Regional anesthesia in Marfan syndrome, not all dural ectasias are the same: a report of two cases

Anesthésie régionale dans le syndrome de Marfan; toutes les ectasies de la dure-mère ne se ressemblent pas : une étude de deux cas

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Abstract

Background The anesthetic management of women with Marfan syndrome and dural ectasia undergoing Cesarean delivery remains controversial. We present two cases of patients with Marfan syndrome and dural ectasia where neuraxial anesthesia was used successfully.

Case 1 A 31-yr-old G1P0 female with Marfan syndrome presented for elective Cesarean delivery at 35^{3/7} weeks' gestation. The magnetic resonance imaging of her spine revealed significant dural ectasia in the lumbosacral area with a mean lumbar dural sac area of 4.71 cm². Intrathecal administration of 0.75% hyperbaric bupivacaine 9 mg produced only limited perineal analgesia. The epidural catheter was titrated, and 0.5% bupivacaine 150 mg were required to achieve a T4 sensory level.

Case 2 A 34-yr-old G1P0 female with Marfan syndrome presented for elective Cesarean delivery at 37 weeks' gestation. The intrathecal administration of 0.75%

hyperbaric bupivacaine 13.5 mg produced a T5 sensory level. Magnetic resonance imaging of her spine prior to discharge confirmed the diagnosis of moderate dural ectasia with a mean lumbar dural sac area of 3.61 cm².

Discussion The two patients described in this report responded differently to spinal anesthesia, most likely based on the severity of their dural ectasia. Although preoperative magnetic resonance imaging may help to identify patients at risk for a failed spinal, we suggest considering a combined spinal-epidural technique in cases of dural ectasia.

Résumé

Contexte La prise en charge anesthésique de femmes ayant un syndrome de Marfan et une ectasie de la dure-mère au cours d'un accouchement par césarienne reste un sujet controversé. Nous présentons le cas de deux patientes ayant un syndrome de Marfan et une ectasie de la dure-mère chez qui une anesthésie neuraxiale a été réalisée avec succès.

Cas n° 1 Une femme de 31 ans, G1P0, ayant un syndrome de Marfan a été admise pour accouchement par césarienne programmé à 35 semaines et 3 jours de grossesse. L'imagerie par résonance magnétique de la colonne vertébrale a révélé une ectasie significative de la dure-mère dans la région lombosacrée avec une surface moyenne du sac dural lombaire de 4,71 cm². L'administration intrathécale de 9 mg de bupivacaine hyperbare à 0,75 % n'a procuré qu'une analgésie périméale limitée. La dose du cathéter péri-dural a été titrée et 150 mg de bupivacaine 0,5 % ont été nécessaires pour atteindre le niveau sensitif D4.

Cas n° 2 Une femme de 34 ans, G1P0, ayant un syndrome de Marfan a été admise pour accouchement par césarienne programmé à 37 semaines de grossesse. L'administration intrathécale de 13,5 mg de bupivacaine

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hyperbare à 0,75 % a permis d'atteindre le niveau sensitif D5. Une IRM de la colonne vertébrale, effectuée avant le congé de la patiente a confirmé le diagnostic d'ectasie modérée de la dure-mère, avec une surface moyenne du sac dural lombaire de 3,61 cm².

Discussion Les deux patientes décrites dans cet article ont répondu différemment à la rachianesthésie, le plus probablement en rapport avec la gravité de leur ectasie durale. Bien que l'imagerie par résonance magnétique préopératoire puisse aider à identifier les patientes à risque d'échec de la rachianesthésie, nous suggérons qu'une technique combinée de rachianesthésie et d'anesthésie péridurale soit envisagée dans les cas d'ectasie de la dure-mère.

Marfan syndrome is an autosomal dominant connective tissue disorder with a reported incidence of one in 3,000-5,000 individuals.¹ It is caused by mutation in the fibrillin-1 gene located on chromosome 15.² The cardinal features occur in the skeletal, ocular, and cardiovascular systems, but other organs can also be affected, including the skin, lungs, and dura.

