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Use of Shear Wave Elastography in Pediatric Musculoskeletal Disorders

Celik Halil Ibrahim and Karaduman Aynur Ayşe

Abstract

Muscle shear-wave elastography (SWE) is an exciting and rapidly evolving ultrasound technique that allows quantification of muscle stiffness with a non-invasive, non-painful and non-irradiating examination. It has the potential of wider clinical use due to relatively low-cost, providing real-time measurement and, especially for the pediatric population, taking less time and sedation/anesthesia-free. Research indicate that muscle SWE shows promise as an adjunct clinical tool for differentiating between a normal and an abnormal muscle, monitoring the effectiveness of therapeutic interventions, altering the therapeutic intervention, or deciding treatment duration. This chapter will aim to provide an overview of the knowledge about the using of muscle SWE in common pediatric musculoskeletal disorders such as Duchenne Muscular Dystrophy, Cerebral Palsy, Adolescent Idiopathic Scoliosis, and Congenital Muscular Torticollis in the light of current evidence.

Keywords: shear-wave elastography, ultrasound elastography, muscle stiffness, muscle elasticity, musculoskeletal disorders

1. Introduction

Palpation is an ancient diagnostic method that has been used in medical practice for thousands of years since Hippocrates' day. Nevertheless, palpation is still valuable and often preferred today to assess the mechanical properties of a tissue in many clinical settings. However, some disadvantages such as limited tissue accessibility and inherent subjectivity may cause hesitations in its use [1]. To overcome these disadvantages by providing a more objective assessment of tissue mechanical properties, Magnetic Resonance Imaging Elastography and Ultrasound Elastography (USE) are the options offered by today's technology. However, the latter has the potential for wider clinical use because it is relatively low-cost, it provides real-time measurement, it takes less time, especially for the pediatric population, and it is sedation/anesthesia-free. The rapidly evolving evidence appears to be very promising that USE may be used to assess the mechanical properties of a tissue, especially muscle stiffness, that is, the degree of muscle resistance to deformation [2–4].

To briefly explain the basic principle, USE measures deformation in response to force/stress applied to the muscle. There are various USE techniques (strain elastography, Acoustic radiation force impulse [ARFI], Transient elastography, and Shear-wave elastography [SWE]), depending on the way of stress application and the

measurement of deformation. SWE quantitatively measures muscle stiffness based on shear-wave propagation within the tissue and seems to be ahead of other USE techniques due to some prominent features such as less user-dependency, providing quantitative results besides elastogram, and higher reliability and repeatability [1, 4].

SWE shows promise as an adjunct clinical tool for differentiating between a normal and an abnormal muscle, monitoring the effectiveness of therapeutic interventions, altering the therapeutic intervention, or deciding treatment duration. Therefore, SWE, which measures individual muscle stiffness, can provide significant benefits, especially for physiotherapists because many rehabilitation strategies aim to change the muscle structure.

This chapter aims to present an overview of the knowledge about the use of muscle SWE in common pediatric musculoskeletal disorders such as Duchenne Muscular Dystrophy and Adolescent Idiopathic Scoliosis in the light of current evidence.

2. Shear-wave elastography in pediatric musculoskeletal disorders

2.1 Duchenne muscular dystrophy

Duchenne Muscular Dystrophy (DMD), which is the most common childhood neuromuscular disease with a prevalence of 1/3600–9300, is an X-linked recessive hereditary disease characterized by progressive muscle degeneration and weakness [5, 6]. The natural course of DMD includes pathological muscle changes, including a decrease in the number of normal muscle cells, dystrophic changes in myofibrils, and the replacement of normal muscle cells by adipose and fibrous tissues [7].

It is difficult to evaluate pathological changes in muscles and ultimately disease progression in patients with DMD because repeated muscle biopsies are invasive and not easily obtained [8, 9]. Therefore, it is emphasized that there is a need for reliable and non-invasive objective assessment tools that can be used routinely to evaluate disease progression and the efficacy of therapeutic interventions [10]. In response to this need, it has been emphasized in recent years that SWE is a promising clinical assessment tool to evaluate muscle pathology and disease progression in DMD [10–13].

