




Ruptured Filum Terminale Ependymoma as an Uncommon Cause of Refractory Lumbosciatica in a Pregnant Patient: Case Report

Ependimoma del filum terminal roto como causa infrecuente de lumbociática refractaria en paciente embarazada: Reporte de caso

Esteban Espinoza¹ Manuel González¹ Ratko Yurac²

¹Hospital San Camilo, San Felipe, Chile

²Clínica Alemana de Santiago, Vitacura, Chile

Address for correspondence Esteban Espinoza, MD, MSc, Hospital San Camilo, San Felipe, Chile

(e-mail: esteban.espinoza@ncaconcagua.com).

Rev Chil Ortop Traumatol 2024;65(2):e102–e107.

Abstract

Lumbar pain and sciatica are a common complaint during pregnancy. Neoplastic etiologies, although rare, require consideration in the differential diagnosis, particularly in the presence of acute onset, severe symptoms, or neurologic deficits.

We present the case of a 33-year-old woman at 31 weeks gestation presenting with acute, progressive lumbar pain, bilateral sciatica, and motor weakness. Neurological examination revealed bilateral lower extremity motor weakness (M4) in L3, L4, and L5 myotomes, with hyperreflexia and Brudzinski and Kernig signs. Magnetic resonance imaging demonstrated an L2-L3 level expansive intradural lesion compressing the descending nerve roots, consistent with a filum terminale ependymoma. A cesarean section occurred at 33 weeks gestation, followed by a tumor resection three days later. Histopathology confirmed the diagnosis of a myxopapillary ependymoma. The patient experienced a favorable postoperative course, with resolution of sciatica and gradual improvement in motor function. At the four-month follow-up, she regained normal ambulation and could care for her infant independently. She returned to her work duties six months after surgery.

This case highlights the importance of considering spinal tumors in the differential diagnosis of acute lumbosacral radiculopathy with neurologic deficits during pregnancy. Early diagnosis and prompt surgical intervention can lead to a favorable outcome for both mother and baby.

Keywords

- ▶ lumbar pain
- ▶ pregnancy
- ▶ spinal tumors
- ▶ ependymoma

received
November 27, 2023
accepted
August 12, 2024

DOI <https://doi.org/10.1055/s-0044-1790193>.
ISSN 0716-4548.

© 2024. Sociedad Chilena de Ortopedia y Traumatología. All rights reserved.

This is an open access article published by Thieme under the terms of the Creative Commons Attribution-NonDerivative-NonCommercial-License, permitting copying and reproduction so long as the original work is given appropriate credit. Contents may not be used for commercial purposes, or adapted, remixed, transformed or built upon. (<https://creativecommons.org/licenses/by-nc-nd/4.0/>)

Thieme Revinter Publicações Ltda., Rua do Matoso 170, Rio de Janeiro, RJ, CEP 20270-135, Brazil

Resumen

El dolor lumbar y la ciática son síntomas frecuentes durante el embarazo. Sin embargo, las etiologías neoplásicas, aunque raras, deben considerarse en el diagnóstico diferencial, particularmente en presencia de inicio agudo, síntomas graves o déficits neurológicos.

Presentamos el caso de una mujer de 33 años con 31 semanas de gestación que se presentó con dolor lumbar agudo y progresivo, ciática bilateral y debilidad motora. El examen neurológico reveló debilidad motora bilateral en las extremidades inferiores (M4) en los miotomas L3, L4 y L5, con hiperreflexia y signos de Brudzinski y Kernig. La resonancia magnética evidenció una lesión intradural expansiva a nivel L2-L3 que comprimía las raíces nerviosas descendentes, compatible con un ependimoma del filum terminal. Se realizó una cesárea a las 33 semanas de gestación, seguida de resección del tumor tres días después. La histopatología confirmó el diagnóstico de un ependimoma mixopapilar. La paciente experimentó un curso postoperatorio favorable, con resolución de la ciática y una mejora gradual de la función motora. A los cuatro meses de seguimiento, había recuperado la deambulación normal y podía cuidar a su bebé de forma independiente. Regresó a sus actividades laborales seis meses después de la cirugía.