The most important complication of Marfan syndrome is the progressive dilatation of the aortic root and ascending aorta, leading to aortic valve incompetence and aortic dissection.³ Another major feature in the diagnostic criteria⁴ is dural ectasia (DE), defined as the widening of the spinal canal, indentation of the vertebral body posteriorly, thinning of the cortex and laminae as a result of long-standing pressure, widening of the neural foraminae, or presence of meningocele.⁵ The prevalence of DE in patients with Marfan syndrome is estimated to be from 63-92%.⁶ Although DE can occur at different levels of the lumbar spine, it is most common at the L5-S1 level.⁷

The anesthetic management of these patients can be challenging, and the literature regarding the management of these cases is scarce. Spinal anesthesia may result in undesirable hemodynamic changes in patients with significant cardiovascular disease and may fail as a result of an increased volume of lumbar cerebrospinal fluid (CSF).⁸ While epidural anesthesia provides more stable hemodynamics, it may be difficult to perform due to scoliosis, and it has been associated with failure.^{1,9}

We present two cases of patients with Marfan syndrome and associated DE. Both patients presented for elective Cesarean delivery where neuraxial anesthesia was used successfully but performed very differently. Both patients gave written informed consent for publication of this information.

Case 1

A 31-yr-old G1P0 female with clinical features suggestive of Marfan syndrome was referred for anesthesia consult at

34 weeks' gestation in anticipation of an upcoming Cesarean delivery. The diagnosis of Marfan syndrome was not confirmed by genetic testing; however, she had a typical body habitus: height 178 cm, weight 98 kg, and arachnodactyly. The echocardiogram prior to conception showed mild global left ventricular hypokinesia, bicuspid aortic valve with mild aortic regurgitation, mitral valve prolapse, and a dilated aortic root measuring 42 mm. The aortic root was assessed by cardiac magnetic resonance imaging (MRI) during the patient's pregnancy; it increased gradually and measured 48 mm at 32 weeks' gestation. Beta-blocker therapy was initiated for heart rate and blood pressure control. A spine image obtained as part of the cardiac MRI revealed the presence of DE involving the lumbosacral spine and extending into bilateral perineural cysts within the sacrum (Fig. 1). The anteroposterior diameter of the lumbar dural sac ranged from 1.8-2.2 cm with a mean lumbar dural sac area of 4.71 cm² (Table 1). There was a mild cervicothoracic scoliosis. Due to progressive dilatation of the aortic root, a Cesarean delivery was planned at 35^{3/7} weeks' gestation. The patient remained asymptomatic throughout her pregnancy and continued to belong to New York Heart Association (NYHA) Class I at the time of the Cesarean delivery. The Cesarean delivery was performed under a double-segment combined spinal-epidural (CSE) anesthetic. The patient was monitored with a noninvasive blood pressure cuff on each arm, five-lead electrocardiogram, pulse oximeter, and noninvasive cardiac output monitor (NICOMTM Cheetah Medical Inc, Portland, OR, USA). Pre-procedural ultrasound scanning of the patient's spine was performed, and the appropriate intervertebral spaces were identified. With the patient in the sitting position, an end-hole epidural catheter (Arrow FlexTip Plus[®]) was placed at the L3-4 interspace without any difficulty, and an electrical stimulation test¹⁰ was used to confirm its position. The spinal component of the CSE was introduced at the L4-5 interspace. We used 0.75% hyperbaric bupivacaine 9 mg, fentanyl 10 µg, and preservative-free morphine 100 µg. Fifteen minutes following the intrathecal injection, the patient had only limited perineal analgesia as assessed by pinprick sensation using a blunt 25G needle and cold sensation to ice. The epidural injection was carefully titrated over the next 15 min, and 30 mL of 0.5% isobaric bupivacaine were required to achieve a bilateral T4 sensory level. Baseline systolic blood pressure levels were maintained with phenylephrine boluses given in increments of 0.1 mg *iv* as required.

