

Case Report

Extensive Bilateral Patellar Tendon Ossification in a Spinal Cord Injury Patient

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This is the first report of extensive bilateral patellar tendon ossification occurring over a prolonged time after a unilateral knee injury. An 84-year-old Japanese man with a spinal cord injury caused by a burst fracture of the T12 vertebra presented with a bony hard prominence on the left knee, which was injured in a traffic accident when he was 77 years old. Radiography revealed extensive ossification of the bilateral patellar tendons. We review the English literature with a focus on the localization of bilateral heterotopic ossification of the knee in patients who had a central nervous system injury.

Key words: heterotopic ossification, patellar tendon, spinal cord injury, bilateral, knee injury

Heterotopic ossification (HO) is defined as the deposition of bone within soft tissues that do not normally undergo ossification. HO is classified into three types: traumatic HO, neurogenic HO (occurring after traumatic brain injury [TBI], spinal cord injury [SCI], anoxia, or encephalitis), and a rare form of congenital HO [1,2]. HO of the knee is a relatively uncommon condition in patients with an SCI [3,4]. Ossification of the patellar tendon is rare, and has been reported mainly in patients with knee injuries [5,6] or as a consequence of patellectomy [7], intramedullary nailing of the tibia [8], and anterior cruciate ligament reconstruction [9].

To our knowledge, there have been no reports of patients with both an SCI and bilateral ossification of the patellar tendon in the absence of surgical intervention occurring over a prolonged time after a unilateral knee injury. Here we report the case of a patient with

extensive ossification of the bilateral patellar tendons, and we review the English literature regarding the localization of bilateral ossification of the knee in patients with a central nervous system (CNS) injury.

Case Report

An 84-year-old Japanese man presented with a bony hard prominence on his left knee. He had flaccid paresis of both legs (grade A according to the American Spinal Injury Association impairment scale), due to an SCI caused by a burst fracture of the T12 vertebra at 46 years of age. At the age of 77 years, he suffered a contusion of the left knee in a traffic accident, but he did not receive any specific treatment. After the swelling of the left knee subsided, he noticed a gradually enlarging bony prominence over the left patellar tendon. He did not recall any episodes of right knee trauma, and did not notice ossification of the right patellar tendon.

On examination, the bilateral patellar tendons were bony and hard, and completely fused to the tibial tuberosity (Fig.1). The right patellar tendon did not have a bony prominence similar to that identified on the left patellar tendon. In both knees, the range of flexion was 20°-130°, and thus there was no shortening of either patellar tendon associated with ossification. There were no signs of inflammation or ligamentous instability. There was no marked joint contracture at the hips, ankles, or toes.

Radiography and three-dimensional computed tomography revealed extensive ossification of the bilateral patellar tendons, with complete fusion of the inferior pole of the patella to the tibial tuberosity (Figs. 2, 3). The ossification was more extensive on the left side, but the location of the ossification was similar in both knees, with the ossified patellar tendon being hypertrophic and protruding beyond the normal border of the tendon over the anterior patellar surface and tibial tuberosity. The position of the patella was normal, and



Fig. 1 Appearance of both knees with the patient in a sitting position on a bed. There is a bony hard prominence on the left knee.

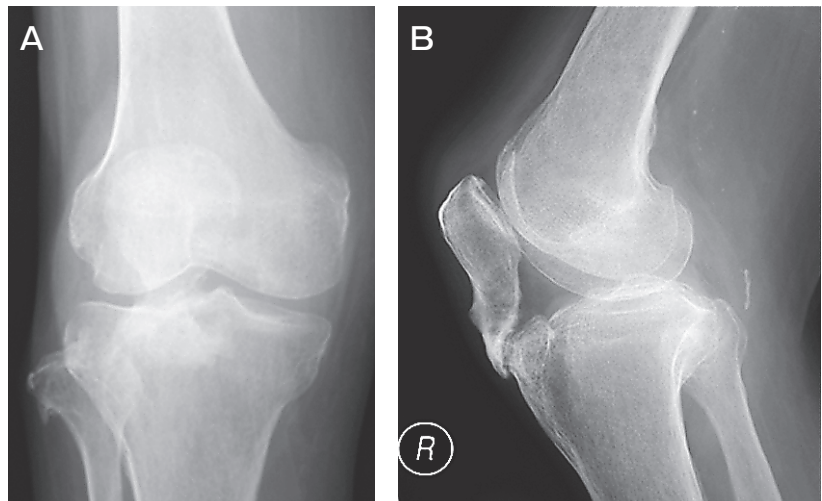


Fig. 2 Anteroposterior (A) and lateral (B) radiographs of the patient's right knee show that ossification has led to complete fusion of the inferior pole of the patella with the tibial tuberosity.