Palabras clave

- ▶ dolor lumbar
- ▶ embarazo
- ▶ tumores espinales
- ▶ ependimoma

Este caso enfatiza la importancia de considerar los tumores espinales en el diagnóstico diferencial de la radiculopatía lumbar aguda con déficits neurológicos durante el embarazo. El diagnóstico precoz y la intervención quirúrgica oportuna pueden conducir a un resultado favorable tanto para la madre como para el bebé.

Introduction

In pregnant patients, low back pain is a frequent cause of outpatient consultation, occurring in up to 56% of cases. Pregnancy increases lumbar lordosis and causes an anterior pelvic tilt to compensate for the enlarged abdominal circumference and the resulting anterior shift of the center of gravity.¹ Typically, low back pain appears in the second trimester of pregnancy, becoming more frequent around 22 weeks.² Diagnosis relies on physical examination and identification of risk factors, considering red flags for neurological, oncological, or infectious symptoms. These findings require additional studies to define therapeutic management.³ There are algorithms for the study and management of a pregnant patient with low back pain, such as the one presented in ► **Figure 1**, adapted and translated from Sehmbi et al.⁴

The prevalence of sciatica in pregnancy has been scarcely investigated, mainly due to low back pain during pregnancy. However, an estimated 17% of pregnant women have sciatica at some point during pregnancy.^{5,6} Compressive lumbar nucleus pulposus hernias are rare, occurring in approximately 1 in 10,000 affected pregnant women, and only 15% requiring surgery.⁷ Detection of other spinal conditions causing symptoms, such as tumors or infections, is less common during pregnancy.⁸

The incidence of all types of cancer is unusual in pregnancy, occurring in approximately 1 in 1,000 pregnancies.⁹ Spinal tumors resulting in symptoms during pregnancy are much rarer than a lumbar nucleus pulposus hernia,

so medical experience is limited, and recommendations for managing these cases are scarce.¹⁰ The treatment of benign tumors can be conservative until delivery, unless these neoplasms produce symptoms and surgical management must be done earlier.¹¹ Malignant tumors require a more aggressive treatment since pregnancy could create a favorable environment for cell growth due to the increased total blood volume, vascularization, and hormonal factors inherent to pregnancy.¹²

Ependymomas are rare tumors of neuroectodermal origin emerging from the ependymal cells of the central medullary canal, filum terminale, choroid plexus, or periaqueductal white matter. Their annual incidence ranges from 1 to 4 per 1,000,000 inhabitants. The myxopapillary subtype is a benign tumor (World Health Organization [WHO] grade I) located virtually exclusively in the region of the conus medullaris, cauda equina, and filum terminale.¹³ The mainstay of treatment for myxopapillary ependymoma is total macroscopic resection, which provides the most favorable outcome and minimizes the possibility of recurrence to approximately 10 to 20%.¹⁴ Mortality is low, with a 5-year survival rate of 85 to 100%.¹⁵ The literature reports a few cases of myxopapillary ependymoma detected during pregnancy due to refractory sciatica or after an epidural anesthetic puncture, as presented in one report.¹⁶

Therefore, the investigation of a spinal tumor in a pregnant patient requires a very high level of diagnostic suspicion and attention to clinical warning signs.¹⁷ Some meningiomas and ependymomas during pregnancy have been described, potentially presenting with lumbar pain and neurological