Lacourpaille et al. and Pichiecchio et al. have found that lower and upper extremity muscle stiffness was higher in children with DMD compared with healthy controls and that SWE was able to distinguish between normal and dystrophic muscles [11, 12]. In another study, it was shown that muscle stiffness increased significantly in children with DMD after a 12-month follow-up period while muscle stiffness was stable in controls, and it was stated that SWE has the potential to be used in monitoring DMD progression [13]. Lin et al. reported that the stiffness of the tibialis anterior and the gastrocnemius medialis muscles decreased from ambulatory to early non-ambulatory stages, whereas the stiffness of the rectus femoris muscle increased. This result pointed out that individual muscles have different alteration patterns in stiffness as the ambulatory function declined and that SWE may be useful in the classification and prediction of ambulation status in children with DMD [10]. Additional information on muscle SWE studies in children with DMD is provided in **Tables 1** and **2**.

2.2 Cerebral palsy

Cerebral palsy (CP), a neurodevelopmental disorder caused by non-progressive immature brain disturbances, affects 2.11 of every 1000 children in high-income

Authors	Participants	Muscles	Findings
Lacourpaille [12]	14 children with DMD (age: 13.3 ± 5.9 y); 13 healthy controls (age: 12.8 ± 5.5 y)	TA, GM, VL, BB, TB, ADM	Significantly higher muscle stiffness in DMD patients compared with controls for all muscles, except for ADM.
Lacourpaille [13]	10 children with DMD (age range: 7–22 y); 9 healthy controls (age range: 7–22 y)	TA, GM, BB, TB, ADM	Significant increase in TA, GM, and TB stiffness over 12 months follow-up in patients with DMD while stiffness was stable in controls.
Pichiecchio [11]	5 preschool children with DMD (age range: 3.2–4.9 y); 5 healthy controls (age range: 3.3–3.9 y)	GM, RF, VM, VL, AM, TA, GMax	Muscle stiffness of the DMD patients was moderately higher than controls in the RF, VL, AM, and GMax muscles, whereas VM, TA and GM muscles of DMD patients were only minimally more stiff.
Lin [10]	39 children with DMD (age: 10.4 ± 3.8 y); 36 healthy controls (age: 9.2 ± 4.1 y)	RF, TA, GM	The stiffness of the RF and GM muscles was significantly higher in DMD patients compared with the control group. The stiffness of the TA and GM muscles decreased from ambulatory to early non-ambulatory stages, whereas the stiffness of the RF muscle increased from ambulatory to late non-ambulatory stages in DMD patients.

GMax, gluteus maximus; AM, adductor magnus; ADM, abductor digiti minimi; BB, biceps brachii; GM, gastrocnemius medialis; GL, gastrocnemius lateralis; RA, rectus abdominis; RF, rectus femoris; TA, tibialis anterior; TB, triceps brachii; VL, vastus lateralis; VM, vastus medialis.

Table 1.
Studies using muscle shear-wave elastography in Duchenne muscular dystrophy.

countries and is often considered to be the most common childhood motor disability [14, 15]. In CP, spasticity caused by an upper motor neuron lesion often leads to muscle weakness, muscle stiffness, and contractures, which eventually restrict general mobility [16]. Recent studies have emphasized the increased intensity of the connective tissue and extracellular matrix in spastic muscles, indicating that muscle stiffness may be a critical factor in the worsening of motor disorder over time in children with CP [17, 18]. These research findings may explain our clinical observations that many children with CP have an increase in muscle stiffness and a decrease in joint range of motion and in the efficiency of walking as they age into adolescence and adulthood. Therefore, it is not surprising that many studies have evaluated muscle stiffness with SWE in children with CP.

Muscle SWE studies comparing healthy children and children with CP have reported that muscle (GM, GL, BB, and SoL) stiffness is significantly higher in children with CP [16, 19–21]. Lee et al. [22] and Boulard et al. [23] have noted that muscle stiffness was higher in the more-affected limb than in the less-affected limb of children with CP. In addition, positive correlation has been reported between muscle stiffness and spasticity [20, 24, 25]. SWE has been used not only to distinguish between spastic and normal muscles but also to evaluate intervention effectiveness. All but one [26] of the studies investigating the effectiveness of botulinum toxin A (BTX) injection have reported a significant decrease in muscle stiffness a month after the injection [24, 25, 27]. In addition, it was stated that muscle stiffness values after 3 months is not significantly different from baseline values due to the duration of the

