Received: 16 July 2022 | Revised: 7 June 2023 | Accepted: 23 June 2023

DOI: 10.1002/ccr3.7881

CASE REPORT

Clinical Case Reports Open Access WILEY

Simultaneous bilateral basicervical femoral neck fractures in a patient with osteomalacia: A case report and literature review

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Key Clinical Message

A thin patient with a history of eating disorders developed basicervical femoral neck fracture bilaterally and simultaneously due to vitamin D deficiency osteomalacia. A careful evaluation in thin patients with thigh pain, including bone biopsy, is required to avoid overlooking osteomalacia.

KEYWORDS

basicervical, bone morphometry, femoral neck fracture, osteomalacia, pain, vitamin D deficiency

1 | INTRODUCTION

Osteomalacia has been a serious health problem in developed countries over a century ago due to malnutrition.[1] However, a histological examination of bone biopsies has shown that the prevalence of osteomalacia at postmortem reached as high as 25% in adult Europeans in the 2000s.[2] In recent years, osteomalacia has no longer been considered a common lifestyle disease, suggesting that patients with osteomalacia may not have been appropriately diagnosed and managed.[3] However, reports examining the prevalence of osteomalacia in hip fractures via bone biopsy from 1974 to 2020 showed results in the range of 2%–37%.[4–6] The prevalence tended to be higher in colder regions and older days. The factors involved are increased ultraviolet B exposure at greater altitudes.[5–7] It may also be related to immigration, race, religion, skin color, and use of topical sunscreen.[7] Osteomalacia has various causes,[3,4] one of which is severe vitamin D deficiency, although both severe and mild vitamin D deficiencies are now uncommon in developed countries.[7] Moreover, poor nutritional intake due to inappropriate dieting has been known to cause osteomalacia due to vitamin D deficiency,[3] which has become a recent problem.

In Japan, the 2019 National Health Survey report by the Ministry of Health, Labour, and Welfare showed that 11.8% of the total population corresponds to thin women with a body mass index (BMI) less than 18.5 kg/m2,[8] and teenagers accounted for the highest percentage at 21.0%. Approximately 10% of middle-aged women ≥60 years are also thin. It is quite common for Japanese women to be thin with a BMI of <18.5 kg/m^2. Japanese women have a strong desire to be thin, and many of them are underweight compared with their Western counterparts. The

risk of low nutrition, bone fractures, and low birth weight owing to emaciation are major concerns in Japanese women as a whole.[9]

Unlike osteoporosis, osteomalacia does not involved changes in bone mass; however, changes in the ratio of bone matrix and bone mineral occur because of calcification disorder.[4] On the contrary, like osteoporosis, osteomalacia promotes an increased risk for fractures. However, nontraumatic bilateral femoral neck fractures are rare even in elderly people with osteoporosis.[10]

Basicervical femoral neck fractures are rare and account for 1.7%–1.8% of all hip fractures. They are characterized by the highest degree of rotational instability among hip fractures.[11,12] Based on our reviews, there were several reports of bilateral femoral neck fractures.[10,13–19] However, simultaneous bilateral basicervical femoral neck fractures have never been reported. Furthermore, no case reports of patients diagnosed with osteomalacia by bone morphometry of nondecalcified specimen are available.

We report an extremely rare case of bilateral basicervical femoral neck fractures due to vitamin D deficiency osteomalacia diagnosed using bone morphometry of the iliac bone taken during surgery.

2 | CASE PRESENTATION

A 68-year-old woman who consulted with an orthopedic practitioner complained of progressive bilateral thigh pains for 3 months without any history of trauma. A pelvic X-ray revealed no abnormalities. She was suspected of having spinal stenosis and prescribed duloxetine hydrochloride and pregabalin for 2 months; however, her symptoms did not improve, and she was referred to our hospital. The patient had a history of an eating disorder during adolescence. During her transition to adulthood, she resorted to excessive dieting due to a poor self-perceived body image. She also had a history of femoral head bone and pelvic fractures 3 years ago. She was suspected of having osteomalacia but refused further evaluation.

