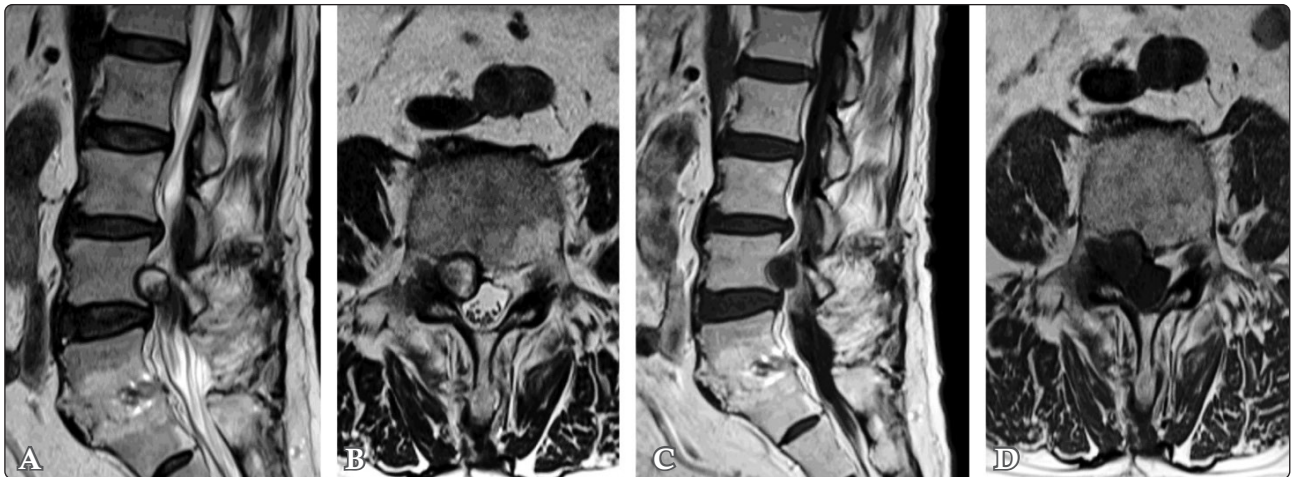


Figure 2. Preoperative sagittal (A) and axial (B) T2-weighted and sagittal (C) and axial (D) T1-weighted MRI images showing facet joint cyst (12 mm in diameter) attached to the medial aspect of the right L4-L5 facet joint and exerting pressure on the posterior aspect of L4 vertebral body and then extends to the spinal canal and intervertebral foramen.