Muscles		Lacourpaille 2015 [12]	Pichiecchio 2018 [11]	Lin 2021 [10]
TA	DMD	23.1 ± 14.7 kPa	11.64 ± 1.35 kPa	2.87 ± 0.77 m/s
	HC	12.5 ± 3.1 kPa	13.54 ± 3.99 kPa	2.70 ± 0.42 m/s
GM	DMD	21.9 ± 12.7 kPa	12.9 ± 3.45 kPa	2.19 ± 0.57 m/s
	HC	14.5 ± 3.5 kPa	12.18 ± 5.56 kPa	1.85 ± 0.28 m/s
VL	DMD	21.5 ± 18.8 kPa	14.26 ± 1.34 kPa	—
	HC	9.6 ± 2.2 kPa	7.8 ± 2.74 kPa	—
VM	DMD	—	15.30 ± 2.43 kPa	—
	HC	—	11.56 ± 3.29 kPa	—
RF	DMD	—	14.44 ± 2.64 kPa	2.10 ± 0.72 m/s
	HC	—	8.10 ± 1.19 kPa	1.77 ± 0.28 m/s
GMax	DMD	—	13.92 ± 2.76 kPa	—
	HC	—	9.92 ± 1.31 kPa	—
AM	DMD	—	14.10 ± 1.31 kPa	—
	HC	—	9.74 ± 3.09 kPa	—
BB	DMD	34.9 ± 23.3 kPa	—	—
	HC	18.9 ± 6.4 kPa	—	—
TB	DMD	8.7 ± 2.1 kPa	—	—
	HC	7.3 ± 1.4 kPa	—	—
ADM	DMD	14.1 ± 7.8 kPa	—	—
	HC	11.9 ± 5.0 kPa	—	—

Table 2.
Muscles stiffness values in Duchenne Muscular Dystrophy.

effect of botulinum toxin [26]. In a study examining the relationship between hip muscle stiffness and hip dislocation, it was reported that there was a correlation between the Reimers Migration Index and the stiffness of the adductor magnus and the iliopsoas muscles [28]. Additional information on muscle SWE studies in children with CP is provided in **Tables 3** and **4**.

2.3 Adolescent idiopathic scoliosis

Adolescent idiopathic scoliosis (AIS), the most common type of scoliosis, is a 3D spinal deformity characterized by the deviation of the spine in the sagittal, frontal, and transverse plane [29]. The etiology of AIS has remained unclear until today even though many hypotheses have been put forward to explain the origin of this deformity [30].

Asymmetric loading on the vertebrae may be one of the causes contributing to the etiology of AIS [31]. Therefore, studies evaluating muscle stiffness with SWE and investigating muscle imbalance have been conducted to clarify the etiology of scoliosis. It has been suggested that this asymmetrical load may be caused by the paravertebral and lateral abdominal muscles consisting of the transversus abdominis (TrA), obliquus internal (OI), and obliquus external (OE) [32, 33]. Liu et al. reported no significant difference in paravertebral muscle stiffness between the concave and

