CASE REPORT

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Heterotopic Ossification in Shoulder and Knee as a Rare Complication of Hemiplegia Following Ischemic Stroke

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ABSTRACT Neurogenic heterotopic ossification (HO) is a complication of neurologic disorders which is characterized by formation of new extra osseous in soft tissue surrounding peripheral joints. HO can cause pain, progressive decrease in range of joint motion and a decline in functional capacity. We herein report a 52-year-old female patient with right hemiplegia due to ischemic stroke. She was admitted to the clinic with pain and limitation of movement in her right knee and shoulder for 6 months. Her imaging studies revealed HO. After 8 weeks of inpatient rehabilitation, shoulder range of motion improved but the contracture in the knee joint remained the same. HO is most commonly seen around the hip joint. However, in the presented case, it differs in that it is observed in the knee and shoulder. HO should be considered in the differential diagnosis of pain and joint limitation in patients with hemiplegia.

Keywords: Heterotopic ossification; ischemic stroke; hemiplegia

Heterotopic ossification (HO) is defined as the pathologic ectopic bone formation within the soft tissues. Acquired HO can occur after musculoskeletal trauma or neurogenic injuries such as spinal cord injury, traumatic brain damage, stroke, or cerebral anoxia.[1] HO is a rare condition in stroke patients, with incidences of HO after a stroke having been reported to as 0.5-1.2%.[2] In the early course of the HO, it can cause pain, fever, swelling, redness, and reduced joint mobility in the affected area. Later course of the disease, a decrease in the range of motion (ROM) and ankylosis of the joint may occur.[3] We herein report a rare and different case of neurogenic HO with knee and shoulder joint involvement without hip joint involvement occurring in the hemiplegic patient.

CASE REPORT

The patient was informed and her written approval was received for a case report presentation. A 52-year-old female patient with right hemiplegia due to ischemic stroke was admitted to the clinic with pain and limitation of movement in her right knee and shoulder for 6 months. The patient had suffered from ischemic stroke one year ago and a history of deep vein thrombosis (DVT) one year earlier. The patient had received a rehabilitation program in the first 2 months following the stroke. Regarding functional status, the patient was able to sit without support but was unable to stand. Her ambulation level was 1 according to Functional Ambulation Classification. She was wheelchair-bound. She performed all activities of daily living with the assistance of caregivers. Upon examination at admission, Brunnstrom's motor stage was 2 in the upper extremities, 3-4 in the hand, and 3 in the lower extremities. In the examination of spasticity, Grade 2 elbow flexor spasticity, Grade 2-3 wrist spasticity, Grade 3 pronator spasticity, Grade 2-3 shoulder joint spasticity, Grade 3-4 knee flexor

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spasticity and Grade 3 ankle flexor spasticity were recorded according to Modified Ashworth Scale. There was flexion contracture and a 50 degree range of joint motion in the right knee (between 90 and 140 degrees). Right shoulder movement was painful and passive shoulder movement was restricted, with 110 degrees in flexion and 90 degrees in abduction. There was no swelling or warmth on the right knee and shoulder. Laboratory tests revealed a WBC: count of 9.4 10^3/μL, an ESR of 12 mm/h, a CRP of 4.6 mg/L, ALP of 94 U/L (normal range: 30-120) and serum CPK of 36 U/L (normal range: 0-145). The patient was first evaluated by conventional radiography and computed tomography (CT) for restriction of the right knee and shoulder joints (Figure 1A, B). The findings of the scintigraphic examination related to the right knee were considered to be mature HO (Figure 1C). Indomethacin 75 mg daily for 3 weeks and alendronate 70 mg per week for 12 weeks were commenced and the physical therapy program was initiated. The rehabilitation program basically comprised of ROM exercises (in the pain-free range), strengthening exercises, transfer activities, and gait training. After 8 weeks of inpatient rehabilitation program and medical treatment, the range of shoulder joint motion improved 20-30 in the direction of flexion and abduction but the contracture in the knee joint did not improve. The orthopedics and traumatology department was consulted for further treatment of the right knee contracture. Arthroscopic debridement of the right knee was recommended; however, the patient did not consent to the surgery. At discharge, there was no noticable improvement of functional or ambulatory status of the patient compared to initial presentation. The patient was discharged with alendronate treatment and a home exercise program comprised of ROM (in the pain-free range) and strengthening exercises.

DISCUSSION

HO is a rare condition in patients with hemiplegia following a stroke and can cause pain, a progressive decrease in the range of joint motion, and a decline in functional capacity.[2] In a cross-sectional study conducted in seven rehabilitation centers, Cunha et al. reported that the annual prevalence of HO was 1.3% in stroke patients who admitted to seven rehabilitation centers over 10 years.[4] The main etiological factors implicated in the development of HO are immobilization and forced manipulation; spasticity, fractures, infections, DVT, and pressure ulcers may also contribute.[5] Our case was also a hemiplegic patient with spasticity and she was immobile for one year. Also, she had a history of DVT 9 months ago.