The remainder of the procedure was uneventful, and the patient delivered a live male infant with Apgar scores 9 and 9 at one and five minutes, respectively. Additional preservative-free morphine 2.5 mg was injected through the epidural catheter for postoperative pain control. The patient

Fig. 1 Case 1: (A) Sagittal view of the spine on a T2 weighted fat suppressed image demonstrating the widened spinal canal that is more pronounced in the lower thoracic and lumbar regions below the conus medullaris. (B) Axial cut at the L2 level on a T2 weighted scan with measurements of the width of the spinal canal in transverse and antero-posterior dimensions demonstrating widening of the spinal canal

Table 1 Dural sac dimensions by magnetic resonance imaging at L1-L5 levels

Level	Case 1			Case 2			Reference values ¹⁵
	Area (cm ²)	Antero-posterior diameter (cm)	Mediolateral diameter (cm)	Area (cm ²)	Antero-posterior diameter (cm)	Mediolateral diameter (cm)	Anteroposterior diameter (cm) with 95% confidence interval ()
L1	3.30	1.8	2.6	2.75	1.6	2.5	1.4(1.2 to 1.7)
L2	5.28	2.4	3.3	3.32	1.9	2.4	1.3(1.0 to 1.6)
L3	4.83	2.2	2.6	3.26	1.6	2.2	1.3(0.8 to 1.6)
L4	4.95	2.2	3.3	4.45	2.2	2.7	1.2(0.7 to 1.6)
L5	5.18	2.2	3.2	4.28	2.0	3.0	1.1(0.6 to 1.6)

was monitored on the labour and delivery floor for 24 hr and then transferred to the ward. On the fifth postoperative day, she underwent surgery for aortic root repair and was discharged home 12 days later.

Case 2

A 34-yr-old G1P0 female with a known diagnosis of Marfan's syndrome was referred for anesthesia consultation at 34^{2/7} weeks' gestation. She had a typical body habitus (height 185 cm, weight 56 kg) and a family history of "Marfanoid" phenotype. The patient had known progressive aortic root dilatation over the last six years; however, it was stable throughout her pregnancy and measured 45 mm. Beta-blocker therapy was maintained throughout her pregnancy. She was asymptomatic and continued to belong to NYHA Class I, and there was no spine imaging to confirm the presence of DE. Cesarean delivery at 37 weeks' gestation was planned. Ultrasound scanning was performed prior to neuraxial anesthesia to identify the appropriate

intervertebral space, and the patient was monitored with standard monitors as well as with NICOM. A two-segment CSE was planned. With the patient in the sitting position, the epidural catheter was inserted at the L2-3 level without any difficulty, and the correct position was confirmed with electric stimulation. The spinal anesthetic was injected at the L3-4 level and consisted of 0.75% hyperbaric bupivacaine 13.5 mg, fentanyl 10 µg, and preservative-free morphine 100 µg as per our standard practice. Fifteen minutes following the injection, a bilateral T5 sensory level was obtained. Surgery was uneventful, and the patient delivered a female infant with Apgar scores 9 and 9 at one and five minutes, respectively. The epidural catheter was not used for supplementary anesthesia during the surgery. The patient was monitored on the labour and delivery unit for 24 h and later transferred to the ward. She was discharged home on the fourth postoperative day without any complications. Prior to discharge, she underwent a spinal MRI, and the diagnosis of DE was confirmed (Fig. 2). The anteroposterior diameter of the lumbar dural sac ranged from 1.6-2.0 cm, with a mean lumbar dural sac area of 3.61 cm² (Table 1).

Fig. 2 Case 2: (A) Sagittal T2 Turbo spin echo sequence of the lumbar spine demonstrating significant dural ectasias with scalloping of the vertebral bodies of the entire lumbar spine and significant dilatation of the spinal canal. (B) Axial cut at the L2 level on a T1 weighted scan with measurements of the width of the spinal canal in transverse and antero-posterior dimensions demonstrating widening of the spinal canal. Note also the thinned pedicles as a typical finding in Marfan syndrome

Discussion

Our results confirm the successful use of neuraxial anesthesia in Marfan patients with DE; however, our results also suggest that the degree of DE may limit the efficacy of spinal anesthesia and require the use of epidural anesthesia.

