CASE REPORT

A healthy patient with bilateral frozen hips preceding bilateral frozen shoulders: a cautionary tale

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SUMMARY

Adhesive capsulitis of the shoulder (frozen shoulder) is a common disease characterised by spontaneous onset of pain and restriction of movement, followed by 'thawing', with complete or near-complete resolution. Adhesive capsulitis of the hip has been reported in around a dozen patients. This report describes an otherwise-healthy middle-aged woman with apparent sequential resolving adhesive capsulitis of all four ball-and-socket joints over a 9-year period, initially affecting each hip and then each shoulder sequentially. The likely hip diagnosis became clear only retrospectively with development of the second frozen shoulder, 5 years after the first pain. All joints subsequently resolved within the expected timeframe and the patient remains healthy, other than having mild hypertension. This case illustrates that, when hip precedes shoulder involvement, there is the potential for the frozen hip to receive alternate diagnoses for which invasive open hip surgery could unnecessarily be recommended.

BACKGROUND

Adhesive capsulitis of the shoulder ('frozen shoulder') has a reported incidence of 2–5% of the general population¹ and accounts for a substantial proportion of shoulder pathology. Adhesive capsulitis is characterised by its spontaneous onset, usually without overt injury; severe pain on internal and external rotation; and a triphasic clinical course characterised initially by pain, followed by limitation of both active and passive movement, and, finally, 'thawing' with complete or substantive resolution, usually within 1–3 years.¹ Diagnosis of adhesive capsulitis of the shoulder is primarily clinical, with blood markers typically normal. Radiographic studies are not diagnostic but may exclude other causes of shoulder pain.¹ CT and synovial fluid analysis are typically normal; X-ray may reveal osteopaenia or osteoporosis of the humeral head, most likely secondary to pain and restriction of movement¹; and ultrasound, MRI and MR arthrography may show thickening of the coracohumeral ligament and increased vascularity in the rotator interval.¹ However, because clinical diagnosis is usually reliable, patients can be reassured that remission is likely and investigations are unlikely to be helpful.

A similar clinical course could be expected for adhesive capsulitis of the hips, the body's other two ball-and-socket joints. However, much less is known about this condition, and its clinical and

radiological findings, because it has been reported in only around a dozen individuals.^{2–5} A 2013 review of previous case reports suggested that ultrasound, CT, MRI and MR arthrography findings can be normal, though they may reveal concomitant pathology⁵ (which may or may not be relevant to the experienced signs and symptoms). We suspect that this debilitating but remitting condition may be systematically underdiagnosed, with the potential for unnecessary investigations and harmful intervention.

Adhesive capsulitis in all four major ball-and-socket joints in an otherwise healthy patient has not previously been reported, to the best of our knowledge.

CASE PRESENTATION

In 2006, a 47-year-old woman presented to her family doctor, with a 10-month history of spontaneous onset of left groin pain. There was no pain on sitting or walking and no mechanical symptoms, such as clicking, locking or joints giving way. However, severe pain was experienced on abduction, external rotation and, particularly, internal rotation. The patient was no longer able to sit cross-legged, and sleep had become difficult. Other than mild hypertension, she was healthy, euthyroid and not diabetic. There was no family history of hip problems, early arthritis, or immune disorders.

The patient had a healthy body mass index of 20 kg/m². Gait was normal and Trendelenburg test negative. The lumbar spine and both knees were normal. By early 2007, the left hip had clinically deteriorated and similar symptoms had developed on the right. Pain was more frequent, particularly with sitting and bending forward. An MRI scan showed no significant abnormality in the articular cartilage of the left hip but did reveal some possible anterior impingement related to the anterior rim of the acetabulum.

An orthopaedic surgeon specialising in hip problems diagnosed femoroacetabular impingement (also known as FAI). This is a clinical syndrome that can arise at any age, starting from adolescence, and is expected to gradually worsen as "the repetitive contact between the femoral neck and the acetabular rim causes damage to acetabular cartilage leading to labral tears ... and progression to early osteoarthritis of the hip."⁶ No alternative differential diagnosis was suggested. The patient declined periacetabular osteotomy to rotate the acetabulum because she felt that this major procedure requiring

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open hip surgery had no guarantee of benefit. No other treatments were offered. Over the following year, symptoms gradually improved, the patient regained near-full movement of both hips, and all pain subsided.

