Evidence of spinal stiffening following fusionless bipolar fixation for neuromuscular scoliosis: a shear wave elastography assessment of lumbar annulus fibrosus

Mathilde Gaume[1,2,5] · Klervie Loiselet[3] · Hedi Chekir[3] · Tristan Langlais[1,2] · Nathalie Boddaert[3] · Sarah Stricker[4] · Stéphanie Pannier[1] · Wafa Skalli[2] · Lotfi Miladi[1] · Claudio Vergari[2]

Abstract

Objectives There are no established criteria for stiffness after fusionless surgery for neuromuscular scoliosis (NMS). As a result, there is no consensus regarding the surgical strategy to propose at long-term follow-up. This study reports the first use of shear wave elastography for assessing the mechanical response of lumbar intervertebral discs (IVDs) after fusionless bipolar fixation (FBF) for NMS and compares them with healthy controls. The aim was to acquire evidence from the stiffness of the spine following FBF.

Patients and methods Nineteen NMS operated on with FBF (18 ± 2y at last follow-up, 6 ± 1 y after surgery) were included prospectively. Preoperative Cobb was 89 ± 20° and 35 ± 1° at latest follow-up. All patients had reached skeletal maturity. Eighteen healthy patients (20 ± 4 y) were also included. Shear wave speed (SWS) was measured in the annulus fibrosus of L3L4, L4L5 and L5S1 IVDs and compared between the two groups. A measurement reliability was performed.

Results In healthy subjects, average SWS (all disc levels pooled) was 7.5 ± 2.6 m/s. In NMS patients, SWS was significantly higher at 9.9 ± 1.4 m/s ($p < 0.05$). Differences were significant between L3L4 (9.3 ± 1.8 m/s vs. 7.0 ± 2.5 m/s, $p = 0.004$) and L4L5 (10.3 ± 2.3 m/s vs. 7.1 ± 1.1 m/s, $p = 0.0006$). No difference was observed for L5S1 ($p = 0.2$). No correlation was found with age at surgery, Cobb angle correction and age at the SWE measurement.

Conclusions This study shows a significant increase in disc stiffness at the end of growth for NMS patients treated by FBF. These findings are a useful adjunct to CT-scan in assessing stiffness of the spine allowing the avoidance of surgical final fusion at skeletal maturity.

Keywords Shear wave elastography · Neuromuscular scoliosis · Fusionless bipolar fixation · Stiffness · Elastic properties

Introduction

Neuromuscular scoliosis (NMS) is due to a lack of muscular control caused by a neurological or degenerative muscular condition. When conservative treatment by bracing fails to control curve progression, patients may require surgery at an early age. In such situations, different growth-friendly techniques have been described [1]. These fusionless techniques allow deformity correction while preserving spinal and thoracic growth. At skeletal maturity, which corresponds to the endpoint of the lengthening protocol, or even frequently before, a definitive posterior spinal fusion (PSF) is usually performed. PSF consists in fusing all vertebral levels with a posterior metallic instrumentation that stiffens the spine and stops almost any intervertebral movement. However, PSF stops also spinal and thoracic growth, and it is at high risk of complications,

✉ Mathilde Gaume
mathilde.gaume@aphp.fr

1 Pediatric Orthopedic Surgery Department, Necker University Hospital, APHP, University of Paris-Cité, Paris, France

2 Arts Et Métiers Institute of Technology, Institut de Biomécanique Humaine Georges Charpak, Université Sorbonne Paris Nord, Paris, France

3 Pediatric Radiology Department, Necker University Hospital, APHP, University of Paris-Cité, Paris, France

4 Neurosurgery Department, Necker University Hospital, APHP, University of Paris-Cité, Paris, France

5 Necker Hospital, 149 Rue de Sevres, 75015 Paris, France

including haemorrhage [2], infections and mechanical complications [3]. Our team used a bilateral fusionless bipolar fixation (FBF), which became an alternative technique for PSF in NMS scoliosis in our department. This technique is based on a telescopic construct including two 5.5-mm-diameter rods on each side bridging the curve and linking two proximal–distal anchors. It is a modular construct which follows spinal growth and allows further gradual correction of residual deformities. The minimally invasive approach minimizes the risk of early fibrosis and auto-fusion, allowing the telescopic construct lengthening for longtime to preserve growth until skeletal maturity. Over the long term with a follow-up of more than 10 years for the first cases, a progressive stiffening was supposed by the continued stability of the spinal correction at skeletal maturity, suggesting that PSF was unnecessary [4, 5].

