Case Report and Literature Review of Periprosthetic Atypical Femoral Fractures After Total Hip Arthroplasty

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Abstract: We describe a case of periprosthetic femoral fracture with 5 major features of an atypical femoral fracture (AFF) and localized cortical thickening at the fracture site, which is characteristic of an AFF. An 81-year-old female patient had undergone cementless total hip arthroplasty for a right femoral neck fracture at the age of 66, and had been taking oral alendronate since then. At the age of 79, she developed spontaneous right thigh pain. Radiographs showed lateral cortical thickening and pedestal formation around the end of the femoral component. She was advised to discontinue oral alendronate and change to eldecalcitol. At the age of 81, she developed sudden severe pain when standing up from a seated position and was not able to walk. Radiographs showed a periprosthetic femoral fracture with 5 major features of AFF at the site of localized cortical thickening. We diagnosed a Vancouver type B1 periprosthetic femoral fracture. She underwent open reduction and internal fixation (ORIF) with an NCB® Periprosthetic Femur Plate System with cable grips. Daily subcutaneous injection of teriparatide and low intensity pulsed ultrasound therapy were performed to stimulate bone healing. She was able to walk without assistance at 4 months after ORIF. Radiographs showed adequate bridging callus and a disappearing fracture line. This case was diagnosed as a periprosthetic atypical femoral fracture (PAFF), because a periprosthetic fracture is excluded from the definition of AFF. Similar to AFF, PAFF exhibits poor clinical outcomes. The approach to treating PAFF should be decided after considering the pathogenesis.

Keywords: torque, total hip arthroplasty, stress fracture, teriparatide, low intensity pulsed ultrasound.

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typically exhibits poor clinical outcomes, and, because its pathogenesis remains unclear, there is no consensus regarding treatment. We review the reported cases of PAFF and discuss its pathological mechanism through our case and the available literature.

Case report

An 81-year-old female patient had undergone cementless total hip arthroplasty (THA) at another hospital for a right femoral neck fracture at the age of 66, and had been taking oral alendronate since then. Neither steroids nor proton pump inhibitor had been used in this patient. At the age of 79, she developed spontaneous right thigh pain and used a cane to walk. Radiographs of her right thigh showed lateral cortical thickening and pedestal formation around the end of the femoral component in her right femur (Fig. 1). She was advised by a doctor to discontinue oral alendronate and change to eldecalcitol for osteoporosis. At the age of 81, she developed sudden severe pain in her right thigh when standing up from a seated position, and was not able to walk. She was taken to the emergency room of our institution by ambulance.

Her right leg was shortened and rotated externally. Her body height and weight were 142.7 cm and 41.0 kg, respectively (body mass index, 20.1 kg/m²). The complete blood count and levels of C-reactive protein and serum calcium and phosphate were within normal range. There were no findings of infection on blood testing. The serum level of tartrate-resistant acid phosphatase-5b (TRACP-5b) was also within normal range (370 mU/dl, normal range: 120–420), but the total pro-collagen type 1 intact N-terminal propeptide (P1NP) level was high (161 ng/l, normal range: 26.4–98.2). Dual energy X-ray absorptiometry T-scores in the 1st–4th lumbar vertebrae and left femoral neck were −2.0 and −2.7, respectively. Radiographs showed a peri-prosthetic femoral fracture with 5 major features of AFF at a site of localized cortical thickening (Fig. 2). There was no apparent loosening of the femoral component on plane radiographs or computed tomography. We diagnosed a Vancouver type B1 periprosthetic femoral fracture.

The patient underwent open reduction and internal fixation (ORIF) with an NCB® Periprosthetic Femur Plate System (Zimmer, Warsaw, IN, USA) with cable grips on the day after the injury (Fig. 3). No bone graft was used. Wheelchair use and non-weight-bearing activity were advised for 3 weeks, and standing or gait exercise using crutches with partial weight-bearing began 3 weeks after surgery. Gait exercise using a walker without weight-bearing limitations began 6 weeks after surgery. Daily subcutaneous injection of teriparatide (Forteo®, Eli Lilly Japan, Kobe) for 6 months
and low intensity pulsed ultrasound (LIPUS) therapy (SAFHS® 2000J ; TEIJIN, Tokyo) were performed from the first week after ORIF to stimulate bone healing. The patient was able to walk without assistance or thigh pain at 4 months after ORIF. Radiographs of the right femur showed adequate bridging callus and a blurred fracture line (Fig. 4). We evaluated the status of the bone union by Mukundan’s method and judged it as bone union at 4 months after ORIF [2].

**Discussion**

We presented a case of periprosthetic femoral fracture with 5 major features meeting the AFF criteria. The patient suffered from the fracture when standing up from a seated position, and radiographs showed localized cortical thickening at the fracture site, which is characteristic of stress fractures. This case was diagnosed as PAFF because periprosthetic fractures are excluded from the definition of AFF. We performed combined therapy with parathyroid hormone (PTH) and LIPUS after ORIF using a locking plate, screws, and cable. Radiographs showed adequate bridging
callus and disappearing fracture line at 4 months post-operatively, and the patient was able to walk without assistance or thigh pain.

