A Rare Type of Atypical Fracture Femur without Bisphosphonate Therapy: A Case Report

Ashwin Suresh[a], Tomichan Mattathil Chacko[b]

a. Undergraduate Medical Student, Al Azhar Medical College and Super Speciality Hospital, Thodupuzha, Idukki, Kerala;
b. Department of Orthopedics, Al Azhar Medical College and Super Speciality Hospital, Thodupuzha, Idukki Kerala*

Published on 1st July 2024

ABSTRACT

Atypical femoral fractures are stress fractures occurring in femoral shaft usually associated with prolonged use of antiresorptive therapy. Various reports have shown that long term use of rheumatoid arthritis medications may also impair mineralization of bone. Here we are discussing a case of atypical fracture femur seen in person who was not on bisphosphonate therapy.

Keywords: Bisphosphonates, Osteoporosis, Atypical Fracture Femur

*See End Note for complete author details

INTRODUCTION

Atypical femoral fractures are stress or insufficiency fractures occurring in femoral shaft usually associated with prolonged use of antiresorptive therapy for osteoporosis. Repeated use of bisphosphonate therapy leads to inhibition of osteoclast function leading to abnormal mineralization of bone resulting in atypical fracture femur.[1] But some researchers had reported atypical femoral fractures in whom bisphosphonates were not used.

CASE REPORT

History

A 44yr old woman who was on treatment for rheumatoid arthritis and hyperthyroidism for 8yrs, presented to emergency department with alleged history of slip and fall from scooter. She was complaining of bilateral hip pain for the last 2 years. She was allergic to Phenergan and Thrombophob.

Examination

The patient was moderately built and nourished with mild pallor. Her left lower limb was shortened and externally rotated with clinical features suggestive of fracture of proximal femur. She also had tenderness over her right proximal thigh. All other systemic findings were normal.

Radiological findings

X-rays showed displaced subtrochanteric fracture of left femur having transverse fracture line and a localized thickening of the lateral cortex at the fracture sites. Radiologically looser zone was seen on right and left hip regions. On right side, lateral cortical thickening and sclerosis present in subtrochanteric region with no evidence of a clear fracture. **Figure 1** and **Figure 2** shows x-ray findings of atypical fracture femure.

Differential diagnosis considered were Atypical fracture femur, Stress fracture, Osteogenic imperfecta, and Pathological fracture. Stress fractures are tiny cracks seen in bone caused by repetitive force among athletes.[12] In this case there was no history of repeated stress. Osteogenic imperfecta is a hereditary condition resulting in repeated fracture due to collagen abnormalities.[13] There were no abnormalities detected in clinical findings or investigations suggestive of pathological fractures.

All haematological and metabolic investigations were normal. Biopsy taken from fracture site on left side

Cite this article as: Suresh A, Chacko TM. A Rare Type of Atypical Fracture Femur without Bisphosphonate Therapy: A Case Report. Kerala Medical Journal. 2024 Jul 1;17(2):110–2.

Corresponding Author:
Dr. Tomichan Mattathil Chacko, Professor, Department of Orthopaedics, Al Azhar Medical College and Super Speciality Hospital, Thodupuzha, Idukki, Kerala E-mail: mtomichan@hotmail.com

Figure 1. **Atypical subtrochanteric fracture left femur**

showed necrotic bony fragments suggestive of micro fracture.

Thus, based on the clinical, radiological and laboratory findings, the diagnosis of Atypical fracture of proximal left femur and impending fracture right femur was made. She was treated with open proximal femoral nailing of left femur followed by closed prophylactic proximal nailing of right femur (PFNA2) and walking was advised with help of walker assistance after 6 weeks. **Figure 3** shows proximal femoral nailing done on both femur.

DISCUSSION

Atypical fracture femurs are stress type fractures located at subtrochanteric region and diaphysis of the femur which occur with minimal or no trauma and having specific radiographic findings.[1] The risk is increased with bisphosphonate use which may result in increased susceptibility in developing atypical fracture femur.[2,4] The diagnosis follows major and minor criteria described by the Task Force of the American Society for Bone and Mineral Research (ASBMR).[3]

Major Criteria and Minor Features of Atypical Femoral Fracture

Major

- Fracture associated with minimal or no trauma.
- Fracture is non-comminuted.
- Fracture is transverse or short oblique in configuration.
- Be located anywhere along the femur distal to the lesser trochanter and proximal to the supracondylar flare.

Figure 2. **Atypical subtrochanteric fracture left femur along with impending fracture of right subtrochanteric region**

Minor

- Localised periosteal or endosteal thickening of the lateral cortex present at the fracture site.
- General increase in cortical thickness of the femoral diaphysis.
- Bilateral fractures.
- Delayed fracture healing.
- Presence of coexisting conditions such as vitamin D deficiency, hypophosphatasia and rheumatoid arthritis.
- Unilateral or bilateral prodromal symptoms of thigh or groin pain.