It was hypothesized that the origin of this stiffening could be i) a peri-implant bone growth, ii) bone growth within the disc, or iii) a general stiffening of the disc, possibly due to partial mineralization. The present study aims to elucidate this last aspect.

Bony structures of the spine are typically evaluated using computed tomography (CT) and spontaneous bone fusion after FBF has been recently published [6]. MRI has been used to assess disc stiffness, its access is difficult in clinical routine, and it requires a long period in an uncomfortable position, especially in neuromuscular patients.

Ultrasound imaging modalities are non-invasive, known for their rapidity, safety and portability. Particularly, shear wave elastography (SWE) is an imaging technique based on the propagation velocity of the ultrasonic-induced shear wave to quantify the soft tissue elasticity or stiffness [7, 8]. SWS reflects the intrinsic mechanical properties of the tissue, and a higher SWS is associated with a stiffer tissue [9]. It has been previously applied to evaluate IVD in adolescent idiopathic scoliosis, and it showed that IVDS in patients had higher shear wave speed (SWS, i.e. stiffer tissue) than asymptomatic controls [10, 11]. Furthermore, it was shown that this increased SWE can detect changes in IVD, and in particular disc SWS in idiopathic scoliosis tended to normalize one year after fusion surgery [12].

This study reports the first use of SWE for assessing the mechanical properties of IVD in NMS after FBF and compares them with asymptomatic controls. The aim was to acquire evidence from the stiffness of the spine following FBF at long-term follow-up.

Material and methods

Subjects

Data were collected prospectively from our department within follow-up clinical routine, between January 2021 and June 2022. Parents and children were informed about the measurement protocol and consented to participating before inclusion. Parents signed informed consent, which was approved by the ethics committee (IDRCB 2020-A03598-31, ClinicalTrials.gov Identifier: NCT04969770).

Nineteen NMS patients who underwent FBF from T1 to the pelvis (Fig. 1) were included prospectively in this study (7 girls and 12 boys). Preoperative Cobb angle was 89° ± 20, and 35° ± 1 at last follow-up. They were 11 ± 2 years old at initial surgery and 18 ± 2 years old when SWE was performed. Twelve had cerebral palsy, six were syndromic, and one suffered from spinal muscular atrophy type 2. All were non-ambulatory and had reached skeletal maturity at last follow-up. Patients with less than 5 years postoperative follow-up were excluded.

Eighteen asymptomatic subjects were also included as control group (9 girls and 9 boys, mean age 20 ± 3y).

Fig. 1 The bipolar fusionless technique anchored proximally by a double hooks claw and distally by ilio-sacral screws

Healthy children were included if they had no history of spinal disease, and a systematic clinical examination was performed by a spine surgeon to rule out any diagnosis of scoliosis.

Protocol

Shear-wave speed (SWS) was measured in all included subjects following a standardized protocol determined for lumbar IVD in children and adolescents' idiopathic scoliosis [13].

Patients were in supine position and the probe was placed on the abdomen to image the spine with an anterior approach. The thinness of the abdominal wall in children allows a good quality ultrasound exploration, which is much easier than in adults. It should also be noted that in the operated group, the scoliosis brings the lumbar spine forward, and therefore even closer to the anterior abdominal wall.

Measurements were obtained using a Canon Aplio i800 system [14]. The probe we used was the i8CX1 (or PVI 475BX) which is a convex probe with a frequency range between 1 and 8 MHz (Mode Multi, Range 0–75 kPa). The probe was pushed to 2–3 cm from the disc. The aortic bifurcation was localized to determine the L4 vertebral level.

Ultrasound examination started with a sagittal section under the umbilicus, which allowed to locate the vertebral bodies and the intervertebral discs. The vertebrae are identified by a very hyperechoic linear interface with a posterior shadow cone, in connection with the cortical anterior wall (Fig. 2). The discs are hypoechoic and allow ultrasound to pass posteriorly.

