Case Report

A case report of heterotopic ossification as rare complication in both affected and unaffected side of hemiplegia following stroke

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ABSTRACT

Background: New bone formation in the periarticular regions of large joints is the main character of Heterotopic ossification (HO), most commonly seen in or after spinal cord injury, traumatic injury to brain, burn. It is rare as the complication incidence is less than 1% or less.

Case Reports: This study reports an unusual presentations of HO in a 50 year-old male with right hemiplegia due to subarachnoid bleeding developed HO on all the major joints bilaterally both in effected and non effected side after 7 months. In this case spasticity around hip flexors, adductors along with knee extensors, along with reduced range of motion associated with pain was present. Evaluation was done with Xray and CT. After rehabilitation, the joints motion improved upto 10°.

Conclusions: keeping in mind by seeing the present case, HO should always be kept in mind as a differential diagnosis in all the stroke patients who present with spontaneous joint pains. Both affected and unaffected sides may develop which has to be always kept in mind and proper early treatment has to be given.

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1. Introduction

Formation of lamellar bone within the soft tissue surrounding a joint is called Heterotopic ossification (HO). Generally seen in cases with long history of spinal cord injury (SCI), traumatic to brain causing injury, burns, and direct trauma.^{1,2} Varghese in his study stated that the incidence of Heterotopic ossification after developing stroke was 0.5–1.2%. The aetiopathogenesis of this still remains unclear. It been said that the major factors for developing Heterotopic ossification are immobilization and forcible manipulation of joints to achieve range of motion.^{3,4} Major contributing factors like fracture, spasticity, deep venous thrombosis infection, and development of pressure ulcers.^{5,6} It is mainly categorised into traumatic and neurogenic. Traumatic which generally develops after direct muscle trauma or surgery or any

irregular manipulations. Neurogenic generally develops after neurological lesions, like trauma to brain or any spinal cord injury (SCI).⁷ In post-stroke hemiplegia conditions occurrence of Heterotopic ossification is rare; very few cases were reported in the literature. Development of Heterotopic ossification in all the joints was not reported previously. We are presenting the case of a 50 yr man who had stroke and developed Heterotopic ossification in all his joints.

2. Case Report

Ten months before, a 50-year-old male was brought to the emergency department due to unbearable headache with previous history of seizures and was admitted. The patient followed by generalized seizure and loss his consciousness for about an hour at that time the Glasgow Coma Scale [GCS]: 5/15. He was intubated and mechanically ventilation was given at the time of unconsciousness. CT Brain was

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being done and which showed a subarachnoid bleed I both the cerebral hemisphere. Then he was shifted to ICU and was there for about 2 months. He was regularly monitored and all the care was taken. pt came out of ventilator after 2 months and at that time his gcs was 9/15. follow up Neurological examination showed that the patient developed rt hemiplegia. He was transferred to the observation department and was discharged later on 20th day with GCS 13/15. Two month later, he was brought back to emergency department with drowsiness, vomiting and speech and gait impairment and his condition was deteriorating along with that, spasticity of the right wrist and finger flexors was grade 3 according to the modified Ashworth scale (MAS) and spasticity of the gastrocnemius was grade 3. Active and passive range of motion of all the non-paretic knee was restricted (range 40–90°). In addition, there was swelling and warmth on the both knee. His treatment was complicated with The rehabilitation of the patient mainly comprised exercise and transfer activities, balance and gait training for 2 months by the time he got discharged he developed a grade 2 pressure sore, which treated with regular dressing and got resolved respectively and discharged. Regular physiotherapy was done in the rehabilitation clinic with regular followup was done for about a month. After a week and her clinical observation spasticity developed in hip flexors, adductors and knee flexors and the rom was very difficult to perform and even a slight moment gave pt a very severe pain ;Laboratory examination revealed a WBC count of 6800mm³, ESR of 40mmh¹ and serum ALP level of 147 IU L¹. The patient was evaluated with the x-rays Heterotopic Ossificans was identified in all the major joints [Figure 1]. C T was done to obtain differential diagnosis, and it clarified the development of the heterotrophic Ossificans. The pt was given Warfarin were administered. The whole episode compromised the daily activity of the patient and moment In his limbs so he was transferred to the training and physiotherapy which was not useful that the stage some functional recovery was achieved for about 5 to 20 degree of moment in all the joints on his last visit.

3. Discussion

In post-stroke hemiplegia Heterotopic ossification is very rare, the reporting of such type of cases is very low in the literature.² Heterotopic ossification aetiopathogenesis is not clear till date in such cases. Few authors proposed that the aetiopathogenesis is may be due to neuronal control mechanisms. Some stated that the main responsible for this metabolic control by a potential neurotransmitter might be the cause. Few say that conditions like Heterotopic ossification, Charcot joint, osteoporosis, are conditions that are controlled by same mechanism.⁸ Some say that factors that can stimulate are related to neurogenic Heterotopic ossification such as : prolonged comatose stage, mechanical

