

CASE REPORT

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Complex regional pain syndrome after multiple revision total hip arthroplasty surgeries

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Abstract

Background Persistent pain after total hip arthroplasty (THA) is a real concern for the surgeon and is a significant cause of patient dissatisfaction; periprosthetic joint infection (PJI), aseptic loosening, and instability are among the common causes of painful THA. However, few rare causes have been reported in the literature; the complex regional pain syndrome (CRPS) was reported to occur after primary hip and knee arthroplasties. Case presentation: We present a female patient who had bilateral hip resurfacing when she was 28 years old after being diagnosed with bilateral hip avascular necrosis; over 16 years, she was satisfied. She started developing pain in her right hip in 2019, diagnosed as aseptic loosening, and a revision using a cementless THA was performed. Six months later, she was diagnosed with PJI, which was treated in two stages; after the first-stage revision THA, the patient complained of persistent pain in her lower leg with a burning sensation and change in skin color, not responding to conservative lines. During the second-stage revision THA and after receiving spinal anesthesia, the skin color of the right lower limb changed to normal. The postoperative diagnosis was made as CRPS, treated effectively with sympathetic block. At six months postoperative, the patient was pain-free, and the implants showed proper position. Conclusions: Although rare, CRPS could cause persistent pain after repeated hip surgeries, and the diagnosis is reached mainly by exclusion.

Keywords Complex regional pain syndrome: hip resurfacing arthroplasty, Painful total hip arthroplasty, Revision total hip arthroplasty

Background

Total hip arthroplasty (THA) is one of the most successful surgical procedures, which helps in the management of various end-stage hip joint pathological conditions; however, persistent pain postoperatively is one of the main reasons for patient dissatisfaction (Erivan et al. 2019). Identifying the exact source and cause of pain is paramount for establishing a management protocol for

this particular patient and deciding whether revision surgery is needed (Pietrzak et al. 2018).

Periprosthetic joint infection (PJI), aseptic loosening, wear, and instability were suggested as the main intrinsic reasons for painful THA (Pietrzak et al. 2018; Yucuma et al. 2021). However, with the introduction of metal-on-metal (MOM) bearing surfaces, which are commonly used in hip resurfacing arthroplasty (HRA), new issues emerged, mainly the metal ions release and the subsequent related altered soft tissue reaction or what is called pseudotumor or atypical lymphocytic vasculitis and associated reactions (ALVAL) (Su 2020).

Interestingly, rare extrinsic causes of painful THA have been suggested in the literature, including complex regional pain syndrome (CRPS), which is characterized by pain distal to the site of physical trauma,

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unproportionate to the procedure performed, and little or no improvement is observed over time (Royeca et al. 2019; Zanotti et al. 2017; Hossain and Andrew 2010; Söylev and Boya 2016). The current case report describes a rare presentation of CRPS after multiple THA revision surgeries.

Case presentation

A female patient of 44 years old presented in June 2019 with persistent right hip pain; she gave a history of bilateral sequential HRA (one year apart, starting with the more affected right hip) performed when she was 28 years old after she was diagnosed with steroid-induced bilateral hip avascular necrosis; the surgeries were performed by the senior author. Over 16 years, the patient was satisfied with both her resurfaced hips, and she got married and had two siblings. Her right hip pain started insidiously with no history of trauma and increased gradually in intensity; upon clinical evaluation, she had limited right hip internal rotation and painful last degrees of all hip movements with intact hip abduction and abductor muscles strength power grades as five; however, her plain radiographs were unremarkable (no evidence of femoral neck stress fracture) and workup for PJI was negative (Fig. 1). At that time, as she had a MOM articulation, concerns related to metal ions release and ALVAL reaction were raised; unfortunately, we did not have access to serum metal ion analysis or metal artifact reduction sequence (MARS) magnetic resonance imaging (MRI) evaluation; we performed a superficial ultrasound (US) evaluation of the hip and gluteal area to detect whether there was a noticeable swelling or collection, and this turned to be negative (Garbuz et al. 2014). Conservative management was prescribed for her, and she was advised to follow up if the pain persisted. After six months of follow-up, she still has pain episodes; the plain hip radiographs showed a radiolucent line around the acetabular component (Fig. 2A). A revision THA was decided, which was performed in January 2020 through the same approach (modified direct lateral); the soft tissues appeared normal with no evidence of metallosis, the acetabular cup was loose, tissue samples were obtained and sent for culture and sensitivity, and a cementless THA was implanted (Fig. 2B and C). The culture and sensitivity results showed positive results for gram-negative bacilli; antibiotics were prescribed per organism sensitivity for six weeks. The patient presented after six months postoperatively with pain, tenderness, swelling, and redness at the surgical incision site; the erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP) were elevated, the diagnosis was established as an infected THA, and the decision

