

# World Journal of *Clinical Cases*

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## Myeloid sarcoma with ulnar nerve entrapment: A case report

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### Abstract

#### BACKGROUND

Myeloid sarcoma (MS) is relatively rare, occurring mainly in the skin and lymph nodes, and MS invasion of the ulnar nerve is particularly unusual. The main aim of this article is to present a case of MS invading the brachial plexus, causing ulnar nerve entrapment syndrome, and to further clinical understanding of the possibility of MS invasion of peripheral nerves.

#### CASE SUMMARY

We present the case of a 46-year-old man with a 13-year history of well-treated acute nonlymphocytic leukaemia who was admitted to the hospital after presenting with numbness and pain in his left little finger. The initial diagnosis was considered a simple case of nerve entrapment disease, with magnetic resonance imaging showing slightly abnormal left brachial plexus nerve alignment with local thickening, entrapment, and high signal on compression lipid images. Due to the severity of the ulnar nerve compression, we surgically investigated and cleared the entrapment and nerve tissue hyperplasia; however,

subsequent pathological biopsy results revealed evidence of MS. The patient had significant relief from his neurological symptoms, with no postoperative complications, and was referred to the haemato-oncology department for further consultation about the primary disease. This is the first report of safe treatment of ulnar nerve entrapment from MS. It is intended to inform hand surgeons that nerve entrapment may be associated with extramedullary MS, as a rare presenting feature of the disease.

### CONCLUSION

MS invasion of the brachial plexus and surrounding tissues of the upper arm, resulting in ulnar nerve entrapment and degeneration with significant neurological pain and numbness in the little finger, is uncommon. Surgical treatment significantly relieved the patient's nerve entrapment symptoms and prevented further neurological impairment. This case is reported to highlight the rare presenting features of MS.

**Key Words:** Myeloid sarcoma; Ulnar nerve entrapment syndrome; Acute nonlymphocytic leukaemia; Sarcoma; Upper limb surgery; Case report

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**Core Tip:** The main aim of this article is to present a rare case of myeloid sarcoma (MS) invading the ulnar nerve and causing ulnar nerve entrapment syndrome. The patient's symptoms were relieved by surgical release of the nerve bundle and compressed tissue, and the disease was diagnosed definitively by pathological examination. The case illustrates the rare presenting features of extramedullary MS, enriches the clinical literature, and highlights that it is essential to suspect MS with a history of acute nonlymphocytic leukaemia.

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## INTRODUCTION

Myeloid sarcoma (MS), also known as granulocytic sarcoma and chloroma, is a rare haematological tumour consisting of a solid extramedullary mass of immature cells of the myeloid lineage, capable of destroying preexisting tissues and organs. It is more common in males than in females and is often associated with acute nonlymphocytic leukaemia (ANLL), which is frequently the primary manifestation of disease relapse[1-3]. MS develops mainly in the skin and lymph nodes but may affect almost any tissue or organ[4,5], and usually presents as an isolated mass of extramedullary infiltrates of leukaemia. To date, it is rare to encounter MS affecting the brachial plexus and producing ulnar nerve compression symptoms in the literature[6,7]. The main aim of this article is to present a case of ulnar nerve entrapment syndrome caused by MS invasion of the brachial plexus, furthering the clinical understanding of the potential for MS to involve peripheral nerves.

## CASE PRESENTATION

### Chief complaints

The patient was a 46-year-old man who complained of painful numbness in the left little finger for 4 mo with no apparent cause.

### History of present illness

The numbness and pain in his left little finger had gradually worsened over the past 4 mo but was not of serious concern.

### History of past illness

The patient had a 13-year history of ANLL and a 20-year history of psoriasis; both diseases had been

fully treated previously by systemic therapy.

### **Personal and family history**

Notably, there was no family history of the same disease.

### **Physical examination**

Physical examination of the whole body revealed no palpable lymph nodes. Specialized examination of the left hand revealed atrophy of the first dorsal interosseous muscle, numbness of the little finger and ulnar aspect of the palm, weakness of the ring and little finger, inability to adduct fingers, positive paper-clip test results, and loss of manual dexterity; all other examination outcomes were normal.