Authors	Participants	Muscle(s) and Stiffness Values		Findings
Lallemant-Dudek [19]	16 children with CP (age: 8.3 ± 2.8 y); 29 healthy controls (age: 12.1 ± 3.3 y)	GM Stretched GCM; CP: 8.8 ± 4.1 m/s Controls: 2.9 ± 0.7 m/s GCM at rest CP: 3.1 ± 1.8 m/s Controls: 1.8 ± 0.6 m/s	BB did not provide original data	Intra-rater ICC: 0.90 and inter-rater ICC: 0.92 in healthy controls. Stretched GM was significantly stiffer in CP than in controls. GM at rest did not show any difference between groups, nor did BB either at rest nor stretched.
Vola [20]	21 children with CP (age range: 3–16 y); 21 healthy controls (age range: 3–14 y)	SoL CP: 8.1 ± 2.3 kPa Controls: 4.8 ± 1.7 kPa		Muscle stiffness is significantly higher in children with CP compared with controls. High positive correlation ($r = 0.74$) between muscle stiffness and spasticity (MAS).
Lee [22]	8 children with CP (age: 9.4 ± 3.7 y)	GM more affected: 5.05 ± 0.55 m/s, less-affected: 4.46 ± 0.57 m/s	TA more affected: 3.86 ± 0.79 m/s, less-affected: 3.22 ± 0.40 m/s	Muscle stiffness of the GM and TA in the more-affected limb was higher than in the less-affected limb.
Brandenburg [16]	13 children with CP (age range: 2–12 y); 13 healthy controls (age range: 2–12 y)	GL CP: 15.0 (11.6, 17.5) kPa Controls: 7.8 (6.1, 11.0) kPa		Muscle stiffness is significantly higher in children with CP compared with controls.
Boulard [23]	11 children with CP (age: 11.1 ± 1.7 y)	GM more-affected: 10.2 ± 2.6 kPa less-affected: 7.9 ± 1.5 kPa		Muscle stiffness of the stretched GM in the more-affected limb was higher than in the less-affected limb.
Bilgici [21]	17 children with CP (age: 9.25 ± 2.68 y); 25 healthy controls (age: 10.40 ± 2.76 y)	GM CP: 3.17 ± 0.81 m/s Controls: 1.45 ± 0.25 m/s		Muscle stiffness is significantly higher in children with CP compared with controls.
Doruk Analan [28]	25 children with CP (age: 4.07 ± 2.25 y);	AM, IP AM: 2.65 ± 1.03 m/s IP: 2.61 ± 0.85 m/s		AM and IP muscle stiffness show correlation with the Reimers Migration Index (0.70 and 0.71, respectively).

IP, Iliopsoas; AM, adductor magnus; BB, biceps brachii; GM, gastrocnemius medialis; GL, gastrocnemius lateralis; SoL, Soleus; TA, tibialis anterior; MAS, Modified Ashworth Scale; kPa, kilopascal.

Table 3.
Studies using muscle shear-wave elastography in cerebral palsy-1.

the convex side of scoliosis [32]. In a study examining the stiffness of the lateral abdominal muscles (TrA, OI, and OE), it was stated that there were no differences between healthy control and AIS groups in terms of muscles stiffness at rest and during isometric contraction and also there was no muscles stiffness asymmetry between the concave and the convex sides of scoliosis [33]. In addition, the same

Authors	Participants	Muscle(s) and Stiffness Values		Findings
Bilgici [24]	12 children with CP (age: 8.58 ± 2.48 y);	GM Before: 3.20 ± 0.14 m/s After: 2.45 ± 0.21 m/s		Significant decrease in muscle stiffness a month after BTX injection. Muscle stiffness is correlate with MAS ($r = 0.578$).
Brandenburg [26]	10 children with CP (age range: 2.1–8.8 y);	GL (The article provided the change values, not the original values.)		Notable, but non-significant, decrease in muscle stiffness 1 month after BTX injection. Baseline muscle stiffness is not significantly different from 3 month after BTX injection.
Bertan [27]	17 children with CP (age: 4.6 ± 1.2 y); 16 children with CP (Control group) (age: 4.4 ± 1.2 y)	GM Before and after values for CP: 2.32 ± 0.50 and 2.08 ± 0.47 m/s Before and after values for controls: 2.34 ± 0.53 and 2.32 ± 0.48 m/s	GL Before and after values for CP: 2.07 ± 0.37 and 1.90 ± 0.31 m/s Before and after values for CP: 2.10 ± 0.48 and 1.98 ± 0.40 m/s	Significant decrease in muscle stiffness of GM and GL 1 month after BTX injection in study group while no significant decrease in the control group.
Dağ [25]	24 children with CP (age: 6 ± 2.8 y);	GM Before: 45.9 ± 6.5 kPa After: 25.0 ± 5.7 kPa		Significant decrease in muscle stiffness 1 month after BTX injection. Muscle stiffness is correlate with MAS and MTS ($r = 0.77$ and 0.79 , respectively).

GM, gastrocnemius medialis; GL, gastrocnemius lateralis; MAS, Modified Ashworth Scale; MTS, Modified Tardieu Scale; BTX, botulinum toxin; kPa, kilopascal.