After a 90° rotation of the probe, the operator switched in axial section in the plane of an intervertebral disc. The medullary canal with the roots can be seen behind the disc. The posterior joints are also visible laterally (Fig. 3). The probe was subsequently moved up or down to measure SWS in the annulus fibrosus of L3–L4, L4–L5 and L5–S1 IVDs [15]. For L5-S1 disc, a greater inclination of the probe was necessary to be in the plane of the disc. Children with a transitional anomaly were excluded from both groups.

For each IVDs (L3–L4, L4–L5 and L5–S1), three circular regions of interest (ROIs) of 8 mm diameter were defined in the periphery of the intervertebral disc at the annulus (Fig. 4). This procedure was repeated three times, and the average was calculated to obtain a single SWS value.

Measurement reliability was assessed on a subset of the first five NMS and healthy patients by two confirmed radiologists. Then, only one of them according to his availability performed the subsequent measures.

Statistics

Intra-operator repeatability and inter-operator reproducibility were determined according to ISO 5725 standard [16]. Five sets of three measurements were repeated by two trained radiologists. The probe was repositioned after each set of measurements. Repeatability and reproducibility were calculated in terms of standard deviation of uncertainty. Intraclass correlation coefficient (ICC) was determined both intra- and inter-operator; an ICC greater than 0.75 was considered to signify good agreement.

Differences were analysed with Wilcoxon rank sum tests and correlations were quantified with Spearman's rank correlation coefficient, since variable did not follow normal distributions (Shapiro–Wilk test).

Differences between NMS and healthy groups were analysed with Mann–Whitney tests and differences between vertebral levels with Kruskal–Wallis tests. Correlations were quantified with Spearman's rank correlation coefficient.

Significance was $p < 0.05$. Analyses were performed using Microsoft Excel 2016.

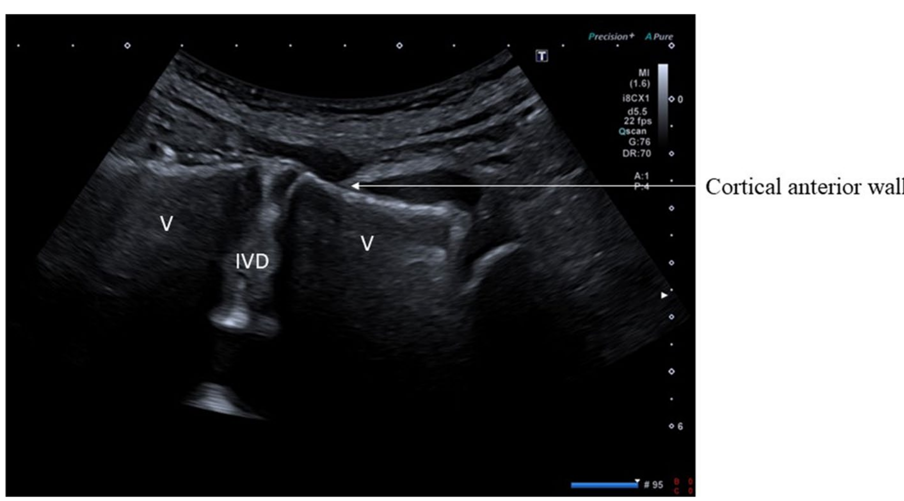

Fig. 2 Sagittal ultrasound section. V = vertebrae (V); IVD = intervertebral discs