The etiology of DE is unclear. The predominant presence of DE at the lowest vertebra supports the theory that the development of ectasia is influenced by hydrostatic pressure on a weakened dura.¹¹ Patients are usually asymptomatic, but some may develop lower back pain, headache, proximal leg pain, weakness and numbness above and below the knee, and genital/rectal pain.⁷ Dural ectasia has also been reported in patients with neurofibromatosis type 1, Ehler-Danlos syndrome, ankylosing spondylitis, trauma, tumours, and scoliosis.^{12–14}

The severity of DE can be evaluated by computed tomography or MRI using either quantitative (actual dural sac measurements) or qualitative (evaluating the prevalence of different features of DE, i.e., scalloping) criteria. There are no universally accepted quantitative criteria for diagnosing DE, and various methods have been used, including evaluation of dural sac ratio,¹⁵ assessment of dural sac diameter at S1 and L4 levels, nerve root sleeve diameter,¹⁶ and lumbar pedicle width.¹⁷ According to Fattori *et al.*,⁵ the degree of DE can be classified as mild, moderate, or severe based on radiologic imaging. The severity of this condition is suggested to be age related, which supports the theory that an ongoing pulsatile pressure of the cerebrospinal fluid on the lumbar and sacral canal causes progressive ballooning of a congenitally defective dural wall.¹⁸

The anesthetic literature on parturients with Marfan syndrome and confirmed DE is scarce. In three case reports, neuraxial technique was attempted but failed to produce a block adequate for surgical procedure, and in one case report with radiologically confirmed DE, epidural anesthesia was used successfully for elective Cesarean delivery (Table 2).

On the other hand, there are case reports where neuraxial anesthesia was used successfully in parturients with Marfan syndrome undergoing both Cesarean and vaginal delivery; however, diagnosis of DE was neither suspected clinically nor confirmed radiologically.^{19,20}

The presence of DE may explain variability in the volume of cerebrospinal fluid, and as a result, different response to spinal anesthesia. Greater than normal volume of cerebrospinal fluid in the lumbar theca is postulated to restrict the spread of intrathecally injected local anesthetics and cause a “failed” spinal.²¹ Good correlations have been observed between the volume of lumbar CSF, measured by MRI, and the intrathecal spread of drugs in non-pregnant patients.^{22,23}

Our patients exhibited different degrees of DE, and their dural sac dimensions at various lumbar levels are presented in Table 1. The anteroposterior dural sac diameters were measured perpendicular to the long axis of the dural sac at the midcorpus and lower endplate level of L1 through S1. Reference values were obtained from Oosterhof *et al.*, where the authors measured dural sac dimensions in 44 healthy volunteers.¹⁵ Although, the difference in the dosing of spinal bupivacaine in our two cases makes the comparison between the effect difficult, in our view, the reason for a “failed” spinal in Case 1 was the greater severity of the patient’s DE. Even a low dose of hyperbaric bupivacaine is expected to result in a T3–4 sensory block in healthy parturients.²⁴

It is likely that the dimensions of the epidural space in patients with DE are also enlarged along with the dural sac. Despite the dural sac being a soft-tissue structure, it exerts sufficient pressure on the vertebra over time to cause bony changes of the spine.¹⁷ Therefore, an increased circumference of the bony canal may result in an enlarged epidural space, requiring a higher volume of local anesthetic to achieve sufficient levels, as was observed in Case 1. Further observations, however, are necessary to confirm this hypothesis.