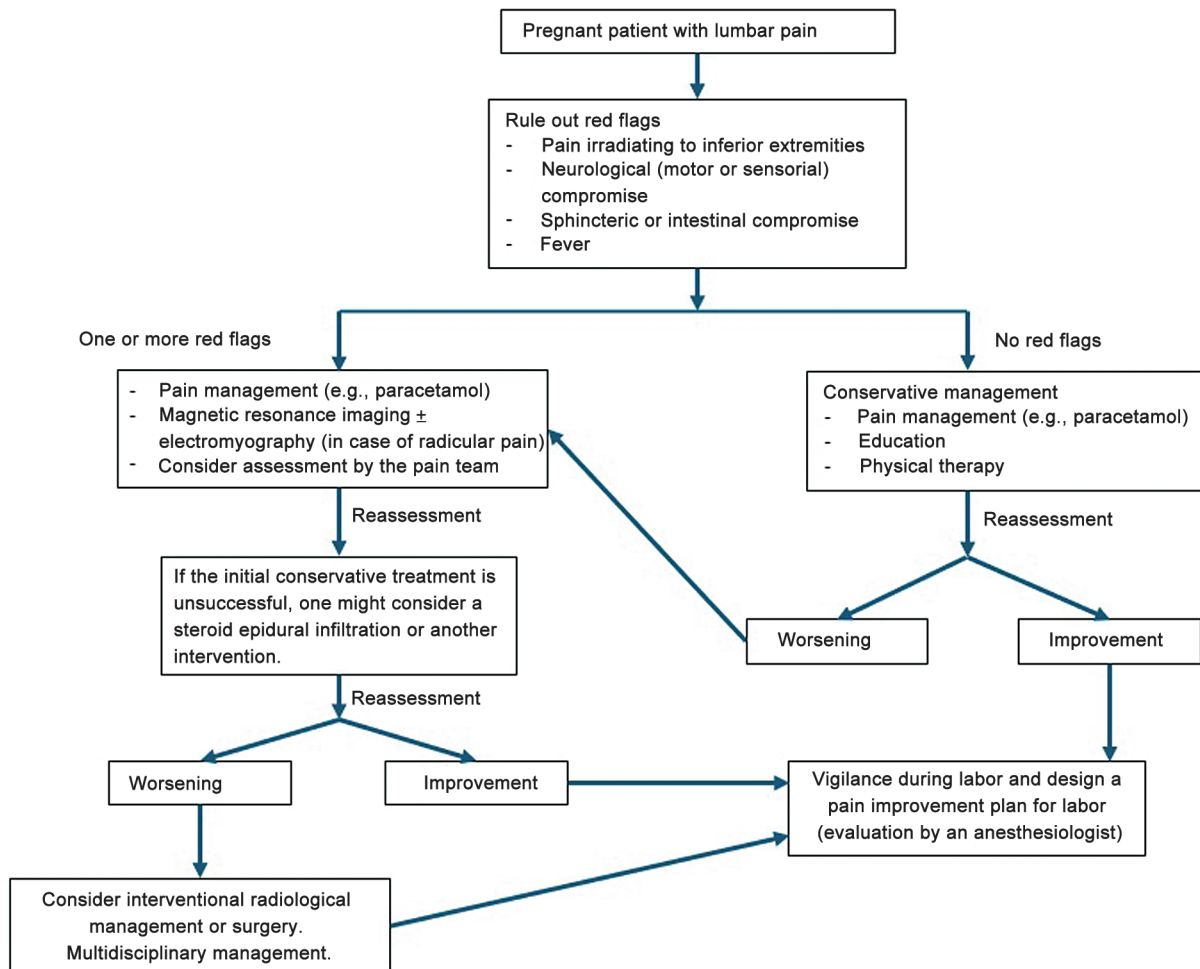


Fig. 1 Proposed algorithm to study a pregnant patient with low back pain, translated and adapted from Sehmbi et al.⁴

conditions depending on their location in the spinal canal. One must consider that these patients may present residual functional deficits after surgical resection.¹⁸

