Epidural chloroma and spinal cord compression

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To the Editor: Chloroma is a rare solid tumor that was formed sometimes from malignant myeloid blast cells outside the hematopoietic system; these are known as chloroma or granulocytic sarcoma.[1] Chloroma can be diagnosed anywhere outside of the hematopoietic system, such as the head and neck, skeletal system, breast, uterus, and central nervous system.[2] This tumor is common in leukemic patients.[3] Chloroma presents as an epidural mass resulting in spinal cord compression with neurologic symptoms.[4] Spinal lesions occur most frequently at the thoracic level; however, the most common primary symptoms are low back pain and radiating pain in the legs.[5]

A 68-year-old male from China without other specific disease visited the Hematology Clinic of Seoul St. Mary's Hospital for evaluation of chronic myeloid leukemia. He was diagnosed with chronic myeloid leukemia based on the bone marrow pathology report (findings consistent with chronic myelogenous leukemia, BCR-ABL1 positive, chronic phase). The bone marrow cytogenics report revealed 46 X,Y, t(9;22)(q34;q11.2).[1] After diagnosis, there was nothing but medication for chemotherapy for chronic myeloid leukemia treatment; Nilotinib 300 mg bid from 22nd July to 13th August 2015, and then it took continuously up to 400 mg qd from 28th August 2015. A thoracolumbar magnetic resonance image showed focal bone marrow-replacing lesions at T11 and the sacrum, which were suspected to be related to chronic myeloid leukemia. He started chemotherapy and continued treatment. However, he had low back pain and radiating pain in both legs. He also complained of numbness in both feet and was referred to our pain clinic for management. Thoracolumbar computer tomography showed mild discogenic endplate degeneration in T7–8, a small Schmorl node in the superior endplate of T12, and diffuse bulging disc with ligamentum flavum thickening from L1 to S1 [Figure 1A]. His pain intensity was 9/10 on a numeric rating scale. We performed caudal epidural block with 0.2% lidocaine, hyaluronic acid, dexamethasone, and sacroiliac joint steroid injection with 0.4% lidocaine and triamcinolone, together with gabapentin for pain control. Two months later, his pain intensity had decreased from 9/10 to 3/10, but he still had low back pain and radiating pain in both legs Notwithstanding he could walk without a stick and his pain had much improved in comparison with the first visit. Six months later, he again visited our pain clinic. He had experienced no pain for several months and so had discontinued treatment. He had been employed as a construction worker for 1 month, and again began to experience low back pain. He had radiating pain and numbness in both legs. He also complained of weakness in both legs. His pain intensity was 7/10. We performed caudal epidural block with 0.2% lidocaine, hyaluronic acid, dexamethasone, and transforaminal steroid injection, at the L4–5 and L5–S1 levels with 0.2% lidocaine, hyaluronic acid, and dexamethasone [Figure 1B], and again prescribed appropriate medications including gabapentin for pain control. Four days after transforaminal steroid injection, he visited our pain clinic again. He complained of severe weakness in both legs and dysuria and dyschezia; his pain intensity was 4/10. The results of a complete blood cell count, blood coagulation test and blood chemistry were normal. His physical examination showed paresthesia and hypoesthesia below T12 and motor weakness of both lower extremities. He could not urinate by himself, so we inserted a Foley catheter. Voluntary anal contraction was also absent. We started steroid therapy and emergency thoracolumbar magnetic resonance imaging was performed. The results showed T2 hyperintensity with impeded water diffusivity in the epidural and paraspinal spaces of T11–T12, resulting in cord compression of T10–T12, which was suggestive of chloroma. Hemangiomas were also detected at T12 and L4 [Figure 1C]. In addition, rapid progression to paraplegia and magnetic resonance imaging made us suspicious of chloroma more clearly; on this basis, emergency surgery for decompression was performed. The frozen section removed during surgery confirmed chloroma. He underwent urgent total laminectomy and extradural spinal cord tumor removal for decompression. The pathologic findings revealed atypical small, round cell infiltration. Steroid