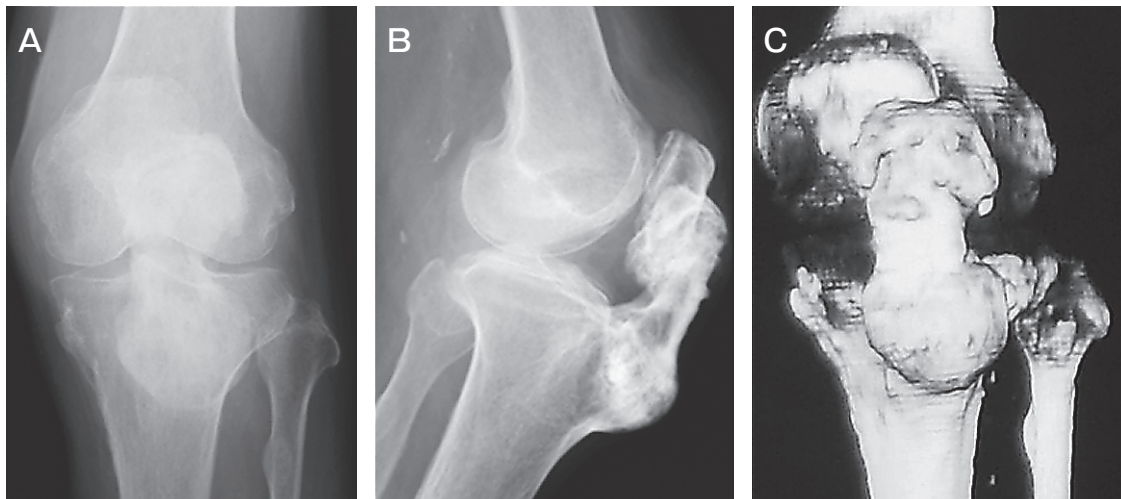


Fig. 3 Anteroposterior (A) and lateral (B) radiographs of the left knee and 3D CT (C) show that ossification is localized to the lateral two-thirds of the patellar tendon.

the patient did not have patella alta. Complete blood count, biochemical tests including calcium, phosphate, and alkaline phosphatase, and chest radiography demonstrated no abnormalities.

Among the patient's other joints, the left shoulder showed calcific tendinitis with a small calcific deposit at the subacromial bursa. Surgery was not performed for either knee because the ossification caused little functional disability. Informed consent was obtained from the patient for the publication of this study.

Discussion

This case demonstrates two important clinical lessons. First, extensive ossification of the patellar tendon in patients with an SCI is extremely rare. To our knowledge, this is the first report of extensive bilateral patellar tendon ossification occurring over a prolonged time after unilateral knee injury. Second, the interesting aspect of our patient's case is the development of HO of the contralateral patellar tendon following the occurrence of unilateral traumatic patellar tendon HO.

Clinically significant neurogenic HO develops in approx. 20% of patients with an SCI. It always occurs below the level of the lesion [3, 4] and is detected from a mean of 12 weeks after injury [10]. The hip was the joint most frequently involved, accounting for 52.2%,

followed by the shoulder (24.4%), the knee (12.2%), and the elbow (8.5%) [11]. A case-control study of SCI patients revealed that neurogenic HO was more common in the patients with complete SCI, rostral injuries, or associated thoracic trauma [12]. On the other hand, traumatic HO differs from neurogenic HO with respect to its location and other aspects. It usually commences at the time of injury and may be radiographically evident by 3 weeks post-injury [4]. In our patient, we speculated that the left patellar tendon represented traumatic HO, and the right patellar tendon involvement was due to subsequent traumatic HO with preexisting neurogenic HO.

This case report demonstrates that patients with SCI can develop extensive bilateral patellar ossification. In a retrospective study, Seipel *et al.* evaluated 1,463 patients in a rehabilitation setting, with most patients suffering from neurological disorders including SCI and TBI [11]. They reported that 13 of 17 patients with multiple manifestations of HO presented with bilateral symmetric involvement of corresponding joint regions, and one patient had involvement of both knees. Coelho *et al.* reported that HO was bilateral in 14 of 33 patients with HO after a traumatic SCI. The knee joint was affected in 6 patients, but HO was bilateral in only one of them (1/6; 16.7%) [13]. Fuller *et al.* retrospectively reviewed 17 consecutive patients with CNS injuries who had

Table 1 Brief summary of bilateral HO of the knee in patients with central nervous system injury