Case Presentation

We present the case of a 33-year-old patient with no history of chronic conditions, 31 weeks pregnant, who attended a gynecological emergency service with a 1-day history of acute low back pain, rapidly progressive, associated with bilateral sciatica and motor impairment of the lower extremities. Due to refractoriness to pain with analgesic management and motor impairment, the patient was admitted to optimize pain management and complete the study. Her clinical examination revealed a Glasgow coma scale (GCS) of 15, no fever, normotension, and signs of bilateral radicular irritation in the lower extremities, motor weakness (M4) due to paresis of L3, L4, and L5 myotomes, and Brudzinski and Kernig signs. Laboratory tests showed a white blood cell count within normal ranges, slightly elevated C-reactive protein levels (22 mg/dL), no electrolyte disorders, and blood sugar levels within normal range. As such, we requested a magnetic resonance imaging (MRI) of the lumbar spine, detecting an intraspinal expansive lesion at the L2-L3 bone level of 3.1 × 2.3 cm compressing the descending roots, in addition to subarachnoid hemorrhage in the dural

cul-de-sac explaining the observed meningeal signs. These findings were consistent with an intraspinal tumor of the filum terminal ependymoma type as a first diagnostic possibility (→ Fig. 2).

The patient was informed and agreed to perform an elective cesarean section at 33 weeks to plan tumor resection. The cesarean section occurred with no intercurrents, and we scheduled the tumor resection surgery for 3 days later. A lumbar laminotomy accessed the dural sac, revealing a highly vascularized neoplastic lesion intraoperatively, dependent on the filum terminale, separable from descending roots, and immersed in serosanguinous cerebrospinal fluid (CSF), consistent with subarachnoid hemorrhage shown at MRI. We performed a complete macroscopic resection (→ Fig. 3), corroborated by a postoperative MRI 24 hours after surgery (→ Fig. 4). A final biopsy confirmed the diagnosis of WHO I myxopapillary ependymoma.

The patient had a favorable course with bilateral lumbosciatica elimination and lower limb paresis maintenance. She was discharged one week later with a rehabilitation plan. In successive outpatient follow-ups, the patient regained normal walking at 4 months, recovering M5 muscle strength in the left lower limb, maintaining M4 on the left side L4 and L5, and having no issues caring for her baby. She returned to work at 6 months.

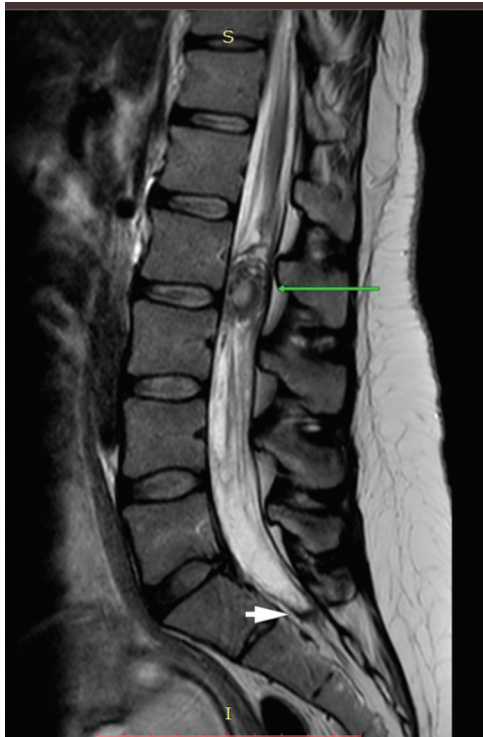


Fig. 2 Lateral lumbar spine T2-weighted MRI. The green arrow indicates a tumor lesion at the L2-L3 level with compression of descending roots, and the white arrow shows the blood level in the dural cul-de-sac, indicating subarachnoid hemorrhage.



Fig. 4 A postoperative lateral T2-weighted magnetic resonance imaging demonstrates complete tumor resection.