Table 4.
Studies using muscle shear-wave elastography in cerebral palsy-2.

research group found that stiffness measurements of the lateral abdominal muscles were carried out with high reliability/agreement during contraction, while the reliability of the stiffness measurements ranged from moderate to excellent at rest [34]. Another issue that attracts the attention of researchers is the involvement of respiratory muscles in scoliosis. By altering the biomechanics of the rib cage, scoliosis can affect the intercostal muscles (ICMs), thoracic expansion, and ultimately respiration. However, Pietton et al. investigated the stiffness of the ICMs in healthy control and AIS groups and reported that there was no significant difference between groups, although the AIS group displayed a trend toward higher stiffness of the ICMs than in the healthy group [35]. Additional information on muscle SWE studies in children with CP is provided in **Table 5**.

2.4 Congenital muscular torticollis

Congenital muscular torticollis (CMT) is a common muscular disorder occurring at or shortly after delivery as a result of the unilateral shortening of the

Authors	Participants	Muscle(s) and stiffness values	Findings
Linek [34]	35 children and adolescents with AIS (age: 12.8 ± 2.8 y)	OE, OI, TrA OE: 21.1 ± 9.19 kPa OI: 14.6 ± 4.24 kPa TrA: 13.8 ± 4.22 kPa	At rest, the reliability of stiffness measurements ranged from moderate to excellent in all examined muscles (ICC: 0.56–0.94). During contraction, muscles stiffness was measured with high reliability/ agreement (ICC: 0.63–0.91).
Linek [33]	108 children and adolescents with AIS (age range: 10–17 y); 151 healthy controls (age range: 10–17 y)	OE, OI, TrA For AIS: OE: 16.6 ± 5.64 kPa OI: 15.5 ± 4.63 kPa TrA: 14.1 ± 3.50 kPa For healthy controls; OE: 16.2 ± 5.67 kPa OI: 15.2 ± 4.03 kPa TrA: 13.9 ± 3.06 kPa	There were no differences between control and AIS groups in the muscles stiffness at rest and during isometric contraction. There were no muscles stiffness asymmetry between convex and concave body sides.
Liu [32]	40 children and adolescents with AIS (age: 10–18 y)	paravertebral muscles Concave side: 18.27 kPa Convex side: 14.31 kPa	No significant difference in muscle elasticity between the concave and the convex sides.
Pietton [35]	16 children and adolescents with AIS (age: 13 ± 2.5 y); 19 healthy controls (age: 12.6 ± 1.7 y)	ICMs AIS: 2.2 ± 0.3 m/s Healthy controls: 2.1 ± 0.4 m/s	SWE is feasible and reliable in the assessment of the ICMs of healthy individuals and those with scoliosis (ICC: 0.85 and 0.83, respectively). Although the AIS group showed a tendency toward higher ICMs stiffness than in the healthy group, there was no significant difference between groups.

OE = external oblique muscle; OI = internal oblique muscle; TrA = transversus abdominis muscle; ICMs, intercostal muscles; kPa, kilopascal.

Table 5.
Studies using muscle shear-wave elastography in adolescent idiopathic scoliosis.

sternocleidomastoid muscle (SCM), which results in clinical symptoms including head lateral tilt toward the ipsilateral side and head rotation to the opposite side [36, 37]. Long-lasting severe CMT can lead to asymmetric cranial and facial structures. Although the exact etiology of CMT remains unclear, endomysial fibrosis with collagen deposition and the migration of fibroblasts to individual muscle fibers are involved in the pathogenesis of CMT [38]. Ultimately, muscle fibrosis and increased stiffness can reduce the elasticity and function of the muscles, which leads to the range of motion deficit and contracture. Although CMT is a muscle-derived condition, the insufficient number of muscle SWE studies addressed the SCM stiffness indicates a large gap in the literature.

It was reported that the stiffness of the affected SCM was positively correlated with the degree of PROM deficit of neck rotation in the affected side [39–41] and affected SCM stiffness was significantly higher than that of the

