MINIREVIEWS

1 Omicron variant (B.1.1.529) of SARS-CoV-2: Mutation, infectivity, transmission, and vaccine resistance
   Ren SY, Wang WB, Gao RD, Zhou AM

12 Hepatitis B virus reactivation in rheumatoid arthritis
   Wu YL, Ke J, Zhang BY, Zhao D

23 Paradoxical role of interleukin-33/suppressor of tumorigenicity 2 in colorectal carcinogenesis: Progress and therapeutic potential
   Huang F, Chen WY, Ma J, He XL, Wang JW

ORIGINAL ARTICLE

Case Control Study

35 Changes in rheumatoid arthritis under ultrasound before and after sinomenine injection
   Huang YM, Zhuang Y, Tan ZM

43 Benefits of multidisciplinary collaborative care team-based nursing services in treating pressure injury wounds in cerebral infarction patients
   Gu YH, Wang X, Sun SS

Retrospective Study

51 Outcomes and complications of open, laparoscopic, and hybrid giant ventral hernia repair
   Yang S, Wang MG, Nie YS, Zhao XF, Liu J

62 Surgical resection of intradural extramedullary tumors in the atlantoaxial spine via a posterior approach
   Meng DH, Wang JQ, Yang KX, Chen WY, Pan C, Jiang H

71 Vancomycin lavage for the incidence of acute surgical site infection following primary total hip arthroplasty and total knee arthroplasty
   Duan MY, Zhang HZ

79 Distribution of transient receptor potential vanilloid-1 channels in gastrointestinal tract of patients with morbid obesity
   Atas U, Erin N, Tazegul G, Elpek GO, Yildirim B

91 Value of neutrophil-lymphocyte ratio in evaluating response to percutaneous catheter drainage in patients with acute pancreatitis
<table>
<thead>
<tr>
<th>Page</th>
<th>Title</th>
<th>Authors</th>
</tr>
</thead>
<tbody>
<tr>
<td>104</td>
<td>Influence of overweight and obesity on the mortality of hospitalized patients with community-acquired pneumonia</td>
<td>Wang N, Liu BW, Ma CM, Yan Y, Su QW, Yin FZ</td>
</tr>
<tr>
<td>117</td>
<td>Minimally invasive open reduction of greater tuberosity fractures by a modified suture bridge procedure</td>
<td>Kong LP, Yang JJ, Wang F, Liu FX, Yang YL</td>
</tr>
<tr>
<td>128</td>
<td>Increased levels of lactate dehydrogenase and hypertension are associated with severe illness of COVID-19</td>
<td>Jin ZM, Shi JC, Zheng M, Chen QL, Zhou YY, Cheng F, Cai J, Jiang XG</td>
</tr>
<tr>
<td>136</td>
<td>Age, alcohol, sex, and metabolic factors as risk factors for colonic diverticulosis</td>
<td>Yan Y, Wu JS, Pan S</td>
</tr>
<tr>
<td>155</td>
<td>Characterization of focal hypermetabolic thyroid incidentaloma: An analysis with F-18 fluorodeoxyglucose positron emission tomography/computed tomography parameters</td>
<td>Lee H, Chung YS, Lee JH, Lee KY, Hwang KH</td>
</tr>
</tbody>
</table>

**Clinical Trials Study**

| 166  | Low-dose intralesional injection of 5-fluorouracil and triamcinolone reduces tissue resident memory T cells in chronic eczema | Wu Y, Wang GJ, He HQ, Qin HH, Shen WT, Yu Y, Zhang X, Zhou ML, Fei JB |

**Observational Study**

| 189  | Predicting adolescent perfectionism: The role of socio-demographic traits, personal relationships, and media | Livazović G, Kuzmanović K                                              |
| 205  | Novel m.4268T>C mutation in the mitochondrial tRNA\(^{\text{Ile}}\) gene is associated with hearing loss in two Chinese families | Zhao LJ, Zhang ZL, Fu Y                                                  |