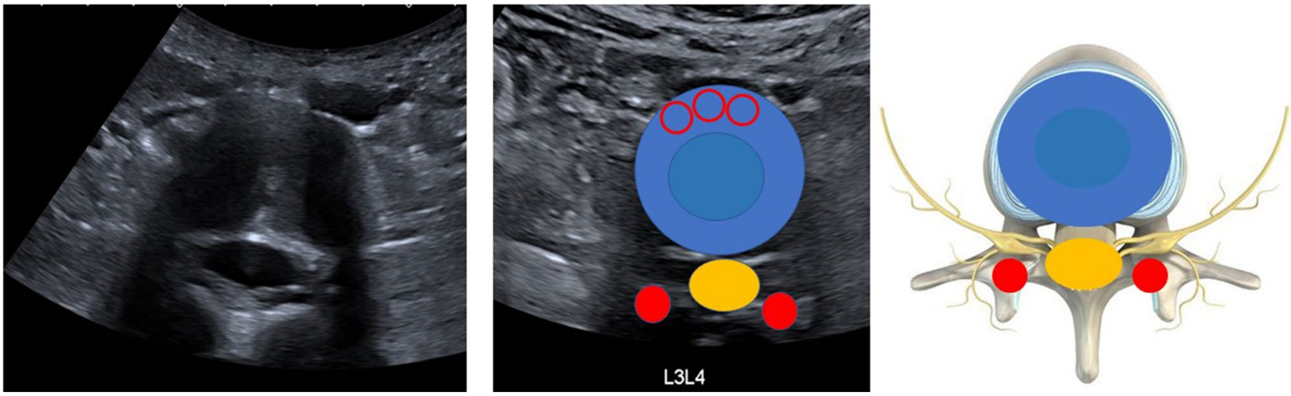

Fig. 3 Axial ultrasound section in the plane of an intervertebral disc. The medullary canal with the roots can be seen behind the disc. The posterior joints are also visible laterally. Disc in blue, spinal canal in yellow, post-articular in red. Red circles: Area of interest in the annulus fibrosus

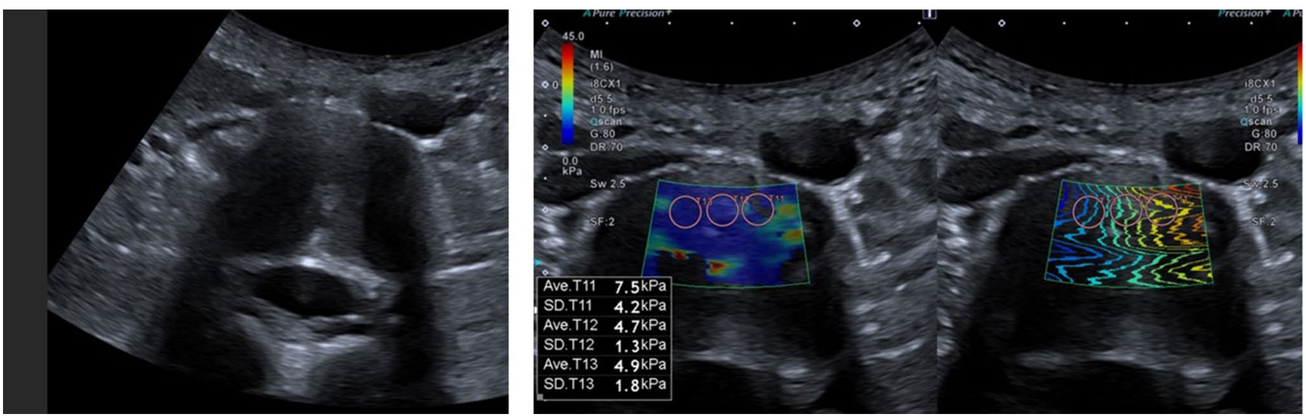

Fig. 4 Ultrasound and corresponding elastogram with three circular regions of interest defined in the periphery of the intervertebral disc at the annulus

Results

Intra-operator repeatability was 4.3%, and inter-operator reproducibility was 5.2%. Intraclass correlation coefficient was higher than 0.8 for each operator, showing "good agreement" for NMS patients and healthy patients (mean ICC = 0.89 and 0.91, respectively).

On average, the investigation lasted less than 30 min per patient, including the installation from the wheelchair to the examination table.

Figure 5 reports SWS in annulus fibrosus at the three-disc levels for the two groups (asymptomatic and NMS).

Healthy group SWS

There was no significant difference between levels L3–L4 (7.05 ± 1.3 m/s), L4–L5 (7.3 ± 1.1 m/s) and L5-S1 (8.5 ± 1.6 m/s, $p > 0.05$), Fig. 5), so values were pooled to obtain an overall average of 7.5 ± 2.6 m/s. The difference in average SWS between girls and boys was not significant ($p = 0.2$). No correlation of SWS was found with age.