Authors	Participants	Muscle(s) and stiffness values	Findings
Park [39]	20 infants with CMT (age: 0.71 ± 0.25 mo); 12 healthy controls (age: 0.65 ± 0.27 mo)	SCM infants with CMT: 3.65 ± 0.75 m/s Healthy controls: 1.50 ± 0.30 m/s	In the CMT group, the stiffness of the affected SCM was significantly higher than that of the unaffected SCM and than that in the control group. The stiffness of the affected SCM was positively correlated with the degree of PROM deficit of neck rotation in the affected side ($r = 0.77$). The intrarater reliability: ICC = 0.923.
Hwang [41]	22 infants with CMT (age: 1.16 ± 0.66 mo)	SCM Initial assessment: 2.33 ± 0.47 m/s After 3 months: 1.56 ± 0.63 m/s	The SCM stiffness decreased significantly from the initial evaluation to 3 months after the start of the physiotherapy. The initial SCM stiffness showed negative correlations with the degree of cervical rotation and lateral flexion, respectively ($r = -0.642$ and $r = -0.643$)
Zhang [40]	46 late-referral infants with CMT (age: 8.13 ± 1.77 mo)	SCM Affected SCM: 205.53 ± 46.34 kPa Unaffected SCM: 27.91 ± 4.90 kPa	SCM stiffness was positively correlated with the degree of the PROM deficit in neck rotation ($r = 0.82$). The intrarater reliability: ICC = 0.981

SCM, sternocleidomastoid muscle; PROM, passive range of motion; kPa, kilopascal.

Table 6.
Studies using muscle shear-wave elastography in congenital muscular torticollis.

unaffected SCM and than that in the control group [39]. Hwang et al. also reported that SCM stiffness decreased significantly after 3 months of physiotherapy [41]. These studies point out the potential of muscle SWE in the diagnosis and treatment of CMT. Additional information on muscle SWE studies in CMT is provided in **Table 6**.

2.5 Healthy children

Muscle SWE studies in healthy children are important for the reliability and repeatability of the measurement methods and to understand the effect of some individual factors such as sex and age on muscle stiffness. Brandenburg et al. reported that sex, age, BMI, extremity dominancy, and calf circumference were not associated with muscle stiffness in children [42]. Liu et al. compared the gastrocnemius medialis muscle stiffness of different age groups and found that there was no significant difference between sexes and muscle stiffness was the greatest in the older group, followed by the middle-aged group and then the children group [43]. Although these two studies give some clues, we think that further muscle SWE studies in healthy children are required and these studies are important especially for the measurement reliability, standardization of the measurement method, and establishing norm values for muscle stiffness. Additional information on muscle SWE studies in healthy children is provided in **Table 7**.

Authors	Participants	Muscle(s) and stiffness values	Findings
Liu [43]	27 children (age: 7.46 ± 1.46 y), 31 middle-aged individuals (age: 34.65 ± 3.19 y), and 28 older people (age: 62.25 ± 2.72 y).	GM Children: 24.28 ± 7.72 kPa Middle-aged individuals: 28.39 ± 6.85 kPa Older people: 34.89 ± 8.48 kPa	In all groups, passive muscle stiffness increased as the ankle DF increased. There was no significant difference between sexes. No significant difference in muscle stiffness between the three groups in terms of PF angles. The difference in muscle stiffness among the three groups became significant as DF increased. In terms of the ankle angles of DF, the muscle stiffness was the greatest in the older group, followed by the middle-aged group and then the children group (that is, stiffness increases with age).
Brandenburg [42]	20 healthy children (age range: 2–12 y),	GL R: 7.7 ± 2.5 kPa L: 7.8 ± 3.3 kPa	Sex, age, BMI, extremity dominance, and calf circumference did not significantly correlate with muscle stiffness.

GM, gastrocnemius medialis; GL, gastrocnemius lateralis; DF, dorsi flexion; PF, plantar flexion; BMI, body mass index; R, right; L, left; kPa, kilopascal.

Table 7.
Studies using muscle shear-wave elastography in healthy children.

3. Conclusions

Muscle SWE is an exciting and rapidly evolving US technique that allows quantification of muscle stiffness with a non-invasive, non-painful, and non-irradiating examination. It has the potential of wider clinical use because it is relatively low-cost, it provides real-time measurement, it takes less time, especially for the pediatric population, and it is sedation/anesthesia-free. SWE shows promise as an adjunct clinical tool for differentiating between normal and abnormal muscles, monitoring the effectiveness of therapeutic interventions, altering the therapeutic intervention, or deciding treatment duration. Therefore, SWE, which measures individual muscle stiffness, can provide significant benefits, especially for physiotherapists because many rehabilitation strategies aim to change muscle structure. However, some remarkable points such as the insufficient number of studies, the small sample size, the differences in measurement settings and methods between studies, and the lack of norm values for different muscles indicate the necessity for further studies.

Conflict of interest

The authors declare that they have no conflict of interest.

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