Discussion

Lower back pain in pregnant patients is a common condition explained by the many physiological changes occurring during pregnancy. Sciatica in pregnancy is a common symptom, affecting 17% of pregnant patients. Sciatica diagnosis in

pregnant women can sometimes be confused with pregnancy-related muscle cramps or conditions with a vascular origin, since pregnancy is the main contributing factor for varicose veins.¹⁹ Assessment of pregnant patients requires

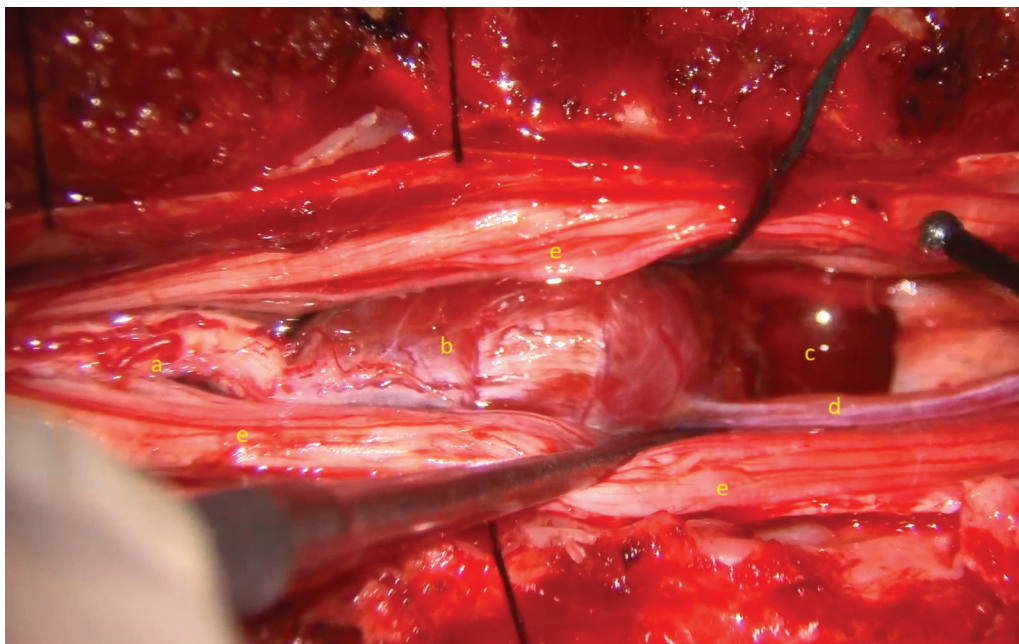


Fig. 3 Intraoperative image of ependymoma. (a) The proximal segment of the filum terminale presents increased thickness. (b) Ependymoma. (c) Presence of cerebrospinal fluid and subarachnoid hemorrhage. (d) Distal segment of the filum terminale. (e) Descending roots.

caution and a thorough physical examination to look for red flags of neurological, oncological, or infectious signs. In our case, the presence of meningeal signs was critical to suspect an unusual pain etiology.

After detecting any red flags, the next step is to confirm the etiological diagnosis with a complementary test that is harmless during pregnancy. The most common conditions in pregnant women with low back pain, or sciatica include herniated nucleus pulposus, vertebral hemangiomas, and stress fractures. Neoplastic etiology is very rare but requires consideration as a potential pain cause during pregnancy. MRI is the most used imaging study in pregnant patients with neurological involvement.⁴

Non-contrast MRI is the safest imaging study modality for pregnant patients, and it offers better resolution than ionizing tests such as fluoroscopy or computed tomography scans.²⁰ To date, there are few reports on the potential MRI effects on pregnancy, and hypothetical risks include fetal teratogenicity, acoustic damage, or effects induced by heat energy absorption in animal models, but with no confirmation in humans.^{21,22} The recommendations of the American College of Radiology (ACR) and the American College of Obstetrics and Gynecology (ACOG) agree that MRI has no association with adverse effects on the fetus but that it must occur prudently and only when the study significantly contributes to the diagnosis and management.^{23,24}

Another safe imaging test in pregnancy is ultrasound, but it does not provide the information needed for most spinal conditions. The indications for pregnant women with low back pain include assessing sacroiliac joint conditions.²⁵ Electrodiagnostic studies are useful complements to imaging for neural compression, correlating MRI findings with the clinical picture, although they are rarely required.⁴