NMS group SWS

There was no significant difference between levels L3–L4 (9.3 ± 1.8 m/s), L4–L5 (10.3 ± 2.3 m/s) and L5-S1 (10.15 ± 2.5 m/s, $p > 0.05$, Fig. 5), and overall average SWS was 9.9 ± 1.4 m/s. No correlation was observed with age at surgery, gender, age at the SWE measurement, time between surgery and SWE measurement, and Cobb angle correction.

Comparison between groups

Differences between NMS and healthy subjects was significant at L3L4 ($p = 0.004$) and L4L5 disc levels ($p = 0.0006$), but not at L5S1 ($p = 0.3$). Difference between overall averages was significant ($p = 5.627E\text{-}6$).

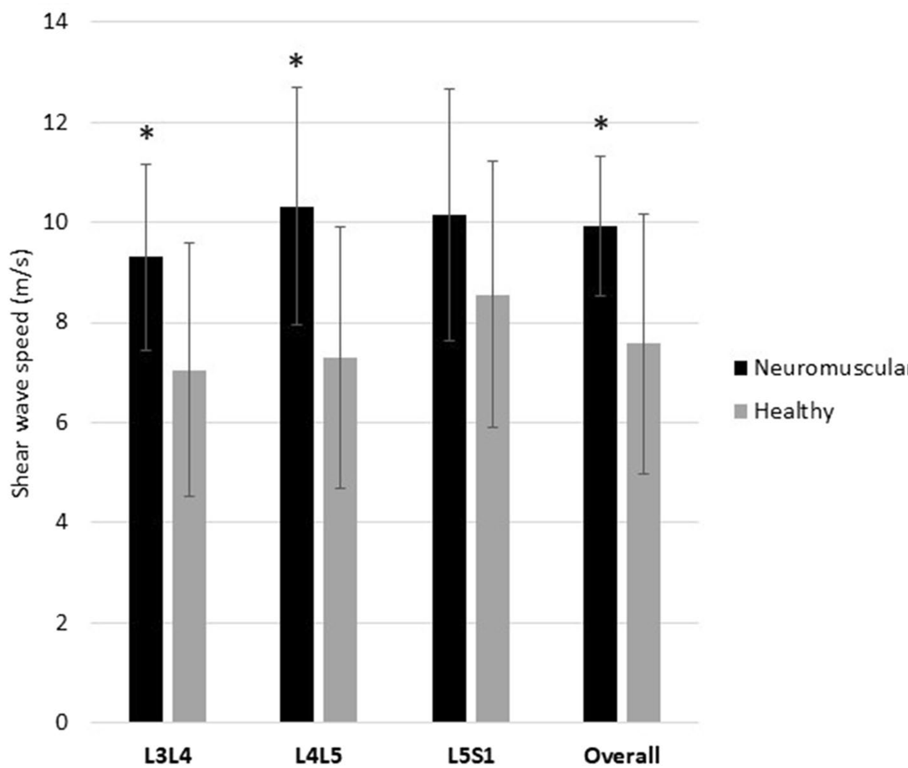

Fig. 5 Shear wave speed comparison in annulus fibrosus at the three-disc levels for the two groups

Discussion

The avoidance of PSF at the end of lengthening programme of growth guidance techniques in NMS has been recently an active area of research. PSF is indeed often regarded by patients as the conclusion of a long burden. Poe-Kochert et al. [17] reported 20% of unplanned reoperation after PSF following traditional growing rods program.

In a series of 100 NMS treated by FBF [18], PSF initially planned at skeletal maturity was not performed for any patients. PSF was avoided according to the stable correction of spinal and pelvic deformities observed during follow-up, and by the strength of this type of fixation [19, 20]. This stability over time suggested a progressive spinal stiffening following FBF. Thus, the spinal growth was possible in the first years of surgery, and the gradual increase in spinal stiffening made it possible to maintain the correction over time.

However, evidence of spinal stiffening following FBF for NMS has never been analysed. In particular, the role of the intervertebral disc (IVD) in this stiffening process is not yet clear.

In this paper, SWE was used to obtain information on lumbar IVD mechanical properties between 5 and 8.1 years after surgery, to shed some light on spinal stiffening after FBF in NMS.