### **Laboratory examinations**

The laboratory results were: White blood cell count (WBC),  $14.14$  (normal range:  $3.50$ - $9.50$ )  $\times 10^9/L$ ; neutrophil count (NEUT),  $12.12$  ( $1.80$ - $6.30$ )  $\times 10^9/L$ ; lymphocyte count (LYMPH),  $0.78$  ( $1.20$ - $3.20$ )  $\times 10^9/L$ ; monocyte count,  $1.22$  ( $0.10$ - $0.60$ )  $\times 10^9/L$ ; eosinophil count (EOS),  $0.00$  ( $0.02$ - $0.52$ )  $\times 10^9/L$ ; %NEUT,  $85.8$  ( $40.0$ - $75.0$ )%; %LYMPH,  $5.50$  ( $20$ - $50$ )%; %EOS,  $0.0$  ( $0.4$ - $8.0$ )%; red blood cell count (RBC),  $4.0$  ( $4.3$ - $5.8$ )  $\times 10^9/L$ ; haemoglobin count (HGB),  $126$  ( $130$ - $175$ ) g/L; haematocrit,  $36.2$  ( $40.0$ - $50.0$ )%; rheumatoid factors,  $22.05$  ( $\leq 18.00$ ) IU/mL; hypersensitive C-reactive protein,  $4.67$  ( $\leq 4.00$ ) mg/L; uric acid,  $463.02$  ( $202.00$ - $416.00$ )  $\mu\text{mol/L}$ ; and interferon- $\gamma$ ,  $34.21$  ( $\leq 20.36$ ) pg/mL.

### **Imaging examinations**

X-rays showed that the bones of the left elbow were regular in shape, with cortical continuity and clear trabecular structures, a normal alignment of the elbow joint with a moderate joint space, and no significant abnormalities in the surrounding soft tissues (Figure 1A and B). Magnetic resonance imaging (MRI) showed a slightly abnormal left brachial plexus nerve alignment with local thickening, entanglement, and a high signal on compression lipid imaging, while the right brachial plexus nerve bundle alignment was normal with no obvious abnormal signal. There were multiple somewhat large lymph nodes in the neck, with the largest being approximately 0.8 cm in short diameter located in the left carotid sheath area (Figure 1C-F).

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## **FINAL DIAGNOSIS**

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MS causing ulnar nerve entrapment.

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## **TREATMENT**

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The patient was placed in a supine position, and an upper arm tourniquet was applied. After routine prepping and draping, a longitudinal incision of approximately 20 cm was made on the medial side of the left upper arm. Intraoperatively, the left ulnar nerve was found to be thickened and degenerated, with severe adhesions to the surrounding tissues. Further observation of the ulnar nerve revealed thickening of the epineurium and hyperplasia of the perineurium, trapping the nerve fibres; therefore, interfascicular neurolysis release was performed with careful separation of the hyperplastic tissue between the nerve bundles. In addition, the lymph nodes in the anterior axillary region were cleared and sent for pathological examination. The incision was closed in layers after irrigation and haemostasis.

Postoperative pathological findings were as follows: Lymphohaematopoietic tumour (both neuroepithelial and lymph nodes), combined with immunohistochemical staining results consistent with MS [CD3 (-), CD20 (-), CD21:FDC network (+), CD5 (-), Ki-67 (+ 70%), TdT (-), LCA (+), MPO (+), CD117 (+), and CD34 (+)] (Figures 2 and 3).