Similar cases have been reported (Table 1) [3–12]. All were females and all had a history of long-term bisphosphonate therapy. Six of 14 PAFF cases were initially treated by surgery, and 8 cases were treated by conservative therapy. During follow-up, treatment in 3 of the 8 cases of conservative therapy converted to surgery due to progression from incomplete to complete fractures. Robinson et al reported that the average time to union in patients with PAFF was 8 months, and the incidence of complications (such as pneumonia, myocardial infarction, pulmonary embolism, wound infection, and death) was 25% [13]. The factors leading to a high-complication rate in PAFF cases are older age at the time of fracture, longer operation time, and longer period of bone union than in AFF cases. PAFF also exhibits poor clinical outcomes, similar to AFF, and treatment should be decided after considering the pathophysiology.

The pathogenesis of PAFF remains unclear. The current hypothesis of the cause of PAFF includes a combination of the effects of suppressed bone turnover and repetitive stress around the stem. The bone metabolism markers of our patient were normal, and there was no evidence of suppressed bone turnover. It is possible, however, that bone turnover was extremely suppressed due to taking alendronate for 13 years. Almost all previous PAFF cases had a history of long-term bisphosphonate therapy and prodromal symptoms, and their radiographs showed localized cortical thickening at the fracture site, which is characteristic of stress fractures (Table 1). Bottai et al showed an accumulation of micro-damage at the distal end of the stem in a histological study of PAFF [14], suggesting that repetitive load during activities of daily living is concentrated on the lateral cortical bone around the end of the stem. Although it is possible that the shape of the femur [15], stem alignment [16], and stem instability [17, 18] are factors in PAFF, the femoral shape in the present case was normal and the varus angulation of the stem was slight. There was also no evidence of stem instability, because spot welds were seen at femoral zones 2 and 6. In addition, this case met the criteria of osteoporosis, which is a risk factor of periprosthetic insufficiency fracture [19], but this fracture had the characteristics of stress fracture, and we considered it not to be an insufficiency fracture.

We should consider what kind of repetitive stress occurs around the distal end of the stem. Robinson et al reported that PAFF patients usually complain of

| Age (y) | Gender | BP Type | BP duration before fx (y) | Treatment of fx | Prodromal symptom | Cortical thickening | AO classification | Reference |
|---------|--------|---------|--------------------------|-----------------|-------------------|-------------------|------------------|----------|
| 78      | F      | ALN     | 9                        | Conservative → ORIF | (+)               | (+)               | 32-A3            | (2)      |
| 79      | F      | ALN → IBAN | 10                        | Conservative → ORIF and Revision | (+) | (+) | 32-A3 | (3)      |
| 52      | F      | ALN     | 5                        | Conservative    | (+)               | (−)               | Incomplete fx    | (4)      |
| 85      | F      | RIS     | Many years               | Conservative    | (+)               | (+)               | Incomplete fx    | (4)      |
| 79      | F      | ALN     | >9                       | Conservative    | (+)               | (+)               | Incomplete fx    | (4)      |
| 69      | F      | IBAN    | 4                        | ORIF → non-union | (+)               | (−)               | 32-A3            | (5)      |
| 72      | F      | ALN     | 10                       | Conservative    | (+)               | (−)               | Incomplete fx    | (6)      |
| 81      | F      | ALN     | >12                      | Conservative    | (+)               | (+)               | Incomplete fx    | (7)      |
| 43      | F      | RIS     | 5.5                      | ORIF → non-union | (+)               | (+)               | 32-A3            | (8)      |
| 74      | F      | RIS     | 6                        | ORIF            | (+)               | (+)               | 32-A3            | (8)      |
| 86      | F      | ALN     | 9.3                      | ORIF            | (−)               | (+)               | 32-A3            | (8)      |
| 82      | F      | ALN     | 3                        | ORIF → non-union | (−)               | (+)               | 32-A3            | (9)      |
| 69      | F      | ALN     | >7                       | ORIF            | (+)               | (+)               | 32-A3            | (10)     |
| 68      | F      | RIS     | 7                        | ORIF            | (+)               | (+)               | 32-A3            | (11)     |