Neurogenic HO tends to occur on the affected side. On the other hand, rarely, HO may occur in hemiplegic patient's nonparetic extremities.[6] The development of HO in the affected extremity may

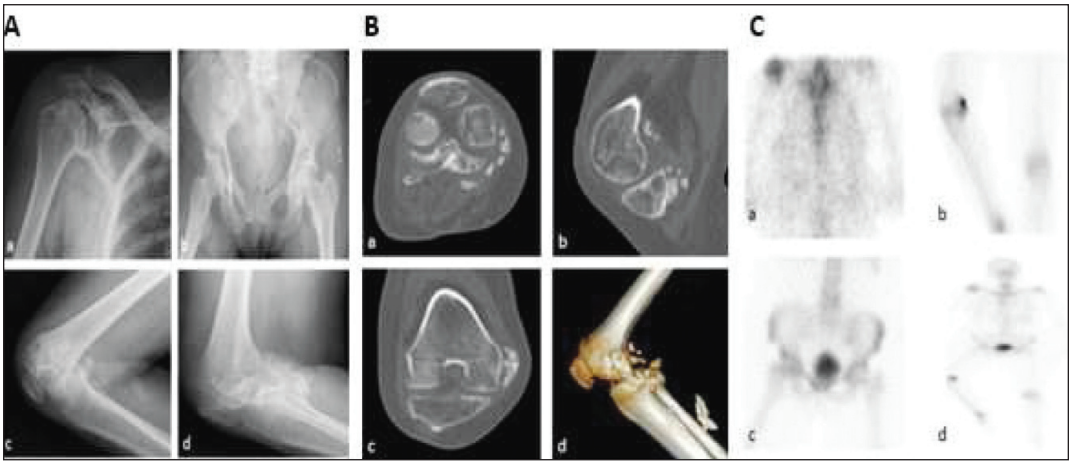

FIGURE 1: A) Plain radiographs of right shoulder (a), hips (b) and right knee (c, d). B) Sagittal and axial images of the right knee on a computed tomography (a, b, c), three-dimensional computed tomography images of the right knee (d). C) Three-phase bone scintigraphy images, right shoulder (a), right knee (b), hips (c), total body (d).

worsen the patient's functional status due to pain and limitation of movement.[7] In our patient, the contracture in the knee joint due to HO was preventing the standing exercises. Neurogenic HO often affects the hip joint. In a study including 61 stroke patients, Genêt et al. reported that HO most often affects the hip joint (76.6%), then elbow joint (11.1%), knee joint (11.1%), shoulder joint (1.2%), respectively.[1] In our case, unexpectedly, HO was detected around the right knee and shoulder without involvement hip joint. Poor positioning, some traumatic factors, and aggressive passive ROM exercises may have been contributed to the HO development. Also, spasticity and DVT may also be responsible for HO formation in the right knee joint.

In hemiplegic patients, the most important target must be to prevent the development of HO but if HO is detected, passive ROM exercises are recommended for preventing the progression of HO.[8] Furthermore, nonsteroidal anti-inflammatory drugs (NSAIDs) and bisphosphonates can be used for preventing HO.[5,8] NSAIDs such as indomethacin, celecoxib and meloxicam are especially effective in the early stages of HO development.[9] Usually, 75 mg per day of indomethacin for 3 weeks is the recommended.[10] Once bone deposition has occurred, the effectiveness of the NSAID decreases, so surgical resection may be required for this stage of HO.[8] Patients who are eligible for surgery are those without pain and swelling in the joints, with normal serum ALP levels and mature HO, based on a scintigraphy.[3] When our patient was admitted to our clinic, she had complaints for 6 months and there was a limited ROM in the knee and shoulder joints. After 8 weeks of inpatient rehabilitation and indomethacin and alendronate medical treatment, the range of shoulder joint motion improved 20-30 in the direction of flexion and abduction but there was no change in the knee joint.

A diagnosis of HO begins with the physical examination and conventional radiography is the most frequently used initial imaging method in patients suspected to have HO. Typically, conventional radiographic and CT findings are characterized by calcification in the periphery of the areas of HO and the relatively radiolucent appearance at the center.[11] Also, ultrasonography and three-phase bone scintigraphy are useful methods for the early diagnosis of HO.[8] In this case, we have used direct radiography, CT to detect HO and three-phase bone scintigraphy to evaluate maturity of HO.

In light of the case presented, HO should be considered in the differential diagnosis in patients with hemiplegia who suffers from pain and limitation of joint movement. In hemiplegic patients, HO is most frequently reported in the hip joint, but it can also occur in other joints without involving the hip joint as in this case report.

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: *Ali Karakaş, Onur Engin, Banu Dilek, Özlem El;* ***Control/Supervision:*** *Banu Dilek, Özlem El;* ***Literature Review:*** *Ali Karakaş, Onur Engin;* ***Writing the Article:*** *Ali Karakaş, Onur Engin;* ***Critical Review:*** *Banu Dilek, Özlem El;* ***References:*** *Ali Karakaş, Onur Engin.*

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