This recent imaging procedure could be particularly attractive for NMS patients, who have difficulties to lie down for a long time in an CT scan or magnetic resonance imaging, because of the spinal deformity or abnormal movements related to their pathology. Overall, measurement time was much shorter than a typical CT scan, and more comfortable for the patient. The measurement reliability indicated that it is feasible in this population with intra-operator repeatability of 4.3% and inter-operator reproducibility of 5.2%.

Until now, SWE has been used in NMS patients for the study of paravertebral muscles [21, 22] but has never been applied to lumbar IVD, which is already well studied in patients with adolescent idiopathic scoliosis (AIS). Langlais et al. [10] showed that AIS presented a stiffer annulus fibrosus than asymptomatic adolescents, with SWSs of 3.5 ± 0.3 m/s vs. 3.0 ± 0.3 m/s, respectively. Values obtained in this study were much higher, which is probably due to the different technologies employed in the two studies (Canon Aplio i800 system in the present series versus Aixplorer Supersonic Imagine).

Therefore, it is important to keep in mind that SWS disc measurement can be very sensitive to the device that is used, and comparison to values obtained with different machines is not relevant, even if the measurement with each machine appears reliable, the technical differences induce a bias of comparison.

However, it can be noted that idiopathic scoliotic patients with no surgery showed a SWS 1.2 times higher than controls, while SWS in this study was 1.3 times higher than controls. This difference is indicative of the fact that idiopathic

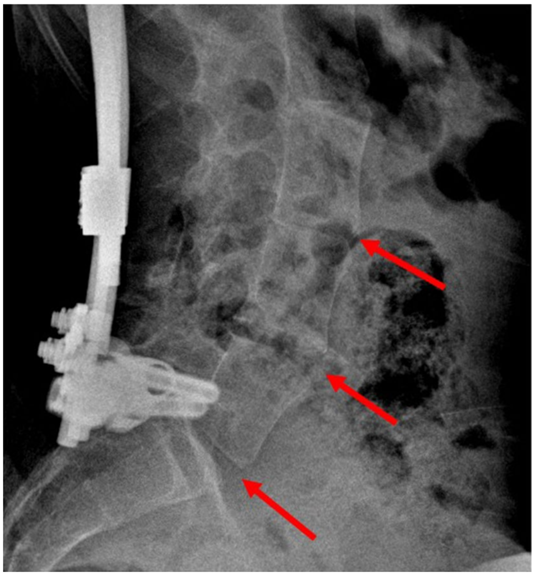

Fig. 6 The absence of disc ossification permitted to obtain an elastographic signal through the anterior approach

scoliosis patients present stiff but mobile spine, while NMS patients show stiffer spine, which raises the question of some degree of autofusion. Besides, a complete ossification of the disc would have made the measurement impossible, since the elastographic signal would not be obtained in bone (Fig. 6).

Unlike previous work on IVD ultrasonography and elastography, microstructural features of the disc were not visible in the present work. This is probably due to the use of a low-frequency probe, which resulted in a spatial resolution too low to distinguish the lamellar structure of the annulus. Assuming an average frequency of 3 MHz and a speed of sound in the annulus fibrosus of 1600 m/s, the spatial resolution should be about 0.5 mm, whereas lamellae in adolescents have a thickness in the order of 0.2 mm [23].

Contrary to Deviren et al. [24], no significant positive correlation was found between the spinal curvature severity and the stiffening of the spine. Vergari et al. [15] compared SWE in AIS before and after surgical intervention, showing that discs outside the fusion tended to normalize one-year post-fusion surgery. This is not in contradiction with the present results, since the discs analysed in this study were within the instrumentation, and therefore, they could not recover their flexibility.

One of the limitations of this study was that measurements were performed with an anterior approach, which does not allow accessing the thoracic discs. However, posterior approach is impossible due to the presence of the bony neural arch, and mediolateral approach is difficult because of the larger depth of the disc in this approach. Even if the post-surgery part of the axial rotation of the discs and vertebra was corrected, we could have measured the medial part of the disc in healthy patients, and the medio-lateral part in scoliotic patient. That is why we sometimes put the probe a little oblique to find the best place to record our elastogram. Our main criterion was to be in the axial plane of the disc and centred on the medullary canal. However, mapping of elastographic properties of the disc around its surface could clarify the potential impact of measurement region. The other limitation is the possible bias in the measurement caused by the modification of the stiffness according to the posture [25, 26]. The stiffness of lumbar IVDs in lying position during the imaging may be underestimated compared to sitting posture on wheelchair. However, this bias was reduced because asymptomatic controls were assessed in the same posture. A group of preoperative patients would have been an interesting addition to this work. However, neuromuscular scoliosis is a relatively rare pathology, so it is difficult to obtain a homogeneous group of patients. We are planning a prospective study to follow a cohort of patients from pre-operative to long-term follow-up, with regular elastography.