The patient had a height of 146 cm, body weight of 35 kg, and BMI of 16.4 kg/m2. Although she had bilateral thigh pain while standing, she was able to walk without a cane at her first visit and had no tenderness in the thigh. Her lower extremity muscle strength and sensory function were generally normal. Her laboratory parameters were as follows: hemoglobin (Hb), 8.2 g/dL (normal range: 11.2–15.2 g/dL); calcium (Ca), 8.5 mg/dL (8.5–10.1 mg/dL); phosphorus (P), 1.5 mg/dL (2.5–4.5 mg/dL); alkaline phosphatase (ALP), 647 U/L (110–320 U/L); ALP3, 64.9% (25.2%–54.2%); albumin, 3.6 g/dL (4.0–5.2 g/dL); intact parathyroid hormone (PTH), 165 pg/mL (10–65 pg/mL); and 25-hydroxy vitamin D (25(OH)D), 6.0 ng/mL (sufficiency: >30 ng/dL). Her fibroblast growth factor 23 level was <10 pg/mL (12.9–52.9 pg/mL). Dual-energy X-ray absorptiometry revealed a significant decrease in bone mineral density (BMD) of the right femoral bone (0.376 g/cm2; T-score, −7.61; Z-score, −4.45) and lumbar spine (0.560 g/cm2; T-score, −4.00; Z-score, −1.64). A pelvic X-ray did not reveal a Looser's zone or obvious abnormalities in both femoral neck (Figure 1). She was suspected of having occult fractures or symptoms associated with lumbar spinal canal stenosis. However, because she did not possess the typical findings of lumbar spinal stenosis, she was also suspected of having metastatic tumors, malignant complications, or a blood disease. A T2-weighted magnetic resonance imaging sequence showed only a slight bilateral signal change in the proximal femur that could be assumed as artifacts with no clear fracture line. We

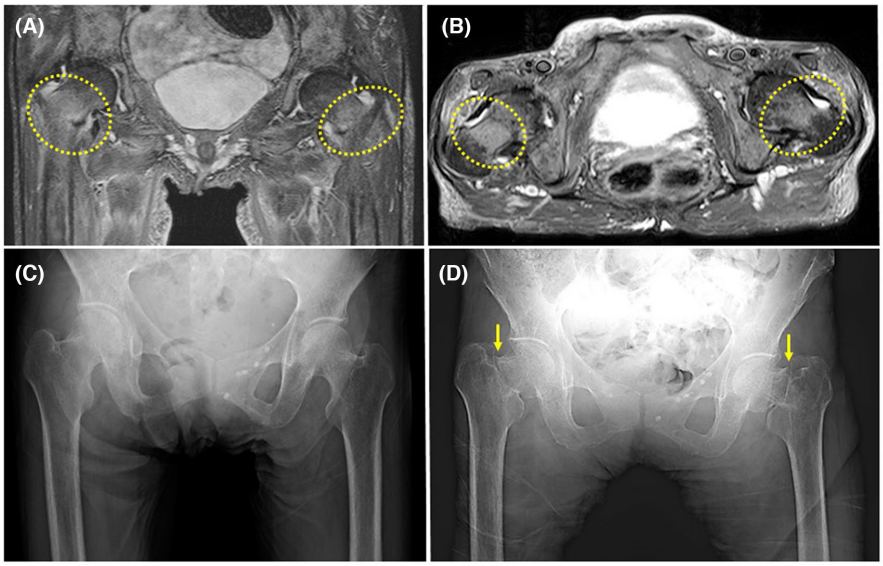

FIGURE 1 Image findings at patient's first and second visit. At the patient's first visit; (A, B) T2-weighted magnetic resonance images showed a slight bilateral signal change in the proximal femur with no clear fracture line (yellow dot circles); (C) no Looser's zone in both femoral necks and no obvious abnormalities on X-rays. At patient's second visit; (D) an X-ray 1 month after the initial examination showed a bilateral basicervical femoral neck fractures with displacement (yellow arrows).

preferred that she was hospitalized for further examination and treatment; however, she was unaware of her illness and refused hospitalization, so we decided to follow up after 1 month. After 1 month, the patient's thigh pain had gradually increased as she was unable to walk at her second visit. An X-ray showed bilateral basicervical femoral neck fractures, which prompted suspicions of a fragility fracture due to a metabolic bone disease, among which osteomalacia was primarily suspected. No observations of malignant disease were noted upon examination, and the laboratory data showed no significant changes compared with the previous results. She was immediately admitted to the hospital, and considering her condition, we decided that fracture treatment was the best option. She underwent osteosynthesis and a short femoral nail fixation for both fractures. Therefore, her bone fragility could not be assessed preoperatively. Thus, we simultaneously performed a bone biopsy on the right ilium to establish a definitive diagnosis. Subsequently, a nondecalcified specimen was prepared (Figure 2). The parameters of bone volume and trabecular thickness were normal on bone histomorphometry (Table 1). However, we found a remarkable increase in osteoid parameters (e.g., osteoid mass, 54.9%; osteoid width, 34.5 μm), which suggested bone mineralization disorder. Bone erosion had also increased significantly (erosion surface, 11.7%), osteoclasts were abundant, and fibrous tissue was not observed. As such, a diagnosis of osteomalacia was established based on the osteoid volume (OV/BV) >10% and thickness (O.Th) >12.5 μm.[20] The pathological findings also showed enhanced bone resorption in the cancellous and cortical bones. Thus, the current case presented a combination pathophysiology of both osteomalacia and osteoporosis.