A topic of special attention is the decision to terminate the pregnancy if spinal surgery is required, for which there is no single consensus. Some recommendations support continuing the pregnancy below 36 weeks before surgery for nucleus pulposus hernias with neurological involvement and performing the procedure in the left lateral decubitus position.⁷ However, this is different when it comes to requiring surgery for neoplastic conditions. Esmaeilzadeh et al.¹⁰ distinguish the surgical management of spinal tumors in the third pregnancy trimester between benign and malignant ones, recommending an elective cesarean section followed by spinal surgery in the former or, if feasible, performing spinal surgery during pregnancy. For malignant tumors requiring adjuvant therapy, these authors suggest an elective cesarean section before spinal surgery. In our case, the patient had a neoplasm with benign imaging characteristics, such as a myxopapillary ependymoma but a rare complication, like a tumor rupture with subarachnoid hemorrhage, made a multidisciplinary (neurosurgery, gynecology, and anesthesiology) approach and decided that interrupting the pregnancy at 33 weeks seemed the best option.

The patient's neurological disorders appeared suddenly and helped in the investigation of an extremely rare condition, such as an intraspinal myxopapillary ependymoma. The patient had a good recovery given the early and planned

treatment, the complete tumor resection, and her proper rehabilitation.

Conflict of Interest

None declared.

References

- Whiles E, Shafafy R, Valsamis EM, et al. The Management of Symptomatic Lumbar Disc Herniation in Pregnancy: A Systematic Review. *Global Spine J* 2020;10(07):908–918. Doi: 10.1177/2192568219886264
- Casagrande D, Gugala Z, Clark SM, Lindsey RW. Low Back Pain and Pelvic Girdle Pain in Pregnancy. *J Am Acad Orthop Surg* 2015;23(09):539–549. Doi: 10.5435/JAAOS-D-14-00248
- Arce-Gálvez L, Méndez-Vega DA, Mancera-Álzate JM, Benavidez-Ramírez A, Rodríguez-Vélez LM. Dolor lumbar en el embarazo, aspectos fisiopatológicos y tratamiento. *Rev Chil Obstet Ginecol*. 2022;87(02):111–121. Doi: 0.24875/rechog.22000004
- Sehmbi H, D'Souza R, Bhatia A. Low Back Pain in Pregnancy: Investigations, Management, and Role of Neuraxial Analgesia and Anaesthesia: A Systematic Review. *Gynecol Obstet Invest* 2017;82(05):417–436. Doi: 10.1159/000471764
- Hall H, Lauche R, Adams J, Steel A, Broom A, Sibbritt D. Healthcare utilisation of pregnant women who experience sciatica, leg cramps and/or varicose veins: A cross-sectional survey of 1835 pregnant women. *Women Birth* 2016;29(01):35–40. Doi: 10.1016/j.wombi.2015.07.184
- Kokkalaki D, Kesharwani P. Prevalence of Sciatica in Pregnancy and Its Impact on Quality of Life - A Cross Sectional Study. *IJSHR* 2022;7(02):127–136. Doi: 0.52403/ijshr.20220419
- Ardaillon H, Laviv Y, Arle JE, Kasper EM. Lumbar disk herniation during pregnancy: a review on general management and timing of surgery. *Acta Neurochir (Wien)* 2018;160(07):1361–1370. Doi: 10.1007/s00701-017-3098-z
- Butenschoen VM, Hitscherich H, Eicker SO, et al. Spine surgery in pregnant women: a multicenter case series and proposition of treatment algorithm. *Eur Spine J* 2021;30(04):809–817. Doi: 10.1007/s00586-021-06726-2
- Pavlidis NA. Coexistence of pregnancy and malignancy. *Oncologist* 2002;7(04):279–287 [published correction appears in *Oncologist* 2002;7(6):585]
- Esmaeilzadeh M, Hong B, Polemikos M, et al. Spinal Emergency Surgery During Pregnancy: Contemporary Strategies and Outcome. *World Neurosurg* 2020;139:e421–e427. Doi: 10.1016/j.wneu.2020.04.019
- Meng T, Yin H, Li Z, et al. Therapeutic strategy and outcome of spine tumors in pregnancy: a report of 21 cases and literature review. *Spine* 2015;40(03):E146–E153. Doi: 10.1097/BRS.0000000000000719
- Vougioukas VI, Kyroussis G, Gläsker S, Tatagiba M, Scheuffer KM. Neurosurgical interventions during pregnancy and the puerperium: clinical considerations and management. *Acta Neurochir (Wien)* 2004;146(12):1287–1291, discussion 1291–1292. Doi: 10.1007/s00701-004-0354-9
- Reni M, Gatta G, Mazza E, Vecht C. Ependymoma. *Crit Rev Oncol Hematol* 2007;63(01):81–89. Doi: 10.1016/j.critrevonc.2007.03.004
- Almatrafi FR, Aseeri AM, Alqahtani MF, et al. Myxopapillary Ependymoma—a Case Report of Rare Multicentric Subtype and Literature Review. *Med Arh* 2023;77(02):150–154. Doi: 10.5455/medarh.2023.77.150-154
- Bagley CA, Wilson S, Kothbauer KF, Bookland MJ, Epstein F, Jallo GI. Long term outcomes following surgical resection of myxopapillary ependymomas. *Neurosurg Rev* 2009;32(03):321–334, discussion 334. Doi: 10.1007/s10143-009-0190-8
- Leidinger W, Meierhofer JN, Ullrich V. [Unusual complication after combined spinal/epidural anaesthesia]. *Anaesthesist* 2003;52(08):703–706. Doi: 10.1007/s00101-003-0525-8