Finally, machine difference can be an obstacle for this application to be standardized. However, it could be possible to normalize standardizing the measurement, for example, by doing a measuring soft-tissue SWE at a specific location as the baseline and use this to normalize the SWE value.

Conclusion

In this study, we present the first attempt to quantify the stiffness of lumbar discs in NMS, compared to healthy patients, using SWE. This technique proved to be a feasible, fast, and reliable means of quantifying disc mechanical properties and stiffening of the spine after FBF. This could be an additional argument when questioning the real need for PSF at skeletal maturity.

Data availability The data are available for 2 years, on request to the corresponding author.

Declarations

Conflict of interest All authors declare no conflicts of interest and financial support with this work. Dr Miladi receives royalties from EUROS company.

References

1. Skaggs DL, Akbarnia BA, Flynn JM, Myung KS, Sponseller PD, Vitale MG (2014) A classification of growth friendly spine implant. J Pediatr Orthop 34(3):260–274

2. Xu D, Ren Z, Chen X, Zhuang Q, Hui S, Sheng L, Li S (2017) The further exploration of hidden bloodloss in posterior lumbar fusion surgery. Orthop Traumatol Surg Res 103(4):527–530
3. Ramo BA, Roberts DW, Tuason D, McClung A, Paraison LE, MooreHG SDJ (2014) Surgical site infections after posterior spinal fusion for neuromuscular scoliosis: a thirty-year experience at a single institution. J Bone Joint Surg Am 96(24):2038–2048
4. Bouthors C, Gaume M, Glorion C, Miladi L (2019) Outcomes at skeletal maturity of a growing rod technique. About a homogeneous series of 34 early-onset of scoliosis. Spine 44(23):1630–1637
5. Miladi L, Gaume M, Khouri N, Johnson M, Topouchian V, Glorion C (2018) Minimally invasive surgery for neuromuscular scoliosis. Results and complications in a series of one hundred patients. Spine 43(16):E968–E975
6. Gaume M, Langlais T, Loiselet K, Pannier S, Skalli W, Vergari C, Miladi L (2023) Spontaneous induced bone fusion in minimally invasive fusionless bipolar fixation in neuromuscular scoliosis: a computed tomography analysis. Eur Spine J 32(7):2550–2557. https://doi.org/10.1007/s00586-023-07745
7. Creze M, Nordez A, Soubeyrand M, Rocher L, Maître X, Bellin MF (2018) Shear wave sonoelastography of skeletal muscle: basic principles, biomechanical concepts, clinical applications, and future perspectives. Skelet Radiol 47(4):457–471. https://doi.org/10.1007/s00256-017-2843-y
8. Gennisson JL, Deffieux T, Fink M, Tanter M (2013) Ultrasound elastography: principles and techniques. Diagn Interv Imaging 94:487–495. https://doi.org/10.1016/j.diii.2013.01.022
9. Eby SF, Song P, Chen S, Chen Q, Greenleaf JF, An KN (2013) Validation of shear wave elastography in skeletal muscle. J Biomech 46(14):2381–2387. https://doi.org/10.1016/j.jbiomech.2013.07.033
10. Langlais T, Vergari C, Pietton R, Dubousset J, Skalli W, Vialle R (2018) Shear wave elastography can evaluate annulus fibrosus alteration in adolescent scoliosis. Eur Radiol 28:2830–3283
11. De Reuver S, Moens AJ, Kruyt MC, Nievelstein RA, Ito K, Castelein RM (2022) Ultrasound shear wave elastography of the intervertebral disc and idiopathic scoliosis: a systematic review. Ultrasound Med Biol 48(5):721–729
