DOI: 10.4274/tod.galenos.2024.29974
Turk J Osteoporos 2025;31(1):47-9

Bilateral Shoulder Osteoarthritis After COVID-19 in a Patient with Hereditary Spastic Paraplegia: A Case Report

Herediter Spastik Paraplejili Bir Hastada COVID-19 Sonrası Bilateral Omuz Osteoartriti: Olgu Sunumu

Burcu Sevingül, Şeyma Alkan, Hilal Devran, Yasemin Tombak, Özgür Zeliha Karaahmet
Ankara Etlik City Hospital, Clinic of Physical Medicine and Rehabilitation, Ankara, Türkiye

Abstract

The study, the first to report such a relationship to date, presents a patient with accelerated osteoarthritis (OA) in weight-bearing shoulder joints after Coronavirus disease-2019 (COVID-19). A 61-year-old man with hereditary spastic paraplegia was diagnosed with bilateral shoulder OA after presenting with bilateral shoulder pain and restricted range of motion. Laboratory tests and X-rays showed no pathology in the pelvis, hands, or knees. However, shoulder X-rays revealed narrowed joint space, subchondral sclerosis, and signs of aging in the capitulum humeri; Unlike the X-ray taken three years ago (before COVID-19). Magnetic resonance imaging revealed bilateral OA and bicipital tendinitis. Further research is needed on COVID-19's impact on joints.
Keywords: Cartilage, COVID-19, glenohumeral osteoarthritis, hereditary spastic paraplegia, osteoarthritis

Öz

Burada daha önce literatürde rastlamadığımız, Koronavirüs hastalığı-2019'dan (COVID-19) sonra ağırlık taşıyan omuz eklemlerinde hızlı gelişen osteoartrit (OA) olan bir hasta sunulmaktadır. Herediter spastik paraplejisi olan 61 yaşında bir erkek hasta, bilateral omuz ağrısı ve eklem hareket kısıtlılığı ile başvurduğunda bilateral omuz OA teşhisi aldı. Laboratuvar testleri ve pelvis, el ve diz direkt grafileri normaldi. Ancak omuz direkt grafisinde, üç yıl önce (COVID-19'dan önce) çekilen grafinin aksine, daralmış eklem aralığı, subkondral skleroz, kapitulum humeri yaşlanma belirtileri gösterdi. Manyetik rezonans görüntüleme, bilateral OA ve biseps tendinitini ortaya koydu. COVID-19'un eklemler üzerindeki etkisi hakkında daha fazla araştırmaya ihtiyaç var.
Anahtar kelimeler: Kıkırdak, COVID-19, glenohumeral osteoartrit, herediter spastik parapleji, osteoartrit

Introduction

Osteoarthritis (OA) is the world's most common joint disease and there is currently no cure. Glenohumeral OA (GHOA) accounts for an estimated 5-17% of patients with shoulder complaints. The etiology of GHOA is multifactorial. These can be divided into nonspecific and specific factors as well as systemic and local factors. Joint damage develops from the interplay between these factors, where local or systemic factors, or non-specific or specific factors, may dominate (1). Current knowledge is lacking about the impact of Severe acute respiratory syndrome-Coronavirus-2 (SARS-CoV-2) on cartilage degeneration and synovial inflammation. This case represents accelerated OA in the weight-bearing shoulder joints after Coronavirus disease-2019 (COVID-19). Symmetrical shoulder OA is rare, late-onset disease is usually seen in early age groups in rheumatologic disorders (2). Early age presentations can be triggered by increased systemic inflammation such as COVID-19.

Case Report

A 61-year-old male was admitted to the rehabilitation unit with the impairment of walking and bilateral shoulder pain with limited range of motion. His past medical history included hereditary spastic paraplegia (HSP), hypertension, diabetes, benign prostatic hyperplasia, and a confirmed COVID-19 diagnosis 3 years ago. He gave a family history of a son and daughter who were diagnosed with HSP. He was unable to walk without a walker. He reports the pain in both shoulders has been present for the last 3 years. On examination, upper extremity muscle strength

Corresponding Author/Sorumlu Yazar: Yasemin Tombak, Ankara Etlik City Hospital, Clinic of Physical Medicine and Rehabilitation, Ankara, Türkiye
E-mail: yasemintombak@hotmail.com **ORCID ID:** orcid.org/0000-0003-0065-5376
Received/Geliş Tarihi: 31.07.2024 **Accepted/Kabul Tarihi:** 18.10.2024 **Publication Date/Yayınlanma Tarihi:** 20.03.2025

Cite this article as/Atıf: Sevingül B, Alkan Ş, Devran H, Tombak Y, Karaahmet ÖZ. Bilateral shoulder osteoarthritis after COVID-19 in a patient with hereditary spastic paraplegia: a case report. Turk J Osteoporos. 2025;31(1):47-9

was evaluated as 5/5. Symmetrical lower limb weakness was predominantly distal and respectively graded in extensor hallucis longus as 1/5, ankle dorsi-flexors as 2/5, hip flexors as 3/5, knee extensors as 3/5, ankle plantar flexors as 5/5. Pain-free flexion and abduction range were respectively 145 and 150 degrees in the right shoulder; Both flexion and abduction range of the left shoulder were 160 degrees with a mechanical pain and crepitation sense during the movements of both shoulders. There was significant spasticity in the hip flexors, hip adductors, and hamstrings respectively scored as 2, 3, 2 using the Modified Ashworth Scale (MAS) (3).