**Randomized Controlled Trial**

| 227  | Zinc carnosine-based modified bismuth quadruple therapy vs standard triple therapy for *Helicobacter pylori* eradication: A randomized controlled study | Ibrahim N, El Said H, Choukair A                                      |
CASE REPORT

236 Acquired coagulation dysfunction resulting from vitamin K-dependent coagulation factor deficiency associated with rheumatoid arthritis: A case report
Huang YJ, Han L, Li J, Chen C

242 Intraoperative thromboelastography-guided transfusion in a patient with factor XI deficiency: A case report
Guo WJ, Chen WY, Yu XR, Shen L, Huang YG

249 Positron emission tomography and magnetic resonance imaging combined with computed tomography in tumor volume delineation: A case report
Zhou QP, Zhao YH, Gao L

254 Successful response to camrelizumab in metastatic bladder cancer: A case report
Xie C, Yuan X, Chen SH, Liu ZY, Lu DL, Xu F, Chen ZQ, Zhong XM

260 HER2 changes to positive after neoadjuvant chemotherapy in breast cancer: A case report and literature review
Wang L, Jiang Q, He MY, Shen P

268 Hyper-accuracy three-dimensional reconstruction as a tool for better planning of retroperitoneal liposarcoma resection: A case report
Ye MS, Wu HK, Qin XZ, Luo F, Li Z

275 Recurrent postmenopausal bleeding - just endometrial disease or ovarian sex cord-stromal tumor? A case report
Wang J, Yang Q, Zhang NN, Wang DD

283 Complex proximal femoral fracture in a young patient followed up for 3 years: A case report
Li ZY, Cheng WD, Qi L, Yu SS, Jing JH

289 Bilateral Hypertrophic Olivary Degeneration after Pontine Hemorrhage: A Case Report
Zheng B, Wang J, Huang XQ, Chen Z, Gu GF, Luo XJ

296 Clinical characteristics and outcomes of primary intracranial alveolar soft-part sarcoma: A case report
Chen JY, Cen B, Hu F, Qiu Y, Xiao GM, Zhou JG, Zhang FC

304 Removal of laparoscopic cerclage stitches via laparotomy and rivanol-induced labour: A case report and literature review
Na XN, Cai BS

309 Cerebral venous sinus thrombosis in pregnancy: A case report
Zhou B, Huang SS, Huang C, Liu SY

316 Eustachian tube teratoma: A case report
Li JY, Sun LX, Hu N, Song GS, Dou WQ, Gong RZ, Li CT
<table>
<thead>
<tr>
<th>Page</th>
<th>Title</th>
<th>Authors</th>
</tr>
</thead>
<tbody>
<tr>
<td>331</td>
<td>Lunate dislocation with avulsed triquetral fracture: A case report</td>
<td>Li LY, Lin CJ, Ko CY</td>
</tr>
<tr>
<td>361</td>
<td>Diagnostic and surgical challenges of progressive neck and upper back painless masses in Madelung’s disease: A case report and review of literature</td>
<td>Yan YJ, Zhou SQ, Li CQ, Ruan Y</td>
</tr>
<tr>
<td>371</td>
<td>Suspected cerebrovascular air embolism during endoscopic esophageal varices ligation under sedation with fatal outcome: A case report</td>
<td>Zhang CMJ, Wang X</td>
</tr>
<tr>
<td>381</td>
<td>An atypical primary malignant melanoma arising from the cervical nerve root: A case report and review of literature</td>
<td>Shi YF, Chen YQ, Chen HF, Hu X</td>
</tr>
<tr>
<td>388</td>
<td>Epidural blood patch for spontaneous intracranial hypotension with subdural hematoma: A case report</td>
<td>Choi SH, Lee YY, Kim WJ</td>
</tr>
</tbody>
</table>
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An atypical primary malignant melanoma arising from the cervical nerve root: A case report and review of literature

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Author contributions: Shi YF drafted the article or made critical revisions related to the important intellectual content of the manuscript; Chen YQ analyzed and interpreted the data; designed the research study; Chen HF approved the final version of the article to be published; Hu X provided substantial contributions to conception and design of the study; All authors issued final approval for the version to be submitted.