12. Vergari C, Chanteux L, Pietton R, Langlais T, Vialle SW (2020) Shear wave elastography of lumbar annulus fibrosus in adolescent idiopathic scoliosis before and after surgical intervention. Eur Radiol 30(4):1980–1985
13. Vergari C, Dubois G, Vialle R et al (2015) Lumbar annulus fibrosus biomechanical characterization in healthy children by ultrasound shear wave elastography. Eur Radiol. https://doi.org/10.1007/s00330-015-3911-0
14. Romano A, Staber D, Grimm A, Kronlage C, Marquetand J (2021) Limitations of muscle ultrasound shear wave elastography for clinical routine-positioning and muscle selection. Sensors (Basel) 21(24):8490. https://doi.org/10.3390/s21248490
15. Vergari C, Rouch P, Dubois G, Bonneau D, Dubousset J, Tanter M, Gennisson JL, Skalli W (2014) Non-invasive biomechanical characterization of intervertebral discs by shear wave ultrasound elastography: a feasibility study. Eur Radiol 24(12):3210–3216. https://doi.org/10.1007/s00330-014-3382-8
16. Moreau B, Vergari C, Gad H, Sandoz B, Skalli W, Laporte S (2016) Non-invasive assessment of human multifidus muscle stiffness using ultrasound shear wave elastography: a feasibility study. Proc Inst Mech Eng H 230(8):809–814. https://doi.org/10.1177/0954411916656022
17. Poe-Kochert C, Shannon C, Pawelek JB, Thompson GH, Hardesty CK, Marks DS, Akbarnia BA, McCarthy RE, Emans JB (2016) Final fusion after growing-rod treatment for early onset scoliosis: is it final? J Bone Joint Surg Am 98(22):1913–1917
18. Gaume M, Vergari C, Khouri N, Skalli W, Glorion C, Miladi L (2021) Minimally invasive surgery for neuromuscular scoliosis: results and complications at a minimal follow-up of 5 years. Spine (Phila Pa 1976) 46(24):1696–1704
19. Gaume M, Persohn S, Vergari C, Glorion C, Skalli W, Miladi L (2020) Biomechanical cadaver study of proximal fixation in a minimally invasive bipolar construct. Spine Deform 8(1):33–38
20. Dubousset J, Gaume M, Miladi L (2021) Ilio-sacral screw pelvic fixation when correcting spinal deformities with or without pelvic obliquity: our experience over 40 years. Spine Deform 9(3):665–670
21. Harmon B, Wells M, Park D, Gao J (2019) Ultrasound elastography in neuromuscular and movement disorders. Clin Imaging 53:35–42
22. Lee SS, Gaebler-Spira D, Zhang LQ, Rymer WZ, Steele KM (2016) Use of shear wave ultrasound elastography to quantify muscle properties in cerebral palsy. Clin Biomech (Bristol, Avon) 31:20–28. https://doi.org/10.1016/j.clinbiomech.2015.10.006
23. Langlais T, Desprairies P, Pietton R, Rohan PY, Dubousset J, Meakin JR, Winlove PC, Vialle R, Skalli W, Vergari C (2019) Microstructural characterization of annulus fibrosus by ultrasonography: a feasibility study with an in vivo and in vitro approach. Biomech Model Mechanobiol 18(6):1979–1986. https://doi.org/10.1007/s10237-019-01189-3
24. Deviren V, Berven S, Kleinstueck F, Antinnes J, Smith JA, Hu SS (2002) Predictors of flexibility and pain patterns in thoracolumbar and lumbar idiopathic scoliosis. Spine (Phila Pa 1976) 27(21):2346–2349. https://doi.org/10.1097/00007632-200211010-00007
25. Chen B, Liu C, Lin M, Deng W, Zhang Z (2021) Effects of body postures on the shear modulus of thoracolumbar fascia: a shear wave elastography study. Med Biol Eng Comput 59(2):383–390. https://doi.org/10.1007/s11517-021-02320-2
26. Sadeghi S, Quinlan K, Eilertson KE, Billy GG, Bible J, Sions M, Cortes D (2019) Changes in shear modulus of the lumbar multifidus muscle during different body positions. J Biomech Eng doi 10(1115/1):4043443