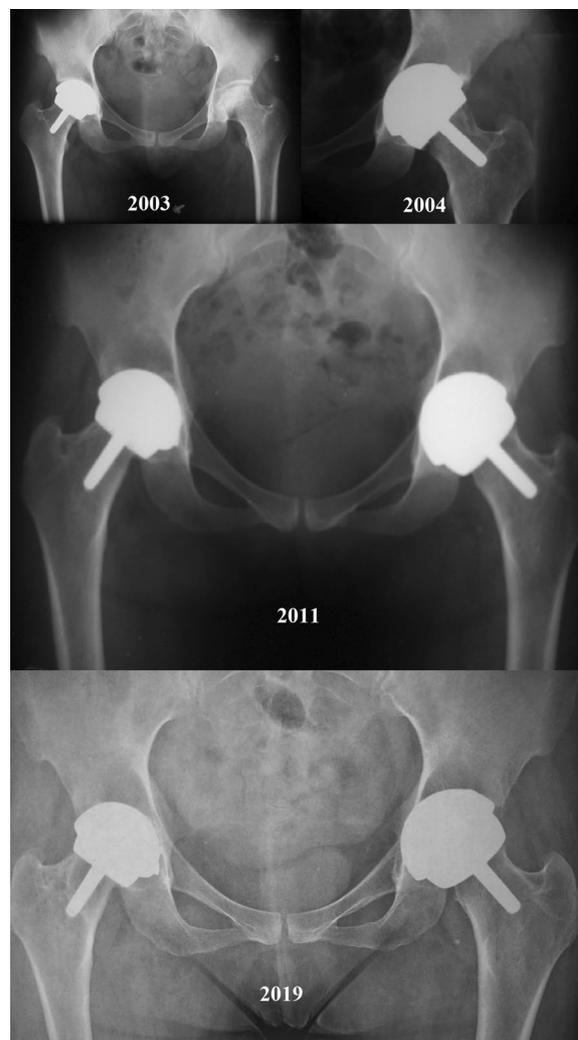


Fig. 1 Plain radiographs showing the patient's follow-ups since she had her hips replaced till she started complaining of right hip pain in 2019

was made to perform a two-stage revision. During the first stage, the acetabular cup was extracted easily; however, the femoral stem was solidly fixed, so we applied a cement ball loaded with antibiotics over the fixed stem (Fig. 2D); intraoperative cultures showed the same previous gram-negative bacilli. Postoperative antibiotics were prescribed according to the sensitivity for six weeks, after which the ESR and CRP levels were normalized; however, the patient reported a new onset of burning sensation started after the first-stage surgery extending from the knee to the dorsal aspect of the foot, increased progressively in intensity and did not respond to analgesics. Upon local examination, there was slight edema of the lower leg and dorsum of the foot with pallor and intact distal pulse. At this stage, we