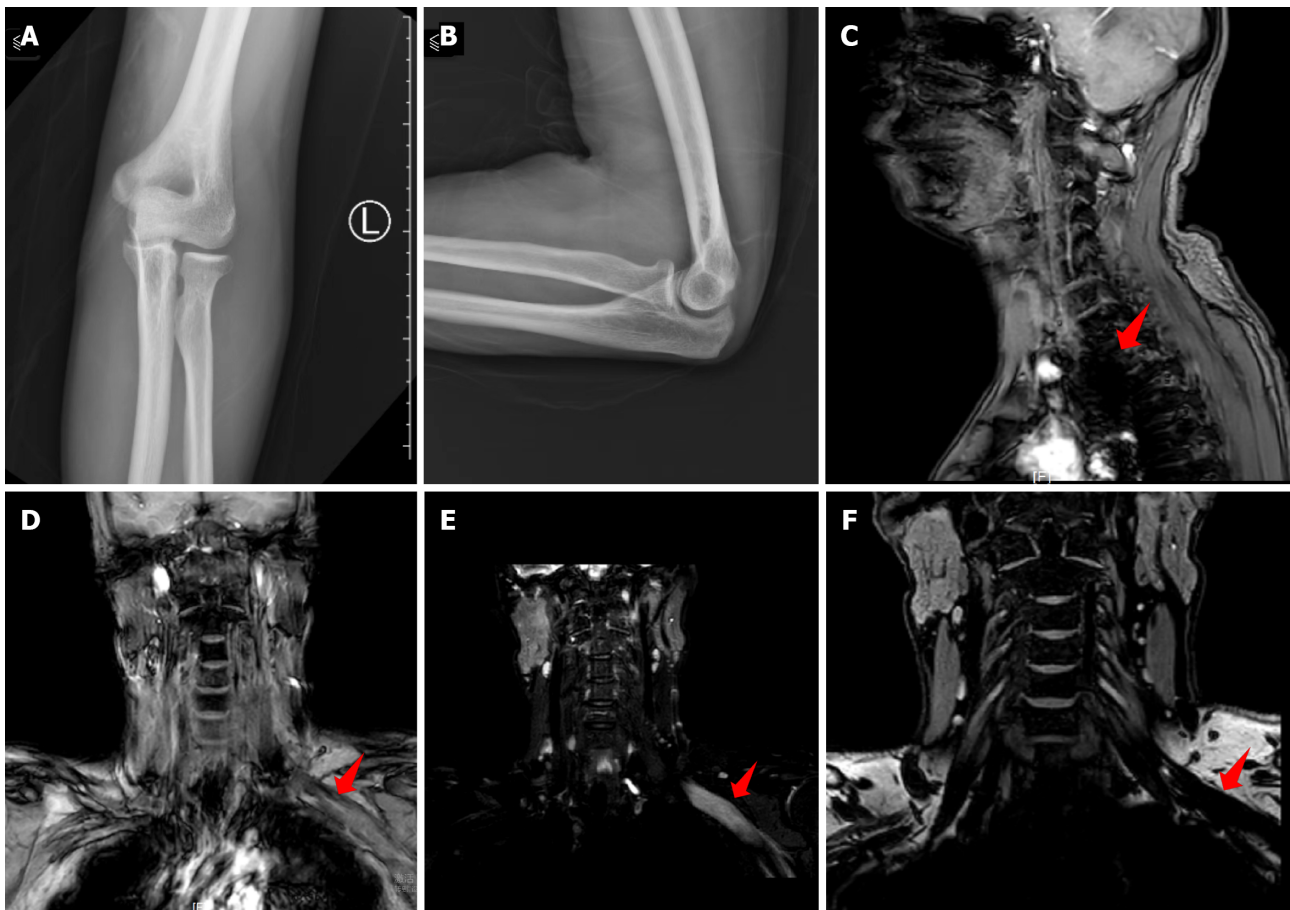
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## **OUTCOME AND FOLLOW-UP**

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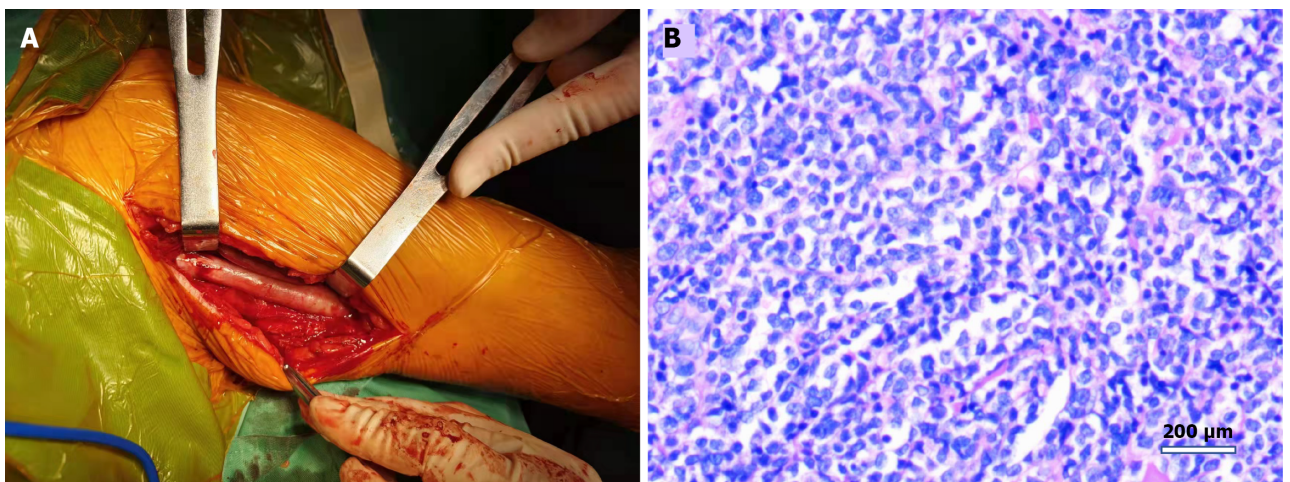
On the first postoperative day, the patient's pain and numbness were significantly reduced. After 1 wk of observation, we actively communicated with the patient and his family and transferred him to the external haemato-oncology department for further treatment of the primary disease. Positron emission tomography/computerised tomography (PET/CT) was performed, and the results were consistent with MS as per the pathology report. At the 2-mo follow-up, the patient had recovered well from the hand symptoms, the numbness had disappeared, and the postoperative visual analogue scale (VAS) score of pain had decreased to 1 from 5 preoperatively.





