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Case report

Atypical femoral fracture as the cause of greater trochanteric pain syndrome – a case report[☆]

Lise Langeland Larsen, MD^a, Jeppe Lange, MD, PhD^{b,c,*}^aDepartment of Orthopedic Surgery, Aarhus University Hospital, Palle Juul-Jensens Boulevard 99, Aarhus 8200, Denmark^bH-HiP, Department of Orthopedic Surgery, Regional Hospital Horsens, Sundvej 30, Horsens 8700, Denmark^cDepartment of Clinical Medicine, Aarhus University, Aarhus, Denmark

ARTICLE INFO

Article history:

Received 6 January 2021

Revised 20 January 2021

Accepted 22 January 2021

Keywords:

Atypical femoral fractures

Greater trochanteric pain syndrome

Lateral hip pain

Magnetic resonance imaging

Looser zone

ABSTRACT

Greater trochanteric pain syndrome may be caused by atypical femoral fractures, and this should be taken into consideration in the diagnostic workout. A 63-year-old woman was referred to our orthopedic outpatient hip clinic with a history of greater trochanteric pain syndrome without known trauma for 1 year. Initially X-ray of the hip and magnetic resonance imaging were found without pathology, and she was given a diagnosis of gluteus medius tendinopathy. As physiotherapy and steroid injections did not resolve her pain, a second look on the magnetic resonance imaging and X-ray revealed a discrete atypical femoral fracture in the lateral cortex with the presence of an isolated Looser zone, which were attributed to her pain syndrome. Two years after onset of symptoms, and with no pain relief on medical treatment, she was treated with an intramedullary nail. One-year postoperative the patient was pain free. This case emphasizes the important utility of magnetic resonance imaging in refractory greater trochanteric pain syndrome.

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Introduction

Greater trochanteric pain syndrome (GTPS) challenges the clinician in the diagnostic setup [1]. Many structures are acknowledged as potential lateral hip pain generators in GTPS.

Trochanteric bursitis has historically been identified as the main pain generator, but also deep gluteal structures, spinal nerve entrapment, hip arthrosis, and hip abductors are included in the spectrum. This highlights the importance of appropriate medical imaging [2–4], especially in refractory GTPS cases [5].

[☆] Competing Interests: The authors have declared that no competing interests exist.

* Corresponding author.

E-mail address: jeppe.lange@clin.au.dk (J. Lange).

<https://doi.org/10.1016/j.radcr.2021.01.044>

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Atypical femoral fractures need to be recognized as a potential lateral hip pain generator in patients with GTPS.

Case

A 63-year-old woman was referred to the senior author by her general practitioner with a history of GTPS for 1 year. The pain had started during the summer of 2016. She stated that the lateral hip pain was not caused by any trauma, but had increased gradually during a period of high activity. Her pain was accentuated when standing, and walking on slopes. But also, classically, no pain was reported, when riding a bicycle. The pain was unilateral, and she had no other musculoskeletal complaints to the affected limb. She had a medical history of myxedema treated with levothyroxine since 2010, nonerosive, sero-negative rheumatoid arthritis treated with methotrexate since 2006, and osteoporosis treated with a 150 mg tablet of Bonviva (Atrahs Pharma) every fourth week, since 2008. The patient reported no history of alcohol or tobacco use.

At the initial physical examination in July 2017 by the senior author, severe pain at palpation were detected at the attachment of the gluteus medius tendon at the greater trochanter, and this was identified like the pain she normally had. There was normal 2-legged squat, negative 1-legged Trendelenburg stance, and negative Obers test. Standard X-ray examination of the hip was described as normal by a dedicated musculoskeletal radiologist.

A diagnosis of GTPS interpreted as caused by gluteus medius tendinopathy was made, and she was treated with a steroid injection in the greater trochanteric bursae and subsequently physiotherapy.

As the treatment had no effect on the lateral hip pain, a magnetic resonance imaging (MRI) scan of the hip was performed in September 2017. The MRI was initially interpreted without any abnormalities in relation to the suspected area of pathology - the hip abductors and the greater trochanteric bursae - by a dedicated musculoskeletal radiologist, and the patient was continued on physiotherapy.

But as the pain persisted, she was seen in December 2017, where scrutiny of the medical imaging did indeed show a minute, very localized inflammation in the lateral femoral cortex on MRI, which had not been identified initially, and the X-ray did reveal an atypical femoral fracture with the presence of a Looser zone in the lateral cortex at the same location of the inflammation (Figs. 1 and 2).