BP: bisphosphonate; F: female, ALN: alendronate, IBAN: ibandronate; RIS: risedronate, fx: fracture, ORIF: open reduction and internal fixation.
dull pain or discomfort on weight-bearing and experience pain when standing up from a seated position [13]. In the present case, the patient could not walk after experiencing severe pain when standing up from a seated position. Based on this, we hypothesized that the pathogenesis of PAFF was a stress fracture due to torque on the diaphysis of the femur (Fig. 5). In general, the torque is generated in the femur on weight-bearing [20], and the torque increases as the hip flexion angle increases [21]. Abdullah et al demonstrated the ability to predict damage formation in hip arthroplasties by finite element analysis using computed tomography images and showed that the damage in torsion configuration is initiated at the distal end of the prosthesis stem [22]. The lateral cortical hypertrophy seen on the radiograph at the tip of the femoral component may have been a reaction to repetitive stress. Moreover, the configuration of all the complete fractures in the PAFF cases reviewed was not spiral (AO classification; 32-A1), but transverse (32-A3) (Table 1). It was suggested that PAFF possibly occurs as a result of shear failure due to torque to the femoral diaphysis. The fracture configuration would be transverse if isometric loading was due to torsion, and spiral if the isometric loading was due to lateral bending [22]. We suggest that a possible cause of PAFF is cumulative micro-damage at the distal end of the prosthesis stem due to increased torque during weight-bearing and standing up from a seated position.

We should carry out PAFF treatment after considering the pathogenesis as follows: 1) suppressed bone turnover due to bisphosphonate, 2) micro-damage accumulation due to repetitive stress at distal end of stem, and 3) torque to the femoral diaphysis (Fig. 5). The time to bone union in this case was 4 months, although previous reports showed that there was a high risk of delayed union or non-union in PAFF [13]. Watts et al reported that they did not observe a consistent effect of teriparatide on healing in an AFF previously treated with bisphosphonates, although we used teriparatide treatment in our case [23]. Warden et al reported that PTH treatment combined with LIPUS was more effec-

**Pathophysiology of PAFF**

1) Suppressed bone turnover due to bisphosphonate (BP)

2) Micro-damage accumulation due to repetitive stress at distal end of stem

3) Torque to diaphysis of femoral

**PAFF treatment after considering the pathophysiology**

1) Suppressed bone turnover

2) Micro-damage accumulation

3) Torque to diaphysis of femoral

BP discontinuation, PTH, LIPUS

Strong rotational fixation

Stem revision as necessary

Excellent outcome

Fig. 5. Pathophysiology of periprosthetic atypical femoral fractures and the treatment after considering the pathophysiology. PAFF: periprosthetic atypical femoral fracture, PTH: parathyroid hormone, LIPUS: low intensity pulsed ultrasound.
tive for fracture healing in rats [24]. In fact, Wakayama et al reported that bone union was confirmed at 4 months after surgery using combined therapy of PTH and LIPUS [12]. Thus, combined therapy with PTH and LIPUS may promote fracture healing in PAFF.

Another important issue is to achieve a strong rotational fixation. The use of intramedullary nails is recommended in order to obtain rotational stability, but this was not an option in our case due to the presence of the stem, so plate fixation had to be performed instead. Regarding the locking plate fixation, the combined use of the screw and cable can obtain a more suitable rotational stability than the use of cable alone [25]. Therefore, we believe that adequate rotational stability was one of the factors for the success of this case. If the situation could not have been solved by the above method, we would have had to consider bone graft and stem revision as well.

**Conclusion**

We described a case of periprosthetic femoral fracture with 5 major features of AFF and localized cortical thickening at the fracture site. Since PAFF also exhibits poor clinical outcomes, similarly to AFF, we should decide treatment after considering the pathophysiology.

**Conflict of interest**

The authors declare that they have no conflict of interest.

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人工股関節全置換術後に非定型大腿骨ステム周囲骨折を生じた1例

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要 旨：2014年にShaneらが提唱した非定型大腿骨骨折（AFF, atypical femoral fracture）診断基準の5つの大項目とAFFの特徴である大腿骨外側皮質骨肥厚を認めた大腿骨ステム周囲骨折の1例を報告する。症例は81歳女性。66歳時に右大腿骨頸部骨折に対し人工股関節全置換術を施行し以後、ビスホスホネートを内服していた。79歳の時に右大腿部痛が出現し、両股関節単純X線正面像で、右大腿骨外側に限局する皮質骨肥厚とpedestal formationを認め、ビスホスホネートを中止し、ビタミンDの内服を開始した。81歳の時、立ち上がった際に激しい右大腿部痛があり歩行不能となった。単純X線像で、AFF診断基準の5つの大項目を満たす右大腿骨ステム周囲骨折を右大腿骨外側皮質骨肥厚部に認めた。骨折型はインプラントのゆるみを伴わない骨折（Vancouver分類type B1）であり、骨接合術を施行した上で、骨癒合促進のために、術後から副甲状腺ホルモン製剤テリパラチドと低出力超音波治療器の使用を開始した。術後4ヶ月で骨癒合を認め、独歩可能となった。大腿骨ステム周囲骨折はAFF診断の除外項目に該当するため、本骨折を非定型大腿骨ステム周囲骨折（PAFF, periprosthetic atypical femoral fracture）と診断をした。PAFFの臨床転帰はAFFと同様に不良であり、PAFFの治療に臨む際には、その病態を考慮した上で方針を決定すべきである。

キーワード：トルク、人工股関節全置換術、疲労骨折、テリパラチド、低出力超音波治療器。

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