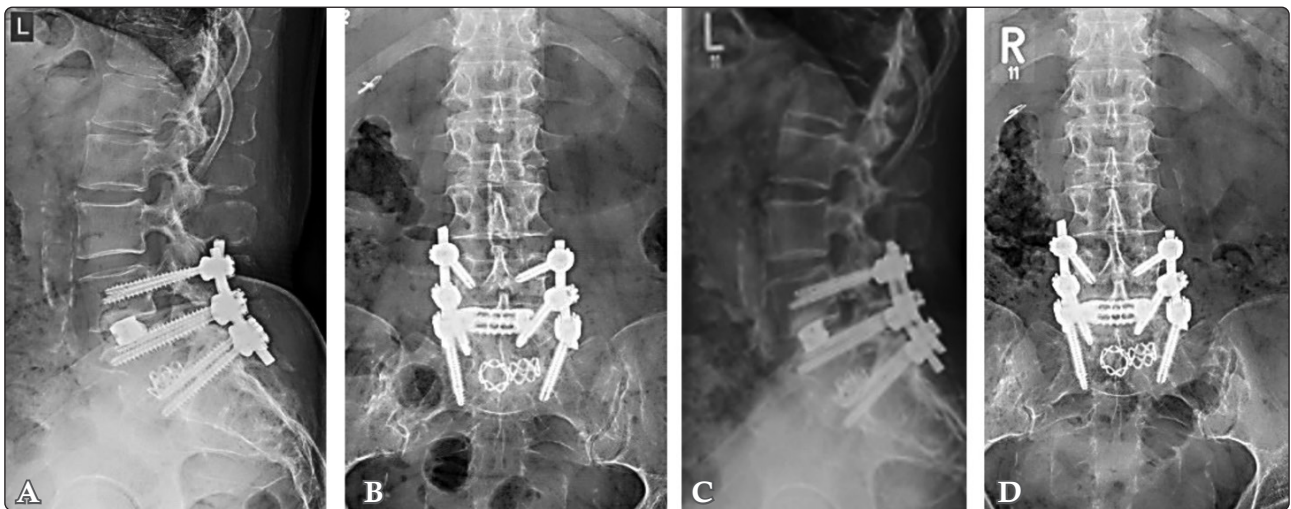


Figure 3. Postoperative X-ray (A, B) showing the interbody fusion of the L4/5 level. One-year postoperative X-ray (C, D) showing fusion of the L4/5 level, no screw loosening, and no signs of adjacent segment disease.

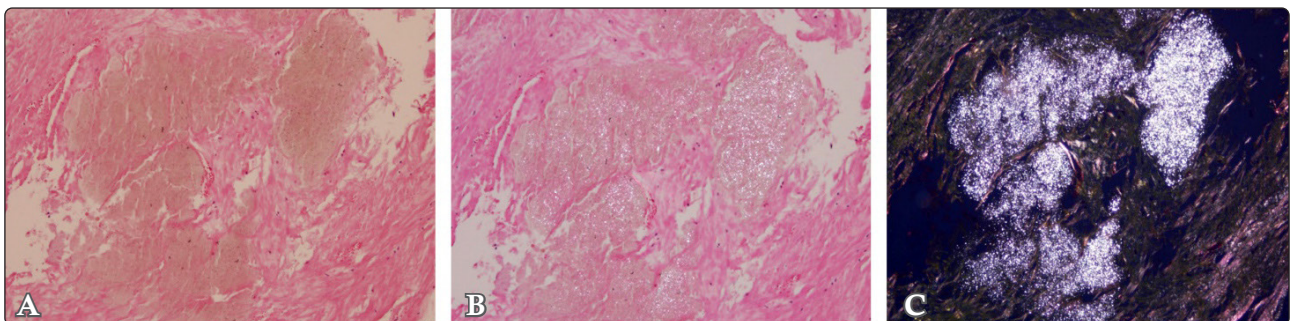


Figure 4. (A) Histopathological photo showing hyaline and fibrous cartilage tissues with degenerative changes. (B, C) Tissue examination under polarized light showed positively birefringent, short blunt crystals of calcium pyrophosphate dihydrate.

DISCUSSION

Cystic lesions of the facet joints are most attributed to degenerative osteoarthritis of the facet joints. They occur most frequently at the level of L4-L5, which is the most mobile lumbar segment, and can be associated with spondylolisthesis.¹ Synovial cysts alone can be asymptomatic regardless of the underlying pathology; however, if they attain sufficient size, they can cause compression-related symptoms such as claudication or radicular pain or cauda equina syndrome.^{5,8} Sudden onset of pain can be associated with intracystic bleeding.^{8,10} In this case, back pain was first reported by the patient, probably due to a pathological process within the facet joint. Then, radicular symptoms occurred six months after the onset of the back pain due to compression of the L4 nerve root by facet joints cysts. Histopathological examination of removed cysts is the standard in our department. We recommend this examination to reveal rare pathologies like CPPD.

This case is the seventh case with a total of 10 cysts described in the literature as facet joint cyst caused by calcium pyrophosphate arthropathy.^{7,3,10} Namazie and Fosbender⁹ provided a comprehensive review of the literature regarding this issue. The most affected level is L4/L5 (7 cysts), followed by L5/S1 (3 cysts). The main clinical presentation is pain either radicular due to nerve root compression, back pain, or claudication due to spinal canal stenosis. Open decompression was performed in six cases (7 cysts), including the case reported in this study, with fusion in only two cases (including also the case reported in this study) and fine-needle aspiration in one case. All the cases recovered postoperatively. There was no recommendation of medical treatment in all publications (Table 1).