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**Figure 1 X-ray and magnetic resonance images.** A and B: No significant abnormality on antero-posterior (A) and lateral (B) X-rays of the left elbow joint; C and D: Sagittal (C) and coronal (D) magnetic resonance images (MRI) showing significant abnormal signal in the left brachial plexus; E and F: Coronal MRI showing significant abnormal signal in the left brachial plexus. The red arrow points to the lesion.



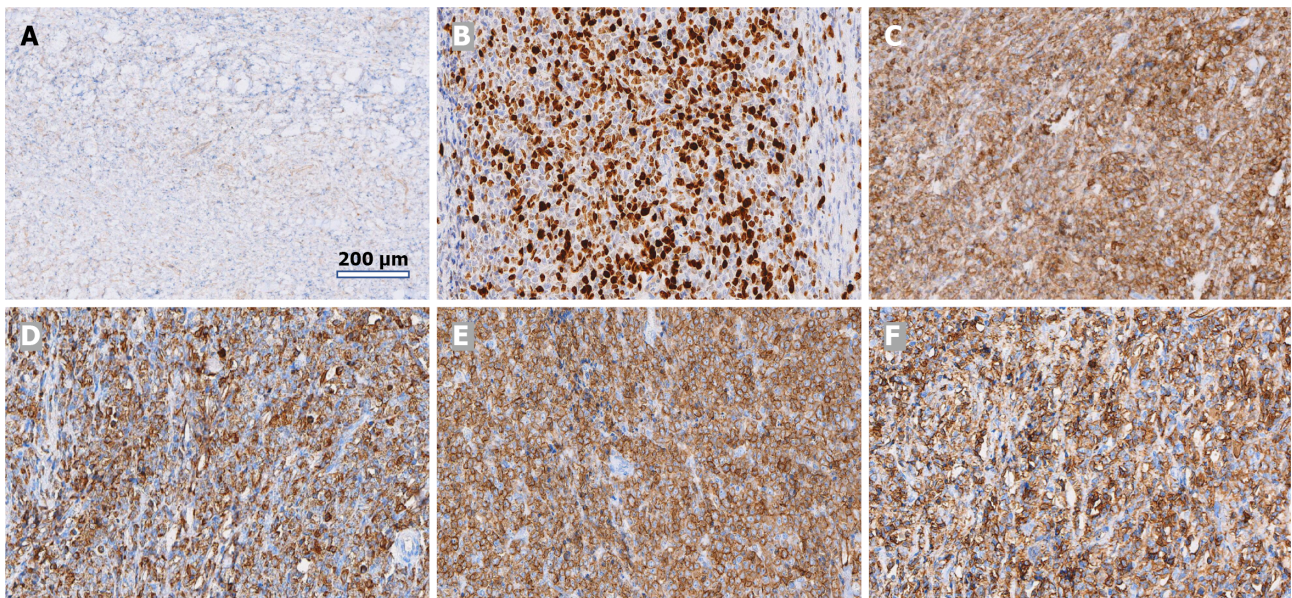
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**Figure 2 Intraoperative and histological images.** A: Intraoperative thickening and degeneration of the left ulnar nerve; B: Hematoxylin and eosin-stained section showing neuroepithelial and lymph node pathology.

