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CASE REPORT

Conservative Management for Spontaneous Lumbar Facet Joint Hemarthrosis in Severe Hemophilia a: A Rare Case Report

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Spontaneous lumbar facet joint bleeding is rarely observed in patients with hemophilia. Since it is categorized as a spinal epidural hematoma, surgery is the main treatment. Conservative treatment was successful in patients with minor or absent neurological deficits. We aimed to present the case of a patient with severe hemophilia who suffered from acute sciatica caused by lumbar facet joint hematoma. Nonsurgical treatment using a multidimensional approach was provided. The hemarthrosis is completely resolved. The patient recovered from clinical neurological symptoms without recurrence throughout the 4-year follow-up period. Spontaneous lumbar facet joint hemarthrosis in patients with severe hemophilia who present with mild neurological symptoms can be conservatively treated if the hemarthrosis is recognized early. Moreover, a multidisciplinary team approach is paramount in achieving good patient outcomes.

Key words: Hemophilia A, spontaneous lumbar facet joint hemarthrosis, conservative treatment

INTRODUCTION

Spinal bleeding is an uncommon site of spontaneous hemorrhage in patients with hemophilia. There are two types of intraspinal bleeding: epidural and subdural hematomas.[1] A rare type of epidural hematoma is the facet joint bleeding, which can arise from direct bleeding, synovial cyst, or ganglion cyst.[2] It must be noted that a facet joint is also a synovial joint. Moreover, hemophilic joint bleeding commonly occurs in the elbows, knees, and ankles, which are large synovial joints, and are believed to be susceptible for bleeding because of their richly vascularized synovial tissue and their exposure to strenuous mechanical forces with an abnormal hemostatic balance.[3] However, there has been no report on the spine as a bleeding site among hemophiliacs. Since hematoma may lead to severe or life-threatening neurological symptoms, most cases reviewed are treated via surgical intervention.[4,5] Here, we report a rare case of lumbar facet joint hemarthrosis in a patient with hemophilia A. He presented with sciatica. He was subsequently conservatively managed, which resulted in the complete resolution of symptoms.

CASE REPORT

The study was conducted with the approval and review of the institutional review board of Tri-service general hospital (No C202105127). (Approval day: 2021.07.07). A 64-year-old man with severe hemophilia A undergoing regular standard half-life factor VIII prophylactic replacement therapy three times a week presented with acute left lower back pain with radiation in the left ankle region in 2016. His target joint was right hip joint. He also had operative history of right total hip arthroplasty and bilateral total knee arthroplasty 20 years ago due to hemophilic arthropathy. He denied any physical injury or trauma. His physical examination results revealed tenderness over the left gluteal region and

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a positive straight leg raising test. The results of the spine radiography showed degenerative changes of the lumbar spine without intervertebral foramen narrowing. Magnetic resonance imaging (MRI) scans revealed a heterointense mass-like lesion around the left L5/S1 facet joint with left S1 root compression [Figure 1a and 2a]. Lumbar facet joint hematoma was first suspected because of the sudden onset of symptoms and history of hemophilia.

The patient received factor VIII replacement therapy (25 IU/kg) consecutively for 14 days and oral analgesic medications for 1 month. Upon 3 months later with improvement of severe low back pain, he further underwent rehabilitation, which included hot pack, short wave diathermy, interferential current therapy, therapeutic exercise, and core muscle strengthening. Left-sided radicular pain and numbness subsided gradually after 8 months. Moreover, follow-up MRI scans showed a reduction in hematoma size [Figure 1b and c, Figure 2b and c]. Through integrated care with the hematologist, physiatrist, and physiotherapist, the patient underwent ongoing prophylactic therapy and rehabilitation without interruption up to 4 years. There was neither recurrent facet joint bleeding nor neurological symptoms throughout the follow-up period [Figure 1d and 2d].

DISCUSSION

To the best of our knowledge, this is the first reported case of spontaneous lumbar facet joint hemarthrosis in a patient with hemophilia A. Management for patients with bleeding at this site is challenging for people with coagulopathy. Hence, the prevention of recurrence is important in delaying the development of hemophilic arthropathy.

In hemophilia, atypical bleeding at the spinal region is very rare. It represents 2% to 8% of central nervous system hemorrhage, which includes intracranial and intraspinal hemorrhage. In general, it only accounts for 3% to 8.7% of bleeding in patients with hemophilia.[1] Spontaneous spinal epidural hematoma (SSEH) is a surgical emergency that should be evaluated carefully and promptly to know the course of treatment to avoid long-term sequelae. Management is more difficult in patients with coagulopathy or a bleeding tendency due to the higher risk for rebleeding rate and extension of the hematoma after surgical decompression.[6] In a review literature for non-operative treatment of SSEH, Groen *et al*.[7] found there was no beneficial factor to facilitate conservative treatment. However, among the 64 patients who