The findings were interpreted as a Looser-Milkman insufficiency fracture, and were attributed as the cause of her GTPS. Watchful waiting combined with limited weight bearing was initiated. At this point the patient had normal vitamin D and parathyroid hormone status, and normal serum calcium and alkaline phosphatase. Thyroid stimulating hormone level were consistent with thyrotoxicosis. A dual-energy X-ray absorptiometry in April 2018 showed osteoporosis with decreased T-scores (lumbar score -1.4 and hip score -2.5). The patient was seen by an endocrinologist and thyrotoxicosis was a suspected contributor to the progression in her osteoporosis.

Over the next 7 months the pain worsened. A renewed X-ray and MRI showed no signs of regression, nor progression,



Fig. 1 – X-ray showing a Looser zone in the lateral cortex 5.4 cm beneath the greater trochanter.

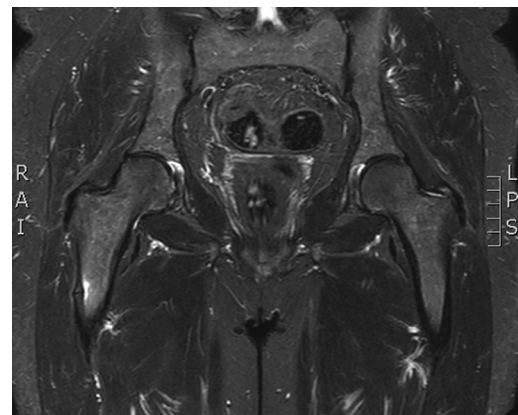


Fig. 2 – Magnetic Resonance Imaging of the Looser zone. Pathological abnormalities (Looser zone and edema of the bone) were seen in STIR and T1 sequences.

of her Looser zone. She was finally treated in September 2018 with a long intramedullary nail for pain relief (Fig. 3), and to avoid secondary fracture [6]. At 1-year follow-up, there were radiological signs of healing (Fig. 4), and the patient was free of any lateral hip pain, and reported no sequelae to the surgery.

Discussion

We describe a case of GTPS, which were attributed to an atypical femoral fracture presumably caused by prolonged bisphosphonate use, with the imaging feature of an isolated Looser zone. The presentation of GTPS in our case was identical to what we normally see in our outpatient hip clinic with gluteal pathologies leading to lateral hip pain: a female over the age of 50 with increasing lateral hip pain, no trauma, and pain over the greater trochanter area on clinical exam.

As in our case, radiological findings of Looser zones are easily overseen, and difficult to diagnose, and MRI can play an important role in early diagnosis [7]. In 1920, Looser described



Fig. 3 – X-ray after insertion of a long intramedullary nail.

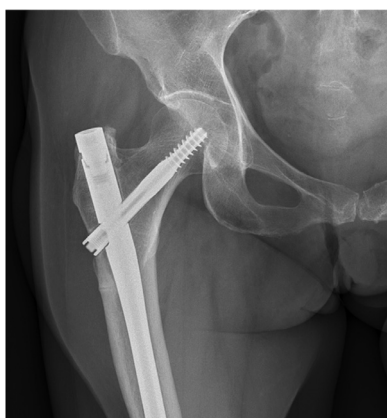


Fig. 4 – One-year postoperative X-ray show radiological signs of healing.

these zones as transverse or oblique fissures or band-like radiolucency in cortical bone [8,9]. They are often bilateral, symmetric, and lie perpendicular to the cortical margins of bones. They are most commonly found at the femoral neck, on the medial part of the femoral shaft under the lesser trochanter, and in the pubic and ischial rami. The margins of the radiolucent bands are well defined, mildly sclerotic, and there is no evidence of formation of callus. They are associated with little or no trauma, and rarely heal without treatment [10]. Looser explained the lesions on the basis of excessive strain, both muscular and postural, on already weakened bone [9]. Presence of a Looser zone is now recognized as a stress fracture [11]. Stress fractures are typically classified into 2 types depending on whether the bone is normal (fatigue fracture) or abnormal (insufficiency fracture). Risk factors include rheumatoid arthritis, metabolic bone disease, corticosteroid therapy, among others [12]. In our case,

this has the anatomic location, radiologic appearance, and clinical features of a bisphosphonate-related insufficiency fracture. Thorough medical examination including repeated medical imaging is recommended, and internal fixation could be offered if an insufficiency fracture is not healing to avoid secondary fracture [6]. In our case, the patient showed no signs of healing or callus on repeat X-ray and MRI, and as no medical or conservative treatment helped the pain, internal fixation was performed with good pain relief.