## DISCUSSION

The incidence of MS in acute myeloid leukaemia (AML) patients ranges from 3%-5% [8]. Although the exact pathogenesis of MS is not fully understood, multivariate analysis has demonstrated that the occurrence of MS is significantly associated with intrinsic properties of myeloid tumour cells (*e.g.*,





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**Figure 3 Histopathology of myeloid sarcoma.** Immunopathological examination shows tissue of lymphohaematopoietic lineage. A: CD21; B: Ki-67; C: LCA; D: MPO; E: CD117; F: CD34.

cytogenetics)[9-11]. MS may manifest in many different parts of the body, including peripheral nerves, and its clinical symptoms depend on the specific location and size of the tumour, making it very difficult for doctors to diagnose based on symptoms alone[12-16].

For early MS diagnosis, biopsy and PET/CT images are useful, with the maximum standardized uptake value (SUV<sub>max</sub>) on PET/CT images for MS detection often in the range of 2.1-9.3, while MRI can only be used to detect tissue abnormalities and not to provide a definitive diagnosis[7,17]. Ultrasound of areas affected by MS can also be valuable in the assessment of mass size and borders[18]. The aim of tissue biopsy is diagnostic clarification, with cytochemical and immunophenotypic analyses, such as myeloid cell markers, anti-myeloperoxidase, and lysozyme, being particularly helpful in gaining a definitive diagnosis[19-23]. Studies have shown that MS is often misdiagnosed as non-Hodgkin's lymphoma, and in the absence of immunohistochemical results, the misdiagnosis rate is 50%-75%[24]. It has also been shown that 88% of patients with untreated isolated MS progress to AML within 1 year[25] and that MS is a precursor to relapse of the primary disease[8,26,27]. Due to its rarity, case reports mostly constitute the literature for MS, and studies on treatment options are mainly retrospective analyses of small sample sizes, with a lack of large randomized prospective studies[28-30]. Therefore, a uniform and consistent approach to optimal treatment has not been established. However, given the risk of progression to leukaemia and the poor prognosis, the main aim of treatment is to delay progression and prolong survival as long as possible[19,31-33].

We review cases of peripheral nerve compression due to MS over the last 10 years (Table 1) and found that similar cases were predominantly observed when MS invaded the eye (3 cases) and spine (12 cases), while invasion of the brachial plexus is very rare. Due to the rarity of MS-associated peripheral nerve compression, the resulting misdiagnosis may lead to a delay in optimal MS treatment[31,34].

The clearest symptom in the present case was ulnar nerve palsy, most typically caused by cubital tunnel syndrome. As our investigations showed that the nerve signal was abnormal, we first explored and released the nerve. This resulted in a significant reduction in the patient's pain, and numbness was significantly reduced on the first postoperative day. The subsequent pathology results were unexpected, but we were able to expedite referral to haemato-oncology for primary disease treatment.

## CONCLUSION

MS causing ulnar nerve entrapment and degeneration through invasion of the brachial plexus is a very rare condition. With a history of ANLL with nerve entrapment features, it is essential to suspect MS and send tissue for pathological analysis, as well as to provide prompt surgical decompression for symptom relief and prevent lasting neurological damage.