Laboratory investigations and the X-ray imaging of the pelvis, hands, and knees did not show any pathology considering ankylosing spondylitis, rheumatoid arthritis, or other rheumatic disease. A plain X-ray of the shoulders showed narrowed joint space, subchondral sclerosis, marginal osteophyte formation, subchondral cyst formation, and aging signs of caput humeri Kellgren-Lawrence graded as 4 which did not exist in the X-ray taken three years ago (Figures 1A,1B). Magnetic resonance imaging of both shoulders performed for differential diagnosis showed bilateral OA and bicipital tendinitis. Prior chest computed tomography revealed post-COVID-19 pulmonary fibrotic-like changes.

In the rehabilitation program, active assistive range of motion exercises, strengthening and stretching of both upper and lower extremities, balance training, and an unweighted treadmill for ambulation have been administrated. The botulinum toxin applied to both hip adductor and gastrocnemius muscles improved one point on the five-point MAS following ten days post injections. On the bilateral shoulder conventional transcutaneous electrical nerve stimulation, with a frequency of 50 Hz, a current transition time of 100 microseconds, and an amplitude density that does not cause contractions or excessive discomfort was performed for 30 minutes. The shoulder joint positions improved from 145° to 165° in flexion and from 150° to 165° in abduction on the right, improved from 160° to 175° in flexion and abduction on the left. His participation in daily living activities was increased with a 60% decreased shoulder pain on the visual analog scale. The Functional Independence measure score improved from 102 to 110.

The study was conducted in accordance with the Declaration of Helsinki. Written informed consent for participation and publication was obtained from the patient.

Discussion

OA is a painful condition caused by a combination of biomechanical and metabolic features resulting in changes in articular cartilage and bone (4). GHOA is less common than OA of the weight-

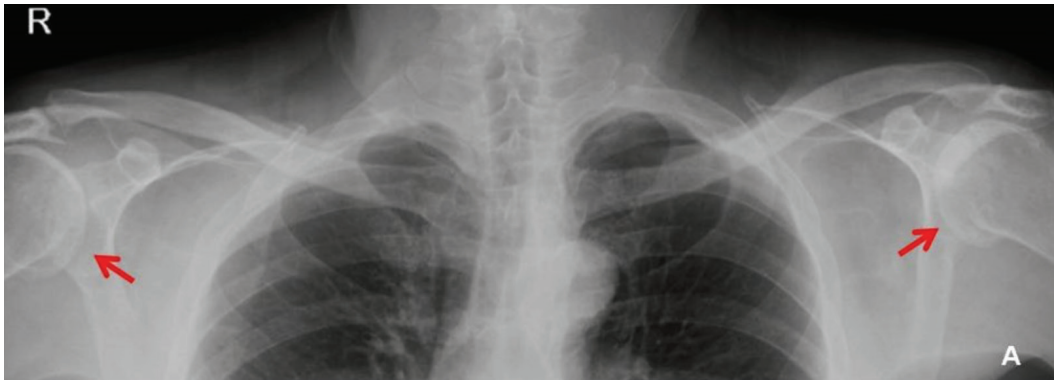

Figure 1A. X-ray of shoulders before COVID-19. A plain X-ray of the shoulders showed a normal joint space

COVID-19: Coronavirus disease-2019

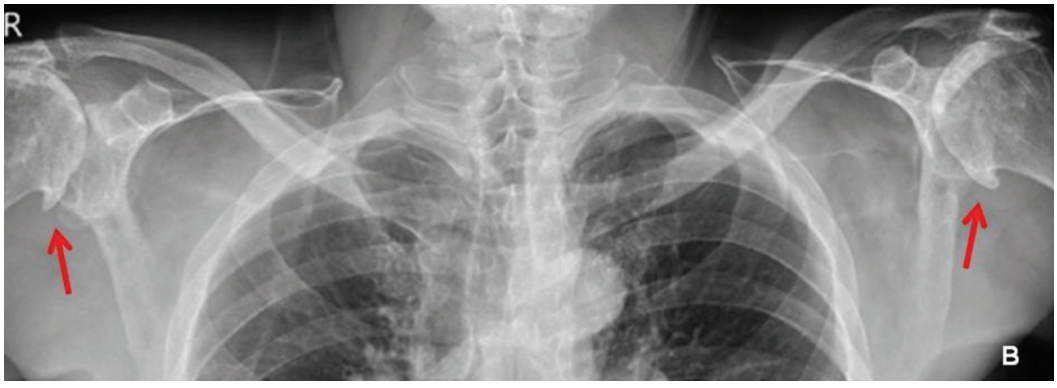

Figure 1B. X-ray of shoulders post-COVID-19. A plain X-ray of the shoulders showed narrowed joint space, subchondral sclerosis, marginal osteophyte formation, and subchondral cyst formation

COVID-19: Coronavirus disease-2019

bearing joints but is a painful condition that can severely affect independence by limiting the pain-free range of motion about the shoulder (1).