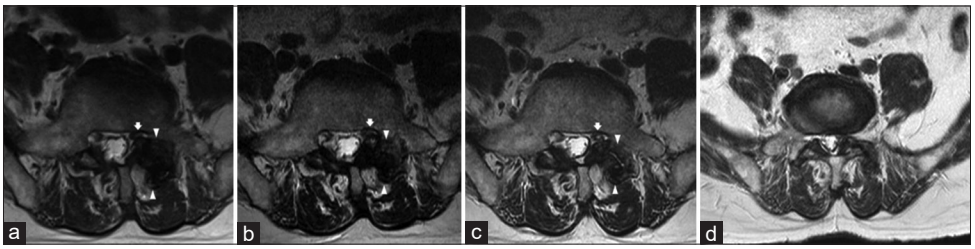

Figure 1: Magnetic resonance imaging of lumbar spine, sagittal T2-weighted images (a-c), axial T2-weighted fat spin echo (FSE) image (d). (a) Sagittal magnetic resonance images showing a heterointense mass-like lesion (arrowheads) located in the L5/S1 facet joint. Lumbar facet joint hematoma was first considered due to sudden onset of symptoms and hemophilia history. There is no herniated intervertebral disc or dehydrated disc. (b and c) Hematoma was gradually resolved at 3 months (b) and 9 months (c) after conservative treatment. (d) The sagittal image shows complete resolution of hematoma at 4-year follow-up

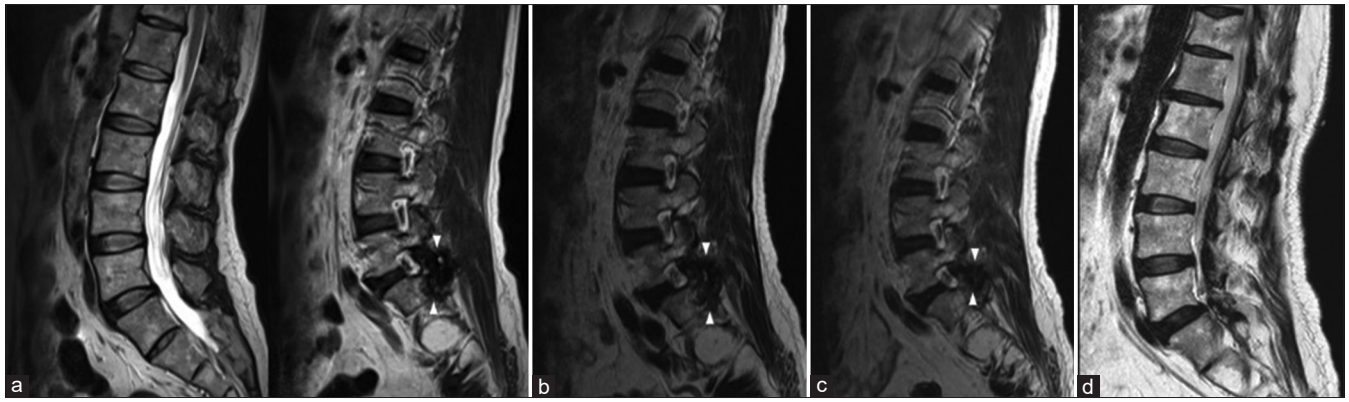

Figure 2: Magnetic resonance imaging of lumbar spine, axial T2-weighted images (a-c), axial T2-weighted fat spin echo (FSE) image (d). (a) Axial magnetic resonance images showing a heterointense mass-like lesion (arrowheads) located in the posterolateral spinal canal around left L5/S1 facet joint with left S1 root compression (arrows). (b and c) Gradual resolution of hematoma was observed at 3 months (b) and 9 months (c) after conservative treatment. (d) The axial image shows complete resolution of hematoma at 4-year follow-up

underwent conservative treatment, 54 had good outcomes. Patients with good outcomes experienced mainly minor neurological deficits.

The origin of the bleeding in common SSEH is the epidural venous plexus. Contrastingly, facet joint bleeding mostly arises from hemorrhage synovial cysts.[5,6] Synovial cysts may be associated with degenerative spinal changes, such as facet joint osteoarthritis and spondylolisthesis. Hemorrhagic synovial cysts are less understood. In fact, only trauma and anticoagulation therapy have been mentioned as possible etiologies.[8] Regardless of the cause, surgical decompression through laminectomy and cyst excision is the mainstay treatment.[5] Spontaneous resolution has only been reported in patients with synovial cysts.[9,10]

Our patient had severe hemophilia. Therefore, he was at risk for spontaneous bleeding, which still occurred despite prophylactic clotting factor substitution.[3] Because of surgical risks and absence of neurological deterioration, we decided to manage the patient conservatively. We closely followed him up to detect any clinical symptoms and to determine the hematoma size. The good outcomes may be due to the following reasons: (1) early detection of the hematoma on MRI; (2) absence of the hematoma's size increase; (3) achievement of hemostasis after the administration of clotting factor replacement therapy; and (4) improvement of function and reduction of pain by ingesting oral analgesics and undergoing physiotherapy. At present, nonsurgical management with the goal of relieving pain and preserving function is critical to prevent the development of hemophilic arthropathy for hemophilia. A multimodal approach, which includes a combination of pharmacotherapy, education, physiotherapy, and exercise, is needed.[3] In line with this, our patient constantly receives integrated care.

CONCLUSION

There is a lack of confirmed prognostic factors for the nonsurgical treatment of SSEH. This requires further investigation. We shared a conservatively treated case of a spontaneous lumbar facet joint hemarthrosis in a patient with severe hemophilia. Conservative management was possible partly due to early recognition. Moreover, good outcomes and the delay of the progression of hemophilic arthropathy were achieved through a multidisciplinary team approach.

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Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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