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Abstract

BACKGROUND
Primary melanomas affecting the central nervous system are very rare, and melanomas originating in the spinal canal or origin of the spinal nerve root are even rarer. As a consequence, not much is known about this.

CASE SUMMARY
Here we report a case of primary malignant melanoma originating in the cervical spinal cord nerve root. A 64-year-old woman presented with symptoms of numbness in the right side of the neck, pain, and hypoesthesia in the right upper limb which persisted for 1 year. Neurological examination showed that the superficial sensation in the right upper limb had decreased with muscle strength of grade 4. Magnetic resonance imaging examination revealed a mass (approximately 2.5 cm × 1.4 cm × 1 cm) in the right side of the spinal canal in the C-2 plane. Based on findings obtained during operation, perioperative examination, pathological diagnosis, and the diagnostic criteria of primary central melanoma proposed by Hayward, the mass was confirmed to be a melanoma of intraspinal nerve root origin.

CONCLUSION
This is the first case of primary malignant melanoma originating from cervical spinal cord nerve roots and spread along the inside and outside of the spinal canal. The clinical relevance of this case is discussed to provide new insights into the differential diagnosis of intraspinal tumours. Further studies are needed to better understand the mechanisms driving the growth pattern and development of this type of tumour.

Key Words: Spinal cord; Primary melanoma; Nerve root; Primary neoplasm; Case report

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Core Tip: Primary malignant melanoma arising from the cervical nerve root, and spreading along the inside and outside regions of the spinal canal is clinically rare. The clinical symptoms and imaging features of this case are atypical and can easily be misdiagnosed. In this case report, additional clinical characteristics and differential diagnoses are presented.

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INTRODUCTION
Primary central nervous system melanomas are very rare, and account for about 1% of all melanomas[1]. In addition, primary malignant melanoma in the spinal canal is even rarer with only several cases reported so far[2]. Majority of such cases show nerve root involvement. Herein, we present an unusual case of primary malignant melanoma originating from the nerve root in the spinal canal without a history of irradiation exposure. To date, there have been only 5 similar cases[3,4]. However, as far as we know, the patient described in this report presents the first case of primary malignant melanoma in which the growth occurs along the inside and outside of the spinal canal.

CASE PRESENTATION

Imaging examinations
Magnetic resonance imaging (MRI) examination showed a mass (approximately 2.5 cm × 1.4 cm × 1 cm) in the right side of the spinal canal in the C-2 plane. The mass exhibited high signal intensity on T1-weighted images and isointense on T2-weighted images. The signal intensity did not vary significantly between the T1-weighted images after contrast enhancement. The boundary was clear and spinal cord appeared significantly compressed and displaced to the left side (Figure 1). Results shown in Figure 1D indicate that the tumour extended beyond the intervertebral foramen to the outside of the spinal canal.

Laboratory examinations
Her preoperative laboratory examination, electrocardiogram, and lung computerized tomography (CT) findings were normal.

Physical examination
Neurological examination showed that the superficial sensation in the right upper limb had decreased accompanied by a muscle strength of grade 4. A general examination did not find any subcutaneous nodules or skin lesions.

Personal and family history
No positive personal or family history.

History of past illness
The patient had no previous medical history or a family history of malignant melanoma. She denied having any previous irradiation exposure.

History of present illness
The patient presented with progressive numbness in the right side of the neck, pain, and hypoesthesia in the right upper limb that occurred in the previous 1 year.