In 2011, the patient re-presented, at 52 years of age, with pain and loss of movement in the right shoulder. Blood count, erythrocyte sedimentation rate, C reactive protein and antinuclear antibodies were all normal. Plain X-ray showed mild degenerative changes of the acromioclavicular joint without destructive bony change, abnormal soft tissue calcification or significant subacromial space narrowing. Ultrasound showed mild subacromial bursal fluid and thickening without tendinopathy, tear or rupture, and no underlying cortical irregularity. No definitive diagnosis was made or treatment undertaken. Some months later, a diagnosis of adhesive capsulitis was made as the patient entered the 'frozen' stage, and symptoms slowly resolved, again without treatment.

In 2013, she re-presented, at 54 years of age, with similar symptoms in the left shoulder. Adhesive capsulitis of the shoulder was promptly diagnosed.

TREATMENT

The optimal treatment for frozen shoulder remains uncertain. Options include expectant management with stretching and corticosteroid injections. With regard to treatments aiming to rupture the capsular contractures, there is some randomised trial evidence that hydrodilatation (injection of saline into the joint) outperforms manipulation under anaesthetic.¹ This patient opted for hydrodilatation with cortisone injection. Fluoroscopy was consistent with capsular restriction, and the patient reported some relief.

OUTCOME AND FOLLOW-UP

As of July 2015, the patient's left shoulder signs had largely resolved with minimal residual limitation of rotation. In the interim, the patient had learnt that her older sister had also experienced bilateral frozen shoulders. Review of her earlier hip problems (and particularly their resolution over a 2-year period) seemed entirely in keeping with a diagnosis of adhesive capsulitis of both hips; she is now able to sit cross-legged again.

DISCUSSION

This is the first report, to the best of our knowledge, describing adhesive capsulitis sequentially affecting all four major ball-and-socket joints, that is, both shoulders *and* both hips, in a healthy patient. The one previous report was in a patient with the known risk factor of idiopathic hypothyroidism, whose diagnosis was more evident because the frozen hips *followed* the frozen shoulders.⁷

Twin studies demonstrated a strong genetic predisposition to shoulder capsulitis,⁸ in keeping with the frozen shoulders experienced by this patient's sibling. Capsulitis of the hip is so rarely reported that no such familial accounts exist. This study suggests that a genetic predisposition may generalise to hip, as well as shoulder, adhesive capsulitis.

Limitations of this report include the presumptive, retrospective diagnosis of adhesive hip capsulitis, against an alternate diagnosis by a hip specialist, based on contemporaneous clinical examination and radiological findings. Resolution, time course

and the subsequent similar shoulder signs and symptoms do, however, strongly favour capsulitis.

We suggest that adhesive capsulitis of the hips remains both underdiagnosed and under-reported. The course of this case illustrates the potential for incurring unnecessary investigations and treatments, including a recommendation for invasive open-hip surgery requiring intraoperative dislocation that, at best, would incur substantial costs and, at worst, lengthy morbidity and/or disability. This is especially important given that the natural history of adhesive capsulitis is ultimately benign and conservative treatment very acceptable to informed patients.

Learning points

- ▶ Unlike adhesive capsulitis of the shoulder, adhesive capsulitis of the hip is probably under-reported, and may frequently be misdiagnosed.
- ▶ This opens up the possibility of unnecessary invasive investigations and treatments because, as for back pain, imaging such as MRI may reveal abnormalities that are not the cause of the pain.
- ▶ When healthy, mid-life patients present with pain and limited passive hip rotation that are not explained by arthritis or injury, a diagnosis of adhesive capsulitis should be entertained.
- ▶ This may engender a period of watchful waiting and explanation, along with a stretching programme.
- ▶ Although adjuncts may include corticosteroid injection, hydrodilatation and/or manipulation under anaesthetic, none has yet been tested for hips with this condition.

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