Fig. 1: Shows the effected both knee in the final follow up

Fig. 2: Shows effected left elbow

Fig. 3: Shows effected rt elbow

Fig. 4: Shows HO over both the sides

ventilation, increased muscle tone, and limited movement in the involved extremity.^{9,10} Some authors stated in such patients immobilization, passive ROM exercises, transfer activities and repeated micro-traumas might be the cause.^{3–5,8,10} few of the recent literature has shown that Heterotopic ossification may be traumatic.¹¹ Michelsson et al.^{3,4} said that immobilization and forcible mobilization are the most important causes of developing Heterotopic Ossification. In our patient, passive range of motion was done in both limbs equally, But none of it explains why HO developed in the non-affected side. The major regions like hips, shoulders, pelvis, elbows and knees are generally involved so as in our case.¹² If the patient has hemiplegia, general site of HO is the hip, where commonly flexor (anterior) or adductor (medial) compartments^{12–16} are affected. Nakajo and Endo¹⁷ in their study stated that HO developed in few of their patients with hemiplegia, most of them occurred around the hip, knee and elbow. In our cases, HO is atypical due to late development at 6 months' after developing stroke, generally the onset of HO ranges from 4–12 weeks, mostly occurred by 2 months.¹⁸ HO in neurologic disorders generally develops over affected side limbs. Garland et al.⁵ in his study stated that hemiparesis is the region where HO occurs. Whereas on the other hand, Kocaaga et al.¹⁹ stated and reported the development of HO in nonparetic knee. In our case, either neurogenic or traumatic or both factors together may have role in developing HO in an affected side other than affected side. It is hence important to keep in mind that the HO may develop both in affecting and non affecting side hence proper observation and early treatment is necessary to prevent HO formation. There are few studies which are against ROM in treatment protocol,^{3,13,20–22} gentle ROM which comes within the pain-free range should be done to patients.

4. Source of Funding

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5. Conflict of Interest

None.

References

1. Lal S, Hamilton BB, Heinemann A, Betts HB. Risk Factors for Heterotopic Ossification in Spinal Cord Injury. *Arch Phys Med Rehabil.* 1989;70:387–90.
2. Warren SB. Heterotopic ossification after total hip replacement. *Orthop Rev.* 1990;19:603–11.
3. Pathogenesis of experimental heterotopic bone formation following temporary forcible exercising of immobilized limbs. *Clin Orthop Relat Res.* 1983;176:265–72.
4. Michelsson JE, Granroth G, Andersson LC. Myositis ossificans following forcible manipulation of the leg. A rabbit model for the study of heterotopic bone formation. *J Bone Jt Surg.* 1980;62:811–5.
5. Garland DE. A Clinical Perspective on Common Forms of Acquired Heterotopic Ossification. *Clin Orthop Relat Res.* 1991;263:13–29.
6. Kocaaga Z, Bal S, Gurgan A. Hemiplegia and heterotopic ossification on the non-paretic extremity: A case report. *J Rehabil Med.* 2007;39(6):500–2.
7. Johns JS, Cifu DX, Keyser-Marcus L, Jolles PR, Frattin MJ. Impact of clinically significant heterotopic ossification on functional outcome after traumatic brain injury. *J Head Trauma Rehabil.* 1999;14(3):269–76.
8. Jones KB, Mollano AV, Morcuende JA, Cooper RR, Saltzman CL. Bone and Brain: A Review of Neural, Hormonal, and Musculoskeletal Connections. *Iowa Orthop J.* 2004;24:123–32.
9. Whyte J, Hart T, Laborde A, Rosenthal M. Rehabilitation issues in traumatic brain injury. *Phys Med Rehabil.* 2005;2.
10. Zeilig G, Weingarten HP, Levy R, Peer I, Ohry A, Blumen N. Heterotopic ossification in guillain-barré syndrome: incidence and effects on functional outcome with long-term follow-up. *Arch Phys Med Rehabil.* 2006;87(1):92–5.
11. Lotta S, Scelsi L, Scelsi R. Microvascular changes in the lower extremities of paraplegics with heterotopic ossification. *Spinal Cord.* 2010;39:595–8.
12. 1991. Available from: <https://pubmed.ncbi.nlm.nih.gov/1899635/>.
13. Stover SL, Hataway CJ, Zeiger HE. Heterotopic Ossification in Spinal Cord-Injured Patients. *Arch Phys Med Rehabil.* 1975;56(5):199–204.
14. Taly AB, Nair KP, Jayakumar PN, Ravishankar D, Kalaivani PL, Indiradevi B. Neurogenic Heterotopic Ossification : A Diagnostic and Therapeutic Challenge in Neurorehabilitation. *Neurol India .* 2001;49(1):37–40.
15. Baron M, Stern J, Lander P. Heterotopic Ossification Heralded by a Knee Effusion. *J Rheumatol.* 1993;10(6):961–4.
16. Genêt F, Jourdan C, Schnitzler A, Lautridou C, Guillemot D, Judet T, et al. Troublesome Heterotopic Ossification After Central Nervous System Damage: A Survey of 570 Surgeries. *PLoS One.* 2011;6(1):e16632. Available from: <https://pubmed.ncbi.nlm.nih.gov/21304993/>. doi:10.1371/journal.pone.0016632.
17. Gurcay E, Ozturk EA. Heterotopic ossification as rare complication of hemiplegia following stroke: Two cases. *Brain Inj.* 2013;27(13-14). doi:10.3109/02699052.2013.831123.
18. Rehabilitation after hip- and knee-joint replacement. An experience- and evidence-based approach to care. *Am J Phys Med Rehabil.* 2006;85(11):98–118.
19. Kocaaga Z, Bal S, Gurgan A. Hemiplegia and heterotopic ossification on the non-paretic extremity: A case report. *J Rehabil Med.* 2007;39(6):500–2.
20. Ackerman LV. Extra-osseous Localized Non-Neoplastic Bone and Cartilage Formation (So-Called Myositis Ossificans): Clinical and Pathological Confusion With Malignant Neoplasms. *J Bone Joint Surg Am.* 1958;40(2):279–98.

21. Garland DE, Shimoyama ST, Lugo C, Barras D, Gilgoff I. Spinal Cord Insults and Heterotopic Ossification in the Pediatric Population. *Clin Orthop Relat Res.* 1989;245:303–10.
22. Wharton GW, Morgan TH. Ankylosis in the Paralyzed Patient. *J Bone Jt Surg.* 1970;52:105–12.

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