The patient showed no signs of intestinal malabsorption or renal dysfunction; however, she had vitamin D deficiency. Based on these results, a diagnosis of osteomalacia with vitamin D deficiency was established.[3] After 1 month of hospitalization, her Ca, P, and intact PTH levels normalized with regular hospital food (2000 kcal/day). Approximately 2 months after the surgery, she was diagnosed with vitamin D deficiency osteomalacia based on the results of the bone morphometry of a nondecalcified specimen. Therefore, she was prescribed calcitriol at 0.5 μg/day. We observed good fracture fusion 3 months after the surgery, and 6 months thereafter, her Hb and ALP levels normalized. After a further 8 months, the BMD of her lumbar spine improved (1.111 g/cm2; *t*-score, −0.06; *z*-score, 1.69).

3 | DISCUSSION

We describe a case of a 68-year-old woman with BMI of 16.4 kg/m2 who developed bilateral basicervical fractures of the femur without any trauma. It is a very instructive and important case because vitamin D deficiency osteomalacia as a result of excessive dieting caused this uncommon fracture bilaterally and simultaneously.

Several case reports have been available on simultaneous bilateral femoral neck fractures, despite being a rare disease.[10,13–19] The age of the patients varied from teenagers to the elderly in their 60s. Factors contributing to fractures included prevention of sun exposure for religious reasons[13,14] or lifestyle,[10,15] malnutrition and low body weight,[10,16] vitamin D deficiency,[13–15,17] pregnancy,[14,16] diseases causing bone fragility (rheumatoid arthritis,[17] chronic kidney disease,[18] celiac disease,[19] and others),

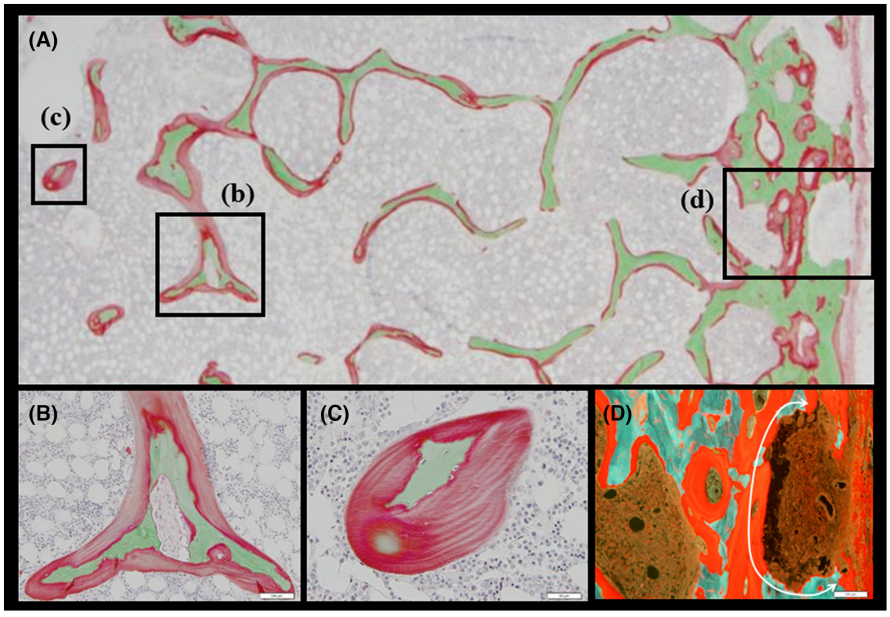

FIGURE 2 Nondecalcified specimens of iliac bone. Pathological findings showed a remarkable increase in the osteoid colored pink. Enhanced bone resorption was also observed in the cancellous (B, C) and cortical bones (D). Resorption image that passes through the trabecula in a tunnel shape (black square line). The island bone of osteoid (black square line). White arrow shows periosteal resorption (black square line). *Goldner stain (natural light); ((A)×30, (B)×150, (C)×300). # Villanueva bone stain (fluorescence); ((D)×150).