Chief complaints
A 64-year-old woman was admitted to our hospital with numbness in the right side of
Figure 1 A: T1-weighted showed high-intensity; B: T2-weighted was equal-signal; C: Contrast Enhancement showed clear boundary, and the spinal cord is significantly compressed and displaced to the left side; D: White arrow the tumor grew out of spinal canal through intervertebral foramen.

the neck, pain, and hypoesthesia in the right upper limb which persisted for 1 year.

MULTIDISCIPLINARY EXPERT CONSULTATION
None.

FINAL DIAGNOSIS
Based on the findings obtained during operation, perioperative examination, pathological diagnosis, and the diagnostic criteria of primary central melanoma proposed by Hayward, the neoplasm was considered to be a melanoma of intraspinal nerve root origin with an atypical growth pattern.

TREATMENT
We adopted the posterior median approach which allowed us to successfully remove the tumour. During the operation, the subdural region appeared black. After opening the dura mater, a black object was seen wrapped in a membranous structure close to the nerve root. The object grew out of the spinal canal through the intervertebral foramen. The lesion was close to the pia mater surface of the spinal cord. However, it was clearly demarcated with spinal cord tissue and pia mater. It had a tough and solid texture, with little blood supply, and did not invade the dura mater (Figure 2A).

OUTCOME AND FOLLOW-UP
After operation, no postoperative neurological deficits were observed and postoperative pathological diagnosis confirmed malignant melanoma (Figure 2B-2E).
Figure 2 A: A black mass (tumor) was tough and solid, lack of blood supply; B: Hematoxylin-eosin stain (HE, original magnification ×200); C: HMB45(+); D: S100(+); E: Ki67 (approximately 20%); F: Positron emission tomography/computed tomography scan showed negative.

A follow-up positron emission tomography/CT scan (Figure 2F) 1 mo after surgery showed total excision and no signs of metastasis and residual tumour. The postoperative course was uneventful, and the patient was discharged on the 14th day. At the last telephone follow-up in October 2021, the patient reported having no issues. We believe that her condition is stable based on routine follow-up MRIs.

DISCUSSION

Primary spinal cord melanoma is a very rare entity. Primary melanoma of the CNS originates from aberrant changes in pigment cells of the neural crest or from melanocytic elements of the pia mater during early embryonic development[5,6]. The clinical presentation of this condition varies from person to person, depending on the location of the tumour[7]. Primary spinal melanomas have been reported in different regions including intramedullary, intradural, and extradural lesions[8]. Most of such cases occur within the cervical and thoracic cord[9]. We retrospectively analysed 70 cases of primary spinal malignant melanoma between 1930 and 2021 on the PubMed Medline database. It was observed that tumours that grow from the inside to the outside of spinal canal are very rare (Table 1).
Table 1 Primary spinal melanoma: A review of the available English literature since 1930

<table>
<thead>
<tr>
<th>Tumor feature</th>
<th>Number of patients</th>
</tr>
</thead>
<tbody>
<tr>
<td>Growth pattern</td>
<td></td>
</tr>
<tr>
<td>Communicating the intervertebral Foramen</td>
<td>1</td>
</tr>
<tr>
<td>Extradural</td>
<td>5</td>
</tr>
<tr>
<td>Intradural-extradural</td>
<td>33</td>
</tr>
<tr>
<td>Intradural-intra-and-extra-medullary</td>
<td>4</td>
</tr>
<tr>
<td>Intramedullary</td>
<td>22</td>
</tr>
<tr>
<td>Other</td>
<td>5</td>
</tr>
<tr>
<td>Location</td>
<td></td>
</tr>
<tr>
<td>Cervical</td>
<td>24</td>
</tr>
<tr>
<td>Thoracic</td>
<td>33</td>
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<tr>
<td>Lumbar</td>
<td>10</td>
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<tr>
<td>Sacral</td>
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</tr>
</tbody>
</table>