Although the pathogenesis of CPPD disease is not fully understood, the formation of CPP crystals in the pericellular matrix of cartilage is the essential first step in the disease process. Once CPP crystals are generated, they mediate tissue

damage through multiple mechanisms. Apart from inducing inflammation, CPP crystals have important direct catabolic effects on chondrocytes and synoviocytes, eliciting the production of destructive matrix metalloproteinases and prostaglandins. CPP crystal deposits in articular cartilage also alter the mechanical properties of cartilage, which may cause or accelerate joint damage. Histopathological features of the disease include the conventional degenerative changes and the characteristic positively birefringent, short blunt crystals of calcium pyrophosphate dihydrate by tissue examination under polarized light.¹¹

The first step in treating adjacent segment disease is conservative treatment (pain medication, physiotherapy, and strengthening exercises). The patient presented underwent a trial of conservative treatment for six months before the surgery.

Finally, it is to be noted that the management of the case is the “standard” treatment of spinal canal stenosis; in other words, the discovery of CPPD has not altered the course of treatment. What can be recommended is regular follow-up to detect peripheral joint involvement early.

CONCLUSION

Calcium pyrophosphate arthropathy, although rare, should be included in the differential diagnosis of cystic lesions of the facet joints. Surgical intervention is indicated for compression-related manifestations of the cyst or associated pathology such as instability or spondylolisthesis. Regular follow-up for early detection of joint affections is recommended. So far, there are no evidence-based recommendations regarding the medical treatment of such cases.

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الملخص العربي

تكون حوصلة في المصل التنوعي في الفقرات القطنية نتيجة لمرض كرساتلات الكالسيوم بيروفوسفات: تقرير عن حالة ودراسة مرجعية

البيانات الخلفية: يعتبر مرض النقرس الكاذب أحد الأمراض الأيضية التي تؤثر على الغضاريف المبطنة للمفاصل. ينتج هذا المرض عن ترسب كرساتلات الكالسيوم بيروفوسفات دايهيدرات في غضاريف المفاصل مما يؤدي للإلتهاب المفصل. تتنوع الصور الإكلينيكية للمرض بين: الاكتشاف العرضي لوجود المرض والالتهاب الحاد للمفصل وصولاً الي التهاب مزمن المفاصل مع فقد للغضروف المبطن للمفصل. بعد تأثر مفاصل العمود الفقري نادر الحدوث.

الغرض: في هذه الورقة البحثية يتم تقديم حالة نقرس كاذب في المفصل بين التنوعين العلوي والسفلي للفقرتين القطنيتين الرابعة والخامسة على الجهة اليمنى.

تصميم الدراسة: تقرير عن حالة مرضية مع دراسة مرجعية مختصرة عن الموضوع.

المرضى والطرق: كان المريض يعاني من آلام أسفل الظهر لمدة ثمانية أشهر مع انتشار الألم في الطرف السفلي الأيمن مطابقاً لتوزيع العصب الشوكي القطني الرابع. وقد خضع المريض لعملية تثبيت للفقرتين الخامسة القطنية مع العجزة الاولى قبل عشر سنوات.

أظهرت نتائج الاشعة العادية والمقطعية مع التصوير بالرنين المغناطيسي وجود حوصلة في المفصل التنوؤي الأيمن بين الفقرتين الرابعة والخامسة.

النتائج: تم إجراء عملية توسيع للقناة العصبية مع استئصال الحوصلة ولحام للفقرتين الرابعة مع الخامسة القطنية. لم تحدث مضاعفات أثناء او بعد العملية. تحسنت أعراض المريض بعد إجراء العملية.

اثبت فحص الباثولوجي وجود كرساتلات الكالسيوم بيروفوسفات دايهيدرات. تعتبر هذه هي الحالة السابعة المسجلة علمياً. لم يكن المريض يعاني من أعراض التهاب المفاصل الطرفية الدالة على وجود نقرس كاذب.

الخلاصة: التوصية الطبية: النقرس الكاذب يجب ان يتم إدراجه في احتمالات تشخيص نشوء حوصلات المفاصل التنوئية للعمود الفقري. يوصي بالمتابعة المستمرة للمرضي من أجل تشخيص تؤثر المفاصل الطرفية بشكل مبكر.