- 17 Domazet I, Pašalić I, Nemir J, Peterković V, Vukić M Predictors of Functional Outcome after Spinal Ependymoma Resection. *J Neurosci Rural Pract* 2018;9(03):354–358. Doi: 10.4103/jnrp.jnrp_56_18
- 18 Bitterman J, Donovan J, Lam M. Diagnosis and rehabilitation of a pregnant woman with spinal cord disorder due to spinal cord tumor. *Spinal Cord Ser Cases* 2019;5:8. Doi: 10.1038/s41394-019-0153-4
- 19 Smyth RM, Aflaifel N, Bamigboye AA. Interventions for varicose veins and leg oedema in pregnancy. *Cochrane Database Syst Rev* 2015;2015(10):CD001066. Doi: 10.1002/14651858.CD001066.pub3
- 20 Sabino J, Grauer JN. Pregnancy and low back pain. *Curr Rev Musculoskelet Med* 2008;1(02):137–141. Doi: 10.1007/s12178-008-9021-8
- 21 Lum M, Tsiouris AJ. MRI safety considerations during pregnancy. *Clin Imaging* 2020;62:69–75. Doi: 10.1016/j.clinimag.2020.02.007
- 22 Gatta G, Di Grezia G, Cuccurullo V, et al. MRI in Pregnancy and Precision Medicine: A Review from Literature. *J Pers Med* 2021;12(01):9. Doi: 10.3390/jpm12010009
- 23 American College of Radiology (ACR) Practice parameter for performing and interpreting magnetic resonance imaging (MRI). [Internet]. 2022 [cited 2024-Jun-21]. Available from: <https://www.acr.org/-/media/ACR/Files/Practice-Parameters/MR-Perf-Interpret.pdf>
- 24 Committee Opinion No. 723: Guidelines for Diagnostic Imaging During Pregnancy and Lactation. *Obstet Gynecol* 2017;130(04):e210–e216. Doi: 10.1097/AOG.0000000000002355. [published correction appears in *Obstet Gynecol*. 2018 Sep;132(3):786. doi: 10.1097/AOG.0000000000002858]
- 25 Vleeming A, Albert HB, Ostgaard HC, Sturesson B, Stuge B. European guidelines for the diagnosis and treatment of pelvic girdle pain. *Eur Spine J* 2008;17(06):794–819. Doi: 10.1007/s00586-008-0602-4