Table 2 Summary of published cases of neuraxial anesthesia for Cesarean delivery in parturients with Marfan syndrome and dural ectasia

Reference	Anesthetic technique	Comments
Lacassie <i>et al.</i> ⁸ (1 st case)	Continuous spinal catheter inserted at L4-5 interspace by midline approach in left lateral position and left at 4 cm in the intrathecal space. Incremental top up of the catheter with 21 mg of 0.5% bupivacaine. Highest sensory block T10. Aspiration of CSF was confirmed at all times.	Intrathecal catheter top up was stopped for fear of potential neurologic injury. Fifty minutes later, the patient had a T12 sensory block. General anesthesia was induced for Cesarean delivery. A CT scan revealed ectatic dural sac. Patient had a history of T1-L3 scoliosis repair.
Lacassie <i>et al.</i> ⁸ (2 nd case)	Spinal catheter inserted in left lateral position under CT fluoroscopic guidance at L5-S1 level. The catheter was left at 5 cm in the subarachnoid space. Patchy sensory block to T7 on the left and L1 on the right after 10 mg of incremental top up of 0.5% bupivacaine and 7.5 mg of 0.75% bupivacaine. Aspiration of CSF was confirmed at all times.	General anesthesia was induced due to the patchy block. The images showed dural ectasia and severe kyphoscoliosis. Spinal catheter was used for postoperative analgesia and removed 18 hr after delivery. On day 3, patient complained of most likely a postdural puncture headache but refused the blood patch. The headache resolved on postoperative day 9.
Tan <i>et al.</i> ⁹	Epidural catheter inserted at L3-4 level and failed to produce an adequate surgical block for Cesarean delivery after 15 ml of 2% lidocaine with epinephrine 1:200,000. Subarachnoid block at L3-4 level with 12 mg bupivacaine produced initial bilateral T4 block; however, it was patchy 10 min into the procedure.	General anesthesia was induced due to the patchy block. Postpartum course was complicated with postdural puncture headache. The authors reported constant “loss of resistance” with increasing needle depth. No follow-up imaging reported to confirm suspected diagnosis of dural ectasia.
Kim <i>et al.</i> ²⁶	Epidural catheter was inserted at L3-4 interspace in lateral position. A total dose of 27 mL of 2% lidocaine with epinephrine 1:200,000 and 100 µg of fentanyl was injected incrementally to produce a T4 block bilaterally 20 min after injection.	Uneventful Cesarean delivery. Epidural catheter was used for postoperative analgesia for three days.

CSF = cerebrospinal fluid; CT = computed tomography

Although DE is not an absolute contraindication for epidural anesthesia, caution has been advised in its use for patients with Marfan syndrome and associated DE because of the theoretical risk of accidental dural puncture²⁰ and CSF leak; however, when performed carefully by an experienced anesthesiologist, many benefits of epidural anesthesia, including minimal hemodynamic fluctuations and excellent postoperative pain control, can outweigh the potential risk. There has been some suggestion in the literature to perform an epidural in the lateral decubitus position to minimize the chance of accidental dural puncture. In this position, the ectatic dural sac is thought to be less extended in the region of needle insertion.²⁰ We postulated that the lateral position may hinder the performance of the neuraxial technique and elected to perform the procedure with both patients in the sitting position. Although we did not acknowledge abnormal “loss of resistance” due to structural changes in the ligamentum flavum in either patient, this phenomenon has been suggested as a likely cause of accidental dural puncture.⁹

Patients with Marfan syndrome and associated scoliosis and DE pose many challenges to the anesthesiologist; therefore, we suggest a few details of our technique that could be useful to optimize the neuraxial technique and reduce the risk of complications. This assumption, however, requires confirmation in future observations. A pre-procedural ultrasound assessment of the spine may correctly identify the target interspace, determine the best interspace and insertion point in patients with scoliosis, and determine the depth to the epidural

space; thus, it can potentially minimize the risk of a dural puncture. Electrical stimulation of the epidural catheter may be particularly useful in testing the catheter for double-segment CSEs and also in ascertaining the correct positioning of the catheter in patients with scoliosis.²⁵

In summary, we reported two cases of parturients with Marfan syndrome and DE undergoing elective Cesarean delivery. Each patient responded differently to spinal anesthesia, most likely based on the severity of their DE. Although a preoperative MRI may help to identify patients at risk for a failed spinal, we suggest considering a CSE technique in cases of DE.

Conflict of interest None of the authors has a conflict of interest.

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