Authors	Age (years), gender	Neurologic condition	Medical information, other sites of HO	Site(s) of HO of the knee	Treatment
Fuller <i>et al.</i> [14]	32, M	TBI, tetraplegia	HO: bilateral shoulders, elbows, hips	bilateral, anteromedial	resection
Fuller <i>et al.</i> [14]	24, M	TBI, tetraplegia	R tibia, intramedullary nail	bilateral, anterior	resection
Fuller <i>et al.</i> [14]	41, M	TBI, hemiplegia	R patella, ORIF	R, anterior L, anteromedial	resection
Fuller <i>et al.</i> [14]	36, M	anoxia, tetraplegia	HO: left hip, left elbow	bilateral, anterior	resection
Fuller <i>et al.</i> [14]	19, F	TBI, hemiplegia	None	R, anteromedial L, anterior	resection
Zhang <i>et al.</i> [16]	47, M	viral encephalitis	Coma for 30 days, HO: bilateral hips	bilateral, medial	resection
Saito <i>et al.</i> [17]	26, F	viral encephalitis	HO was observed at 25 days after coma	bilateral, medial	resection
Sugita <i>et al.</i> [18]	42, M	sedation	Intubation with anesthesia for 44 days, Infection after cardiac surgery	bilateral, medial	resection
Ramin <i>et al.</i> [19]	39, M	SCI, paraplegia	HO was observed at 6 months after injury	bilateral, popliteal fossa	resection
Our case	84, M	SCI, paraplegia	L knee, local injury	bilateral, patellar tendon	conservative treatment

*HO, heterotopic ossification; F, female; M, male; R, right; L, left; SCI, spinal cord injury; TBI, traumatic brain injury; ORIF, open reduction and internal fixation.

undergone excision of HO (22 knees, five bilateral [5/22; 22.7%]) of the knees [14]. In a survey of 570 surgeries of HO after CNS injury, HO in 129 knees occurred in patients with SCI (6 bilateral, 6/129; 4.6%) [15].

We reviewed the English literature with a focus on the localization of bilateral HO of the knee in patients who had a CNS injury. We identified only nine reported cases of bilateral HO of the knee in patients with a CNS injury in which HO was clearly described and defined [14, 16-19] (Table 1). These cases of bilateral HO of the knee are presumably thought to occur during almost the same period. In each patient, the site of HO was very similar and symmetrical in both knees, although HO occurred at various locations (mainly on the anteromedial aspect of the knee [4], which accounted for 54.7% of the HO in the patients with a CNS injury and could be explained by particular locations having worse repercussions [15]). Our literature review thus suggests that bilateral HO of the knee in patients with CNS injuries affects a similar site on both sides.

The pathogenesis of HO is incompletely understood. Bidner *et al.* proposed that a failure of the regulation of the immune system, CNS, or the inflammatory response leads to the release of agents that induce ossification, resulting in the development of HO [20]. Chalmers *et al.* studied the inducing capacity of different tissues for HO [21]. He reported that three conditions were required for the development of HO in soft tissues: (1) an inducing agent, (2) osteogenic precursor cells, and (3) an environment permissive for osteogenesis. When these conditions are met, mesenchymal stem cells are recruited and proliferate to differentiate into chondrocytes or osteoblasts, ultimately leading to the development of HO [22].

The cases of patients with an SCI involving HO are classified into two groups based on the clinical course and the radiographic appearance of the lesions [4]. One group, accounting for approx. 10% of the patients, shows moderate to severe HO or true bony ankyloses and an ongoing potential for ossification. The other group, accounting for the other approx. 90% of the patients, has minimal to moderate amounts of HO. We hypothesized that the contralateral patellar tendon ossification in our patient might have occurred secondary to the stimulation of mesenchymal stem cell differentiation by circulating factors derived from the original site of extensive patellar tendon ossification. It is also possi-

ble that a long period of immobilization associated with his flaccid paraplegia due to the SCI [23] or similar positioning of both knees in daily life resulted in the development of bilateral HO at similar sites and with similar morphology.

Our patient's case also emphasize the importance of considering the possibility of unilateral or even bilateral HO, including the possibility of a subsequent occurrence of HO on the contralateral side of the patellar tendon after unilateral knee injury in patients with an SCI. Our patient's case may also provide new information about the underlying mechanisms of bilateral HO.

The management of HO is aimed at limiting its progression and maximizing the function of the affected joint. Nonsurgical treatment is appropriate for asymptomatic HO. In our patient's case, there was no pain or contracture of the knee due to sensory deficit or flaccid paresis by the SCI. However, surgical excision should be considered in cases of joint ankylosis, significantly decreased range of motion, nerve or vessel compression, or skin breakdown by HO. Patient selection, timing of the excision, and postoperative prophylaxis are important components of proper management [24].

In summary, we reported the clinical and imaging findings of a patient with an SCI with extensive bilateral ossification of the patellar tendon. In this patient, subsequent HO of the right patellar tendon occurred following traumatic HO of the left patellar tendon, and the site of HO in both knees was very similar.

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