In terms of clinical manifestation, the most common initial symptom of spinal schwannoma is pain, followed by loss of nerve function[9]. In our case, symptoms of nerve root stimulation were pronounced before operation, which matched the typical clinical manifestations of spinal schwannoma. For the spinal Schwannoma, MRI imaging shows a dumbbell shape and widening of intervertebral foramen[10]. In our case, this growth pattern was first seen in the spinal melanoma. Using MRI, the primary imaging modality used for evaluation of spinal neoplasm[11], the signal of this lesion emerged from under the epidural of spinal cord, as in the case of schwannoma and meningioma[10]. Besides, most of these lesions show signal hyperintensity on T1-weighted images[8,11], rarely iso-intensity on T2-weighted images[12,13], as well as mild to moderate homogenous enhancement[14,15]. It should be that the MRI signal of the lesion showed signal hyperintensity on T1 weighted image and equal signal on T2. The signal intensity after enhancement was not significantly different from that on T1-weighted images. It has been postulated that a higher blood supply in a lesion yields a higher signal intensity after enhancement[16]. The signal hyperintensity on T1-weighted image was due to the concentration of melanin, haemorrhages, and fat deposits[17,18]. The signal characteristics of MRI may easily lead to misdiagnosis. It is important for surgeons to make an accurate diagnosis and be aware of the limitations of the diagnostic value of MRI. For instance, enhanced MRI revealed a pronounced dural tail sign, which is a classic characteristic of meningioma. However, T1-weighted images with hyperintensity and T2-weighted images with hypointensity are typical features for melanoma, and atypical for meningioma. In addition, intra-tumoral bleeding may cause uneven hyper-intensive signal in T1 weighted images. However, it has been reported that enhancement scan of melanoma originating from intramedullary melanoma will be significantly enhanced. Moreover, if there is bleeding in the tumour, the signal will show mixed density[19].

Moreover, blood supply within a tumour seen during the operation was not abundant. As a consequence, we hypothesized that there was no obvious enhancement in the enhanced scan of melanoma originating from the nerve root. These factors make the preoperative diagnosis of atypical cases difficult, and our case reveals the diverse growth patterns associated the imaging findings of primary central nervous system melanoma. This is extremely important for the design of surgical strategies[20]. Thus, when making a preoperative diagnosis based on neuroimaging and clinical experience, there is need to make a more comprehensive prediction of the benign and malignant lesions before preventive measures are applied during operation and the tumour is excised. This only prevents metastasis and influences prognosis of patients.

In most melanomas occurring in the spine, they have primary lesions or metastases in other parts of the spine[21]. Therefore, the present case adds to the understanding on such melanomas. Notably, the positive rate of Ki-67 index in the postoperative pathological examination of present case was more than 20%. In theory, cancer cells of this case will proliferate rapidly and are likely to metastasize[22,23]. However, patients didn’t receive radiotherapy and chemotherapy. No recurrence or metastasis was found during follow-up, indicating that the primary malignant melanoma originating from...
the nerve root may have different biological characteristics[24], or such tumours have a better prognosis than do cutaneous melanomas[25]. As a consequence, the primary malignant melanoma in the spinal canal may be less invasive than in other parts which require more evidence-based medicine from the clinical experience. The choice of treatment[26,27] for such cases is extremely important. Gross total resection has been shown to result in longer progression-free survival and survival compared with no or partial resection[28,29]. Because the gross total resection of the tumour result in good outcomes and a longer longevity[30,31]. Nevertheless, the efficacy of post-operative radiotherapy or chemotherapy is still controversial[32,33].

CONCLUSION

In the present report, we present the first case of a primary malignant melanoma originating from the cervical spinal cord nerve roots and grew from the inside to the outside of the spinal canal. Unlike most primary melanomas of the spinal canal, this case has an unusual tumour origin, growth pattern, and imaging findings. Therefore, it provides new insights into the understanding and differential diagnosis of intraspinal tumours. Further studies are needed to reveal the mechanisms driving the development and growth pattern of such a tumour.

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