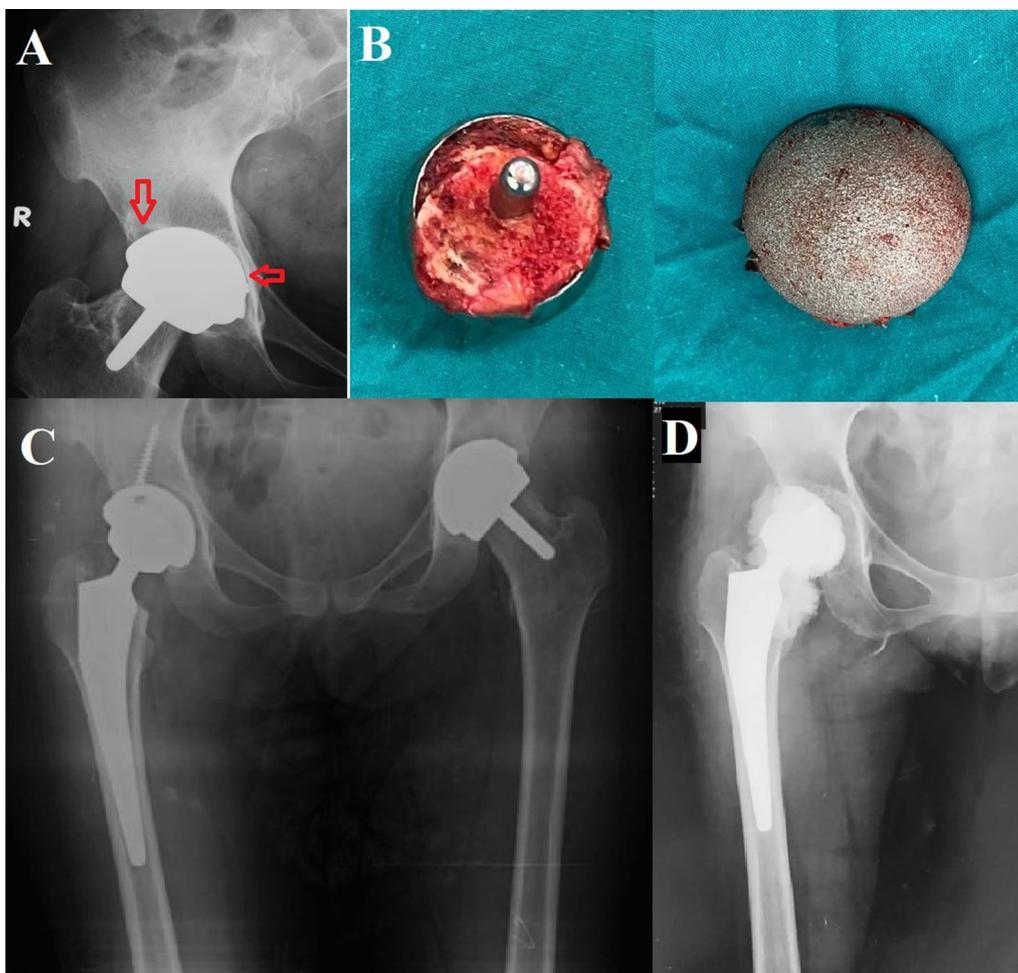


Fig. 2 Revision surgeries performed for the patient. **A** Right hip plain radiograph showing radiolucent lines around the acetabular component (red arrows). **B** An image of the hip resurfacing implants after being removed during the first revision surgery shows the acetabular component's clear back surface. **C** Cementless THA implanted at the first revision surgery. **D** After diagnosing PJI, the first-stage THA cement spacer was performed with the stem left in place, and a cement ball was inserted in the acetabulum

did not have any explanation for these new symptoms except that the pain could be referred from the hip, and the lower leg changes could be due to repeated surgeries. However, during the second-stage revision THA surgery and after the patient received spinal anesthesia, one of the surgeons' assistants and the anesthesiologist observed changing the lower limb skin color to normal, which raised the suspicion of CRPS causing her pain and symptoms. The revision surgery went unremarkable, and the intraoperative obtained samples showed no bacterial growth. The patient was transferred to a pain management consultant who confirmed the diagnosis of CRPS-I based on the patient history and examination, and he decided to treat her with a sympathetic block. After six months of follow-up, the pain was

completely relieved, and the plain radiographs showed a maintained position of the implants (Fig. 3).

Discussion

Analysis of painful THA and how to reach a proper diagnosis to decide on revision had been thoroughly studied in the literature; pain could be due to intrinsic causes such as aseptic loosening, PJI, wear, and instability or extrinsic causes such as peripheral neuropathy referred pain and myofascial/soft tissue pain (Erivan et al. 2019; Pietrzak et al. 2018; Yucuma et al. 2021). However, further issues should be considered when handling a painful HRA, mainly the ALVAL reaction with pseudotumor formation secondary to metal ion release and possible femoral neck stress fractures (Su 2020). Although rare, CRPS



Fig. 3 Radiographs obtained during the last follow-up after six months of having the second-stage revision THA, showing good positioning and stable implants

has been reported to cause unexplained disproportionate pain after total knee and hip arthroplasties (Royeca et al. 2019; Zanotti et al. 2017; Hossain and Andrew 2010; Söylev and Boya 2016).

An essential aspect for diagnosing painful HRA with the possibility of adverse local soft tissue reaction secondary to MOM components wear and metal ions release is to measure the cobalt and chromium blood levels (Su 2020; Lehtovirta et al. 2017; Campbell and Takamura 2019); furthermore, imaging studies such as MARS-MRI scan help evaluate hip soft tissue envelope (Su 2020; Connelly et al. 2018), which we could not obtain in the current case. However, a more economical and simple tool is to use superficial US to rule out the presence of pseudotumor, which showed 100% sensitivity when compared to MRI (Garbuz et al. 2014); however, the main drawback of this method is that it is operator-dependent.

