Dear Editor,

Osteoporosis is a growing problem in our ageing population. With increasing awareness of osteoporosis, it can be diagnosed and treated with first-line drugs such as bisphosphonates. However, there have been increasing reports of atypical fractures resulting from prolonged bisphosphonate therapy. These have largely been described in the subtrochanteric region of the femur and found in the upper extremity. We report a case of bilateral atypical fractures of the ulna in a patient treated with bisphosphonates for 10 years.

Case Report

Our patient was an 89-year-old Chinese female who was ambulant with a walking frame. She had a past medical history of osteoporosis treated with alendronate 70 mg once a week from 2004 to 2011. After a drug holiday of 1 year, she was restarted on alendronate from 2012 to 2014. She also had hypothyroidism (for which she was on thyroxine), hyperlipidaemia and quiescent gout, and was on omeprazole for the last 3 years. She had no other fractures of note in the past and was compliant with her bisphosphonate therapy with improving T-scores of -3.7 in 2007 to -3.5 in 2012. She was admitted to the hospital following a trivial fall onto her buttocks, without any direct trauma to her upper limbs. She did not use her upper limbs to brace the fall and could stand up after. She complained of left upper limb pain and was found to have an atypical ulnar fracture. The initial radiographs (Fig. 1) showed an undisplaced transverse fracture of the ulna with a thickened cortex at the fracture site. The fracture was termed an atypical fracture due to the trivial nature of the fall and relative thickening of the diaphysis of the ulna bone in comparison to the cortex of the radius and the characteristics of the fracture. Blood tests revealed a normal calcium and vitamin D panel. Due to the undisplaced nature of the fracture, she was treated in a cast. Bone mineral density scans were performed and showed osteoporosis with a T-score of -3.1 in both the lumbar spine as well as the proximal femur. Two weeks later, a repeat radiograph was done which showed increasing angulation of the fracture. She was offered surgery at that point but declined and elected to be treated in a cast and be followed up in the clinic. Screening radiographs (Fig. 2) were taken of the contralateral limb which showed an incomplete right ulnar fracture. Further x-rays of her left ulnar fracture showed increasing angulation and minimal callus formation (Fig. 3). She was re-offered surgical treatment at various

Fig. 1. Initial radiographs at the time of injury showing a transverse fracture with thickened cortex. The fracture was at the side on tension in the ulna and at the apex of the curvature.

Fig. 2. Radiographs of the right forearm and bilateral femur showing an incomplete ulnar fracture with thickening of the cortex.
points of her follow-up but consistently declined and was treated with a splint. Subsequent radiographs showed atrophic non-union with minimal callus formation up to 1-year of follow-up (Fig. 4).

Discussion

Long-term bisphosphonate-related atypical fractures are mostly reported in the subtrochanteric and diaphyseal regions of the femur. Recent reports have emerged showing an association with upper extremity fractures. Similar to our study, previous case reports also showed fractures that were characteristic of atypical fractures – non-comminuted, transverse fractures with localised cortical thickening. These fractures were also related to the use of a walking aid on the side of the fracture.

It is theorised that prolonged bisphosphonate therapy suppresses bone remodelling, allowing microfractures to persist and propagate, resulting in stress fractures. Common radiological findings of atypical fractures, as described by the American Society for Bone and Mineral Research, consist of major and minor features. However, these are described for atypical femur fractures. No such classification has been described in atypical upper limb fractures but we observe similar features such as a transverse fracture configuration that is non-comminuted with a thickened cortex or localised periosteal reaction. This was also observed in our patient in her right femur and bilateral ulna fractures.

Atypical upper extremity fractures are rare and largely associated with the use of walking aids for ambulation. We believe that the increased weight bearing requirements shared by the upper limb bones during ambulation with walking aids cause this phenomenon. When using a walking aid, the dorsal aspect of the ulnar undergoes forces in tension (Fig. 5). This is similar to atypical femoral fractures where failure occurs on the tension side. After repetitive stress, there is microscopic failure of the bone in tension, causing microfractures and a localised periosteal reaction. These cause further weakening of the bone and finally, macroscopic failure. This is in keeping with our patient.
who used both her upper limbs to push her walking frame in order to stand as well as ambulate.

With regard to the treatment of non-atypical fracture in patients already on long-term bisphosphonates, the current literature reports a delayed time to fracture healing but no difference in the incidence of non-union.11,12 This is in contrast to atypical bisphosphonate-related fractures where many authors report delayed healing and a non-union rate of up to 26%.13

Conclusion

This case reinforced the need for awareness of atypical fractures in the setting of long-term bisphosphonate therapy. Physicians treating the elderly for osteoporosis should have a high index of suspicion for patients complaining of persistent limb pains, especially looking out for upper extremity atypical fractures in the setting of ambulation with a walking aid. Orthopaedic surgeons treating the patient with an atypical upper extremity fracture will need to factor increased rates of non-union into their treatment algorithm to effectively treat the osteoporotic elderly.

REFERENCES