Repeated use of bisphosphonate therapy leads to inhibition of osteoclast function leading to increased mineralization.[6] The bone produced is biomechanically weak resulting in atypical fracture femur.[11] The relative risk of patients developing an atypical femoral fracture when taking bisphosphonates is high ranging from 3.2 to 50 cases per 1,00, 000.[3] Besides prolonged and continuous use of bisphosphonates, long-term use of glucocorticoids and high BMI are also risk factors of atypical femur fractures.[11] Anterolateral femoral bowing and loss of thigh muscle seem to have an influence on the occurrence of these fractures.[5] Various reports in lit-

Figure 3. **Proximal femoral nailing done for both femur**

erature have stated the occurrence of atypical fracture femur without bisphosphonate therapy.[5] Incidence of other pharmacological treatments which decrease bone turnover[3] leading to atypical femoral fractures are non-Hodgkin's lymphoma,[9] vitamin D deficiency and rheumatoid arthritis. Proton pump inhibitor intake changes resorption leading to different forms of malnutrition and increase risk of fractures.[1] The presence of a genetic metabolic bone disorder like hypophosphatemia[8] and varus hip alignment increase tensile stress leading to the development of atypical fracture femur.[2] Long term use of rheumatoid arthritis medications which may impair mineralization caused by osteomalacia are considered as risk factors.[10] Disease modifying antirheumatic drug use may decrease bone strength and increases the incidence of insufficiency fractures.[3] Radiographic endosteal thickening at the lateral femoral cortex was observed less frequently in patients with atypical fracture femur not treated with bisphosphonates than in patients treated with bisphosphonate.[7] In our case the patient might be suffering from atypical fracture femur due to long term intake of rheumatoid drugs.

CONCLUSION

This case report highlights that some patients may sustain atypical femur fracture even without exposure to bisphosphonate. The absence of bisphosphonate therapy and typical radiographic findings like focal periosteal or endosteal thickening at the lateral femoral cortex, may complicate the early diagnosis and implementation of preventive measures. Prolonged use of steroids and drugs related to treatment of osteomalacia may also contribute to atypical fracture femur. Combination therapy with vitamin D and calcium effectively enhances healing of atypical fracture femur.

END NOTE

Author Information

1. Dr Ashwin Suresh, Third year MBBS, Undergraduate Medical student, Al Azhar Medical College and Super Speciality Hospital, Thodupuzha Idukki, Kerala
2. Dr Tomichan Mattathil Chacko Professor, Department of Orthopaedics, Al Azhar Medical College and Super Speciality Hospital, Thodupuzha, Idukki, Kerala

Conflict of Interest: None declared

Terminology :

Osteoporosis: A bone disease that develops when bone mineral density and bone mass decreases, or when the structure and strength of bone changes

Prophylactic Nailing: A procedure to stabilize the femoral bone that has become weak.

Conflict of Interest: None declared

Financial Support: Nil

Acknowledgement: We would like to thank the patient for cooperation and support.

REFERENCES

1. Silverman S, Kupperman E, Bukata S. Bisphosphonate-related atypical femoral fracture: Managing a rare but serious complication. Cleve Clin J Med. 2018 Nov;85(11):885–93.
2. Georgiadis GF, Begkas DG, Maniatis KA, Vasilakis AE, Chatzopoulos STD, Balanika AP. Atypical Femoral Fracture in a Patient without Bisphosphonate or Denosumab Exposure-A Case Report. J Orthop Case Rep. 2021 Oct;11(10):21–4.
3. Shane E, Burr D, Abrahamsen B, Adler RA, Brown TD, Cheung AM, et al. Atypical subtrochanteric and diaphyseal femoral fractures: second report of a task force of the American Society for Bone and Mineral Research. J Bone Miner Res. 2014 Jan;29(1):1–23.
4. Khan AA, Kaiser S. Atypical femoral fracture. CMAJ. 2017 Apr 10;189(14):E542.
5. Tile L, Cheung AM. Atypical femur fractures: current understanding and approach to management. Ther Adv Musculoskelet Dis. 2020;12.
6. Girgis Christian M., Sher Doron, Seibel Markus J. Atypical Femoral Fractures and Bisphosphonate Use. New England Journal of Medicine. 2010;362(19):1848–9.
7. McKenna MJ, Kleerekoper M, Ellis BI, Rao DS, Parfitt AM, Frame B. Atypical insufficiency fractures confused with Looser zones of osteomalacia. Bone. 1987;8(2):71–8.
8. Saviola G, Prometti P, Ferrari PR, Dalle Carbonare LG. Atypical femoral fracture as first sign of non-hodgkin lymphoma- case report with a brief review of the literature. Clin Ter. 2016;167(6):188–90.
9. Lee SH, Lee YH, Suh JS. Lateral Cortical Thickening and Bone Heterogeneity of the Subtrochanteric Femur Measured With Quantitative CT as Indicators for Early Detection of Atypical Femoral Fractures in Long-Term Bisphosphonate Users. AJR Am J Roentgenol. 2017 Oct;209(4):867–73.
10. Woo SB, Choi ST, Chan WL. Atypical periprosthetic femoral fracture: a case report. J Orthop Surg (Hong Kong). 2016 Aug;24(2):269–72.
11. Koh JH, Myong JP, Yoo J, Lim YW, Lee J, Kwok SK, et al. Predisposing factors associated with atypical femur fracture among postmenopausal Korean women receiving bisphosphonate therapy: 8 years' experience in a single center. Osteoporos Int. 2017 Nov;28(11):3251–9.
12. May T, Marappa-Ganeshan R. Stress Fractures. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2024 [cited 2024 Jun 20].
13. Lewiecki EM. Osteoporosis: Clinical Evaluation. In: Feingold KR, Anawalt B, Blackman MR, Boyce A, Chrousos G, Corpas E, et al., editors. Endotext [Internet]. South Dartmouth (MA): MDText.com, Inc.; 2000 [cited 2024 Jun 20].