Various challenges in diagnosing the exact cause of pain are given in the current case. First, the patient was diagnosed with an aseptic loosening of RHA and being free from ALVAL and pseudotumor formation; however, it turned out to be an infected THA. Second, the CRPS did not develop until the third surgical procedure. Lastly, we did not perform full investigations for the CRPS, such as foot and ankle radiographs, bone densitometry, and bone scan to detect the presence of osteoporotic changes, which were performed in previous reports (Zanotti et al. 2017; Hossain and Andrew 2010), as we did not consider

the possibility of CRPS after multiple THA revision surgeries, unlike previous reports where CRPS occurred after primary THA.

CRPS is a rare but debilitating condition, with an estimated incidence of up to 80,000 per year in the USA, and it mainly affects females (Marinus et al. 2011; Shim et al. 2019). The condition had various synonyms, such as causalgia, sympathetic reflex dystrophy, post-traumatic pain syndrome, and algodystrophy. It is divided mainly into two categories, namely CRPS-I where an injury to a peripheral nerve is absent, and CRPS-II (known as causalgia), where a peripheral nerve injury or damage is evident (Marinus et al. 2011; Chang et al. 2019).

The duration of symptoms onset could vary between patients, which could start immediately postoperatively or after a few weeks; furthermore, there is no gold standard test to diagnose this condition, and the diagnosis is mainly made by exclusion (Crane 2019). However, most physicians rely on patient history and clinical evaluation based on the Budapest consensus criteria for diagnosing CRPS (Oh et al. 2019). Proper diagnosis and management of CRPS require the cooperation of a multidisciplinary team approach with the involvement of a physiotherapist and pain management team, medical management in the form of non-steroidal anti-inflammatory drugs (NSAIDs) and gabapentin could be tried first; however, if the patient is unresponsive, an attempt of trigger-point injections, regional nerve blocks, sympathetic nerve block, and surgical sympathectomy could be attempted (Crane 2019).

Zanotti et al. reported on three patients who developed CRPS after elective primary THA performed through a posterolateral approach; two of the patients responded to medical and physiotherapy management; however, the third patient needed a sympathetic block (Zanotti et al. 2017). This differs from the current case, as the patient underwent three hip surgeries without developing this condition; however, CRPS developed at the time of the fourth surgery. We also obtained good results after sympathetic block, as in one of the cases in the Zanotti et al. report, which was described as effective management if the patient was not responding to medical management (Crane 2019; O'Connell et al. 2016).

Although the exact etiologies behind CRPS are unclear, CRPS-II is known to occur in relation to nerve injury or nerve trauma, in the current case as we used a modified direct lateral approach to the hip, which endangers the superior gluteal nerve (Chomiak et al. 2015; Piponov et al. 2021); however, the safety of the nerve was ensured intraoperatively by limitation of the proximal wound extension less than three cm above the tip of the greater trochanter. The clinical evaluation at each follow-up showed that the gluteal muscles' function was intact. We are unaware of any report regarding

CRPS-II after superior gluteal nerve injury; however, this condition was reported after sciatic nerve injury secondary to intramuscular injection (Bicer et al. 2012).

Conclusions

Although rare, complex regional pain syndrome should be considered when dealing with painful total hip arthroplasty, especially when other causes are excluded and if the pain is persistent, unproportionate, and not responding to usual management lines. The diagnosis is made mainly by exclusion. This condition was reported after elective THA; however, it is possible to present after multiple THA revisions.

Abbreviations

THA	Total hip arthroplasty
PJI	Periprosthetic joint infection
CRPS	Complex regional pain syndrome
MOM	Metal on metal
HRA	Hip resurfacing arthroplasty
ALVAL	Atypical lymphocytic vasculitis and associated reactions
MARS	Metal artifact reduction sequence
US	Ultrasound
NSAIDs	Non-steroidal anti-inflammatory drugs
MRI	Magnetic resonance imaging
ESR	Erythrocyte sedimentation rate
CRP	C-reactive protein

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Author contributions

AMA conceived the case report and performed the surgery. MFA carried out data acquisition and assessment. AAK and MFA performed a literature search, drafted the manuscript, and designed the figures. All authors discussed the final version of the manuscript and approved it.

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Availability of data and materials

All the data regarding the presented case are included within the article.

Declarations

Ethics approval and Consent to participate

The ethical committee of our institution waived ethical approval for this case report as this was considered a part of the usual patients' care (Faculty of Medicine, Assiut University, Egypt (Telephone, Fax: +20882332278, ethics-committee12@yahoo.com, <http://afm.edu.eg>).

Consent for publication

A verbal as well as an informed written consent was obtained from the patient to use her clinical data and images for publication of this case report, no identification of the patients' identity is present neither in the manuscript nor in the images.

Competing interests

The authors declare that they have no competing interests.

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