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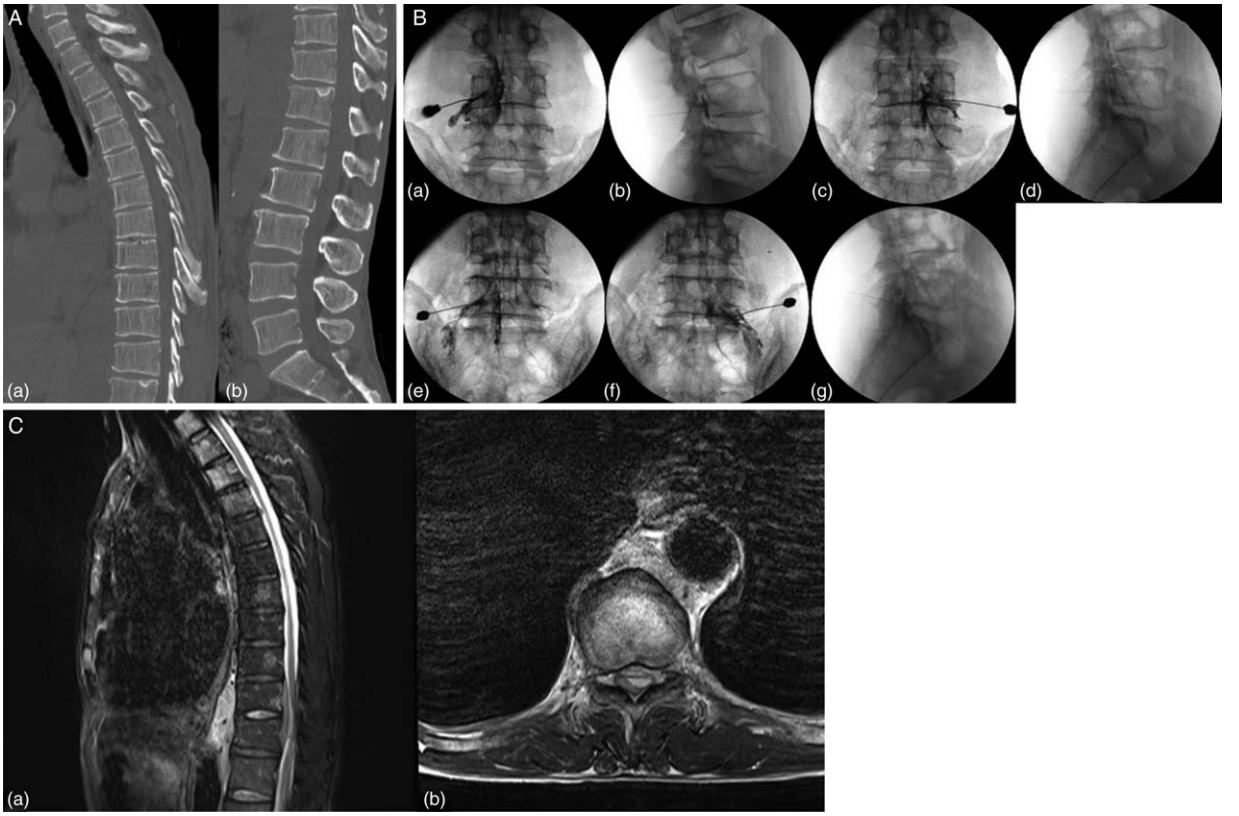

Figure 1: A 68-year-old chronic myeloid leukemic male with epidural chloroma and spinal cord compression. (A) The thoracolumbar computer tomography: C6–T12 (a) and T10–S2 (b), sagittal plane. (B) The transforaminal steroid injection (images with contrast): left anteroposterior (a), left lateral (b), right anteroposterior (c), and right lateral (d) views at L4–5 level; and left anteroposterior (e), right anteroposterior (f), and right lateral (g) views at L5–S1 level. (C) The thoracolumbar magnetic resonance imaging: sagittal plane at T2–L1 (a) and axial plane at T12 level (b).

therapy was continued and the neurologic dysfunction improved slightly. Additional palliative radiotherapy was performed at T9–T12, but the neurologic dysfunction remained. He refused bone marrow transplantation and was discharged and transferred to a local hospital for conservative care 35 days after the operation.

In our case, urgent decompressive laminectomy and tumor removal were performed immediately, followed by radiotherapy, but the paraplegia was not resolved. The patient with known acute or chronic myeloid leukemia who has progressive weakness and paresthesia should be suspected of spinal cord compression due to epidural chloroma.

Chloroma is localized proliferation of myelogenous leukemia cells. This solid tumor is often an initial manifestation of myelocytic leukemia. In our case, the chloroma is chronic myeloid leukemia differently located thoracic epidural area instead of central nervous system.[6] This chloroma didn't show triplegia but paraplesia and occurred in older ages than young, adolescents. Chloroma in the lumbosacral bone after diagnosis of chronic myeloid leukemia had been treated by management with both nerve block and radiation therapy.[7]

The patient, in this case, was 68 years of age. He was diagnosed with chronic myeloid leukemia but did not have signs of chloroma at the time of visiting our clinic. Because there was no involvement of the spine in the MRI prior to visit and it was judged to be cured, he was treated as a simple disc problem with a lumbosacral lesion. But it recurred within 6 months and the spine was invaded by the chloroma. For this series result, we think that if I first recognized the possibility and approached it earlier, it would have had a better impact on prognosis. This disease is uncommon in this age group, and the patient showed no signs of transformation into the acute stage. Paraplegia after transforaminal injection necessitated magnetic resonance imaging to diagnose common complications of nerve block; however, it was shown chloroma compressing the spinal cord. There was no effect by surgery and radiation therapy and the paraplegia was not resolved.

There was found similar to another case that occult epidural chloroma together with acute paraplegia after lumbar puncture.[8] This case was recommended myelography rather than magnetic resonance imaging for diagnosis.

As seen in this case, lifestyle adjustment advice or patient education about their disease was important. If he had a disability in the feces which was one of the most important clues of disease differentiation, we would have recommended to come to the clinic immediately and to take an MRI. This was a good case showing that the delay in treatment and misdiagnosis depending on the ability of the patient's understanding and education about the disease

The ideal treatment for chloroma is unclear. A patient with acute myeloid leukemia or chronic myeloid leukemia who shows progressive neurologic dysfunction should be

suspected of chloroma. In addition, this tumor may occur without hematologic disease or hematologic involvement. Therefore, magnetic resonance imaging may be a good diagnostic approach considering all the possible cases.

Declaration of patient consent

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

Conflicts of interest

None.

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