TABLE 1 Bone morphometry data of the iliac bone.

Parameters	Abbreviation[23]	Unit	Value	Normal value (Ages 65–74)[24]
Bone volume	BV/TV	%	15.57	19.56 ± 5.62
Trabecular thickness	Tb.Th	Mm	109.37	131.3 ± 28.1
Osteoid volume (tissue)	OV/TV	%	8.54	0.36 ± 0.31
Osteoid volume (bone)	OV/BV	%	54.87	1.20 ± 0.87
Osteoid surface	OS/BS	%	86.12	14.0 ± 6.64
Osteoid thickness	O.Th	Mm	34.74	8.31 ± 1.99
Osteoblast number	N.Ob/BS	N/mm	3.11	—
Eroded surface	ES/BS	%	11.72	3.66 ± 1.69
Multinucleated osteoclasts	N.Mu.Oc/BS	N/mm	0.72	—
Osteoclast surface	Oc.S/BS	%	3.63	0.59 ± 0.73
Fibrotic bone marrow volume	Fb.V/TV	%	0	0

corticosteroid use.[17] Johansson et al. reported that low BMI for women was a risk factor for hip and all osteoporotic fractures,[21] with more than half of these cases having involved lean women. We checked the X-rays presented in these papers, and no case of basicervical femoral neck fractures was noted; therefore, we inferred simultaneous bilateral fracture may not occur unless the bone fragility is severe, as in osteomalacia.

The clinical characteristics of osteomalacia include low bone density, bone deformity, pain, muscle weakness, and adverse imaging findings. However, these are nonspecific symptoms that can also be observed in other diseases besides osteomalacia. The diagnostic criteria for osteomalacia have been proposed by Fukumoto.[3] A definitive diagnosis of osteomalacia necessitates several investigations, including blood tests, imaging (e.g., X-ray and bone scintigraphy), and pathological tests (including biopsy). Serum levels of Ca, P, and ALP are essential for a diagnosis of osteomalacia; however, these levels tend to fluctuate depending on factors such as sample collection time and the patient's immediately preceding meal. Reports have also shown that approximately 20% of patients with osteomalacia had no biochemical abnormalities.[4,22] Thus, these parameters may not always be abnormal, which may lead to overlooking the diagnosis of osteomalacia at the time of routine biochemistry.[22]

Given that osteomalacia can be overlooked, bone biopsy remains important. However, this method for diagnosing osteomalacia is invasive, and obtaining a bone biopsy during surgery may be more convenient. While our patient was diagnosed with osteomalacia based on OV/BV > 10% and O.Th > 12.5 μm,[20] other reports have used different bone morphometry data as the diagnostic criteria.[4–6] This may also be a factor that makes range in the prevalence of osteomalacia. Although our patient had no history of trauma or neoplastic lesion and no abnormal bone morphology, bone morphometry of the bone biopsy allowed a definitive diagnosis of osteomalacia.

4 | CONCLUSION

Vitamin D deficiency osteomalacia due to biased nutrition and excessive dieting caused bone fragility, leading to bilateral basicervical femoral neck fractures. As this condition can be overlooked by clinicians, it is necessary for careful evaluation in thin patients to avoid developing insufficiency fractures by performing a comprehensive work-up of insufficiency fractures, including a biopsy.

AUTHOR CONTRIBUTIONS

Kei Yokogawa: Data curation; formal analysis; methodology; writing – original draft. **Keita Nagira:** Conceptualization; formal analysis; writing – original draft; writing – review and editing. **Toru Yonei:** Data curation; investigation; supervision. **Tetsuya Otsuka:** Investigation; methodology. **Hiroshi Hagino:** Formal analysis; resources; supervision; writing – review and editing. **Hideki Nagashima:** Resources; supervision.

ACKNOWLEDGMENTS

We thank Dr. R. Kouno and Dr. K. Yamamoto for their support on tissue collection techniques and histological assessment.

CONFLICT OF INTEREST STATEMENT

The authors do not have any conflicts of interest to disclose.

DATA AVAILABILITY STATEMENT

The data are not available for public access because of patient privacy concerns, but are available from the corresponding author on reasonable request.

CONSENT STATEMENT

Written informed consent was obtained from the patient for the publication of this case report and the attached images.

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How to cite this article: Yokogawa K, Nagira K, Yonei T, Otsuka T, Hagino H, Nagashima H. Simultaneous bilateral basicervical femoral neck fractures in a patient with osteomalacia: A case report and literature review. *Clin Case Rep*. 2023;11:e7881. doi:10.1002/ccr3.7881