**Table 1** Cases of peripheral nerve compression due to myeloid sarcoma over the last 10 years

Ref.	Gender/age (yr)	Leukaemia-Yes (type)/No	Symptoms	Involved segment	Lesion invasion site	Imaging	Bone marrow biopsy	Laboratory test findings	Treatment regimens	Pathology and immunohistochemistry examination
McCarty <i>et al</i> [35]	M/14	No	Headaches, urinary incontinence	Intracranial, extracranial, sacral spine	Cerebral nerve, sacral nerve root	MRI	Positive	WBC: $51 \times 10^9/L$ ; HGB: 113 g/L; platelets: $130 \times 10^9/L$	Radiotherapy, chemotherapy	-
Zhu <i>et al</i> [36]	M/36	No	Abdominal pain, pain in right eye, eye protrusion with inability to close	Pancreas, right eye	Pancreas, right optic nerve	CT, MRI, PET-CT	Negative	Mildly elevated liver and pancreatic enzymes	Radiotherapy, chemotherapy	Ki-67 (+ 80%), P53 (approximately 50%), CD43 (+), MPO (+)
Bai <i>et al</i> [24]	M/29	No	Severe radiating pain in lower limbs, moderate difficulty in urination	Waist	Lumbar spine attachment, spinal cord	CT, MRI, SPECT	Negative	WBC: $6.01 \times 10^9/L$ ; RBC: $5.00 \times 10^{12}/L$ ; HGB: 158 g/L; platelets: $236 \times 10^9/L$ ; CRP: 2 mg/L; erythrocyte sedimentation rate (ESR): 4 mm/1 h; procalcitonin (PCT): 0.27 ng/mL	Lesion excision, chemotherapy	CD33 (+), MPO (+)
Valsamis <i>et al</i> [37]	M/56	No	Pain in right hip radiating down right leg	Right hip	Right sciatic nerve, lumbosacral plexus	Ultrasound, MRI	-	MONO: $0.9 \times 10^9/L$ ; RBC: $3.56 \times 10^{12}/L$ ; HGB: 116 g/L; mean cell volume: 102 fL; CRP: 13 mg/L	-	-
Pandey <i>et al</i> [38]	(1) F/44; (2) F/16; (3) F/63; (4) M/53	(1,2,4) No; (3) Yes (AML)	(1) Neck pain, right arm weakness, and muscle wasting; (2) back pain, bilateral lower extremity tingling, and weakness; (3) lower back pain, bilateral lower extremity weakness, and bowel incontinence; and (4) lower back pain, hip pain, lower extremity weakness, and muscle pain	(1) Cervical spine; (2) thoracic spine; (3) thoracic spine; and (4) lumbar spine	Spinal cord compression	-	(1) Negative; (2) positive; (3) negative; and (4) positive	(1) and (3) Negative; (2) and (4) high WBC	Lesion excision	-
Snoj <i>et al</i> [39]	F/49	Yes [MDS (refractory anaemia with excess blasts)]	Weakness and decreased sensation in right arm, right supraclavicular mass	Left upper limb	Left brachial plexus	Ultrasound, MRI	-	-	Radiotherapy, chemotherapy	-
Yamamoto <i>et al</i> [40]	F/34	Yes (Chronic phase chronic myeloid leukaemia)	Pain in left eye, protrusion of eyeball, blurred vision with headache, and vomiting	Left eye	Left optic nerve	MRI	-	-	-	CD13 (+), CD33 (+), CD117 (+)
Koh <i>et al</i> [41]	M/68	Yes (chronic myeloid leukemia)	Low back pain and numbness in both legs with radiating pain	Thoracic and sacral spine	Spinal cord	MRI, CT	-	-	Chemotherapy	-
Kim <i>et al</i> [42]	M/62	Yes (acute myeloid leukaemia, M2)	Abnormal sensation and weakness of ankle flexion in posterior aspect of left calf and foot	Lower left limb	Sciatic nerve	Ultrasound, MRI	-	-	-	MPO (+)



Rambeloson <i>et al</i> [43]	M/1	No	Bilateral ophthalmoplegia with loss of vision	Bilateral eye	Occulomotor nerve	Myelogram, CT	-	-	-	-
Slouma <i>et al</i> [31]	M/56	No	Progressive back pain and bilateral sciatica	Waist	Lumbar 5 <sup>th</sup> nerve root	MRI	-	WBC: $83 \times 10^9/L$ ; absolute neutrophil count: $60 \times 10^9/L$	Chemotherapy	MPO (+), CD13 (+), CD33 (+), CD14 (+), CD4 (+), CD46 (+)
Anqi <i>et al</i> [44]	M/24	No	Limp, numbness in right lower limb, mild urinary frequency, urinary and faecal incontinence	Lumbosacral region	Lumbar and sacral spine	MRI	-	Normal	Lesion excision, chemotherapy	MPO, CD34, CD43 (+)
Ha <i>et al</i> [34]	F/38	Yes (AML)	Subacute radiating pain in right upper limb, weakness	Right upper limb	Right brachial plexus	MRI, PET/CT	-	-	Chemotherapy, hormone shock therapy	CD13(+), CD14 (+) ,CD15 (+), CD33 (+), CD54 (+), CD64 (+), CD117 (+) , MPO (+)

SPECT: Single-photon emission computed tomography; WBC: White blood cell count; RBC: Red blood cell count; MRI: Magnetic resonance imaging; MS: Myeloid sarcoma; PET/CT: Positron emission tomography/computerised tomography; CRP: C-reactive protein; AML: Acute myeloid leukaemia; HGB: Haemoglobin count; ESR: Erythrocyte sedimentation rate; PCT: Procalcitonin.

## FOOTNOTES

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