MRI is the gold standard in early recognition of stress fractures [7]. We have identified a similar case [13] in which GTPS was believed to be caused by pain generated by a femoral osteoid osteoma, but were only identified after a hip fracture had occurred. This case could have benefitted from a first-line MRI exam.

Our case highlights the application of MRI as an important diagnostic modality, with a low threshold for performing, in refractory GTPS. Atypical femoral fractures must be kept in mind in refractory GTPS.

Patient consent

The patient was informed that data concerning the case would be submitted for publication, and the patient has agreed to this.

REFERENCES

- [1] Fearon AM, Scarvell JM, Neeman T, Cook JL, Cormick W, Smith PN. Greater trochanteric pain syndrome: defining the clinical syndrome. *Br J Sports Med* 2013 Published online. doi:10.1136/bjsports-2012-091565.
- [2] Kong A, Vliet A, Zadow S. MRI and US of gluteal tendinopathy in greater trochanteric pain syndrome. *Eur Radiol* 2007 Published online. doi:10.1007/s00330-006-0485-x.
- [3] Fearon AM, Scarvell JM, Cook JL, Smith PNF. Does ultrasound correlate with surgical or histologic findings in greater trochanteric pain syndrome? A pilot study. *Clin Orthop Relat Res* 2010 Published online. doi:10.1007/s11999-009-1174-2.
- [4] Long SS, Surrey DE, Nazarian LN. Sonography of greater trochanteric pain syndrome and the rarity of primary bursitis. *Am J Roentgenol* 2013 Published online. doi:10.2214/AJR.12.10038.
- [5] Kingzett-Taylor A, Tirman P, Feller J, McGann W, Prieto V, Wischer T, et al. Tendinosis and tears of gluteus medius and minimus muscles as a cause of hip pain: MR imaging findings. *Am J Roentgenol* 1999 Published online. doi:10.2214/ajr.173.4.10511191.
- [6] Rasmussen M, Lange J. Treatment with bisphosphonates as a possible cause of bilateral spontaneous atypical femoral fracture. *Ugeskr Laeger* Jan 9 2012;174(1-2):32–3.
- [7] Berger FH, de Jonge MC, Maas M. Stress fractures in the lower extremity. The importance of increasing awareness amongst radiologists. *Eur J Radiol* 2007 Published online. doi:10.1016/j.ejrad.2007.01.014.
- [8] Looser E. Über Spätrachitis und Osteomalacie. *Klinische, röntgenologische und pathologisch-anatomische Untersuchungen. Dtsch Zeitschrift Für Chir* 1920 Published online. doi:10.1007/BF02798519.

- [9] Looser E. *Über pathologische Formen von Infraktionen und Callusbildungen bei Rachitis und osteomalakie und anderen knochen Erkrankungen.* *Zentralbl Chir* 1920;47:1470–4.
- [10] McKenna MJ, Heffernan E, Hurson C, Mckiernan FE. Clinician approach to diagnosis of stress fractures including bisphosphonate-associated fractures. *QJM* 2014 Published online. doi:[10.1093/qjmed/hct192](https://doi.org/10.1093/qjmed/hct192).
- [11] McKenna MJ, Kleerekoper M, Ellis BI, Rao DS, Parfitt AM, Frame B. Atypical insufficiency fractures confused with looser zones of osteomalacia. *Bone* 1987 Published online. doi:[10.1016/8756-3282\(87\)90073-1](https://doi.org/10.1016/8756-3282(87)90073-1).
- [12] Matcuk GR, Mahanty SR, Skalski MR, Patel DB, White EA, Gottsegen CJ. Stress fractures: pathophysiology, clinical presentation, imaging features, and treatment options. *Emerg Radiol* 2016 Published online. doi:[10.1007/s10140-016-1390-5](https://doi.org/10.1007/s10140-016-1390-5).
- [13] Sferopoulos NK. Subtrochanteric osteoid osteoma: A misdiagnosed case complicated by a hip fracture. *Chinese J Traumatol English Ed* 2016 Published online. doi:[10.1016/j.cjtee.2016.03.006](https://doi.org/10.1016/j.cjtee.2016.03.006).