In this case report, a 61-year-old male patient presented with bilateral GHOA, complicated by HSP. The patient, who had to rely on his arms to walk, naturally put a load on his arms, which may have contributed to the development of OA, with the additional contribution of systemic inflammation such as SARS-CoV-2.

HSP is a clinical picture consisting of symmetrical spasticity and weakness in lower limbs that progresses very slowly, impairment of walking, increased tendon reflexes, and Babinski's sign. There is axonal degeneration prominently in the distal parts of corticospinal tracts and occasionally in the posterior cords. Numbness in the distal lower extremities and impaired vibration sense may be present. While only these symptoms are seen in uncomplicated forms, epileptic seizures, dementia, muscle atrophies, extrapyramidal findings, peripheral neuropathy, and cataracts may be seen in complicated forms. Although the inheritance of HSP is heterogeneous, the dominant form is more common. The prevalence ranges from 0.1 to 9.6 per 100,000 around the world (5).

In patients with HSP, spasticity in lower limbs requires them to use an assistance device to walk, and the weight-bearing shoulder may be at risk of developing OA. A study comparing the rate of shoulder OA between the control group who did not undergo surgery and the group after hip arthroplasty requiring to use of arm support did not find a significant difference (6). Environmental factors such as weight-bearing might lead to the development of the early OA-like phenotype without a viral presence in the joint. The binding of SARS-CoV-2 to angiotensin converting enzyme-2 (ACE-2) is assumed to initiate the endothelial and adipose dysfunction subsequently OA-like phenotype. The overstimulation of immune response in COVID-19 induces OA-like changes similar to metabolic syndrome leading to endothelial and adipose tissue dysfunction. In contrast to the disruption of the renin-angiotensin system pathway via ACE-2 receptors, the nicotinic cholinergic system as the anti-inflammatory pathway stimulates chondrocyte and osteoblast proliferation and restores subchondral bone (7).

Au et al. (8) show damage to the knee joint following in vivo infection with wild-type, Delta, and Omicron variants of SARS-CoV-2. Two patients with post-COVID OA experienced rapid joint damage with cystic lesions at the osteochondral junction, which was replicated in a golden Syrian hamster model. Viral spike proteins leaked into the subchondral bone as a result of increased vascular permeability brought on by SARS-CoV-2-activated endothelin-1 signaling. Histological confirmation of osteoclast activation, chondrocyte dropout, and cyst formation was obtained (8).

Conclusion

HSP is a progressive disease and causes struggle in walking and balance over time, resulting in the need for an assistive walking device. Our case emphasizes the contribution of COVID-19 to the early aging of unusual joints of disabled individuals who need an assistive device.

Ethics

Informed Consent: Written informed consent for participation and publication was obtained from the patient.

Foonotes

Authorship Contributions

Surgical and Medical Practices: B.S., Ö.Z.K., Concept: B.S., Ş.A., H.D., Design: Y.T., Ö.Z.K., Data Collection or Processing: B.S., Ş.A., H.D., Analysis or Interpretation: B.S., Y.T., Ö.Z.K., Literature Search: B.S., Ş.A., H.D., Writing: B.S., Y.T., Ö.Z.K.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declare that this study received no financial support

References

1. Ibounig T, Simons T, Launonen A, Paavola M. Glenohumeral osteoarthritis: an overview of etiology and diagnostics. Scand J Surg. 2021;110:441-51.
2. Stanborough RO, Bestic JM, Peterson JJ. Shoulder osteoarthritis. Radiol Clin North Am. 2022;60:593-603.
3. Yam WK, Leung MS. Interrater reliability of modified ashworth scale and modified tardieu scale in children with spastic cerebral palsy. J Child Neurol. 2006;21:1031-5.
4. Iannone F, Lapadula G. The pathophysiology of osteoarthritis. Aging Clin Exp Res. 2003;15:364-72.
5. Meyyazhagan A, Orlacchio A. Hereditary spastic paraplegia: an update. Int J Mol Sci. 2022;23:1697.
6. Miyake Y, Mitani S, Namba Y, Umehara N, Kawamoto T, Furuichi S. Survey of shoulder osteoarthritis in patients who underwent total hip arthroplasty for hip osteoarthritis. Acta Med Okayama. 2022;76:173-7.
7. Lauwers M, Au M, Yuan S, Wen C. COVID-19 in joint ageing and osteoarthritis: current status and perspectives. Int J Mol Sci. 2022;23:720.
8. Au MT, Ni J, Tang K, Wang W, Zhang L, Wang H, et al. Blockade of endothelin receptors mitigates SARS-CoV-2-induced osteoarthritis. Nat Microbiol. 2024;9:2538-52. Erratum in: Nat Microbiol. 2024;9:3391.