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Educational article

Childhood incontinence and pelvic floor muscle function: Can we learn from adult research?

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Summary
Little research has been undertaken regarding pelvic floor muscle function and dysfunction in children. Apart from dysfunctional voiding, whether specific dysfunction of these muscles contributes to different types of childhood incontinence is not documented. The two areas of practice, that is adult and paediatric incontinence, are quite different; nevertheless, there may be lessons to be learned from the extensive work done in pelvic floor dysfunction and rehabilitation in adults. In this paper key points regarding pelvic floor muscle function and dysfunction in adults, and lessons that can be learned from how studies are reported and how muscle function is measured are presented. The aim is to prompt deeper thought regarding the direction in which further research in children may proceed. There are challenges in first validating muscle measurement standards and making the work relevant to children across different age groups with differing cognitive and motor abilities.

Introduction

Children cannot be treated as little adults. This is particularly true of paediatric incontinence where bladder and bowel dysfunctions differ from adult problems and management can be affected by behavioural issues and the fact that we are dealing with growing and developing individuals. It would not be wise for health practitioners in one area of practice to ignore what has been learned by their colleagues in the other, particularly as there is an area of overlap in the young people who are transitioning into adulthood and adult services.

In 2008 Delancey et al. [1] developed the Lifespan Model, and described a graphical tool to integrate pelvic floor function related to pelvic floor disorders in three phases, the first being "development of functional reserve during an individual's growth". A young person who develops this reserve may be less predisposed to pelvic floor dysfunction in adult life. In the area of female sexual dysfunction it has been stated that "primary pelvic floor muscle hyperactivity in children and adolescents is one of the most neglected predisposing factors to later dyspareunia and vulvar vestibulitis" [2]. A better understanding of how to measure pelvic floor muscle (PFM) function in children and therefore how to retrain these muscles has clinical relevance not only for the treatment of incontinence in the young person but also falls into the category of preventative medicine.

This paper summarises some aspects of current thinking and evidence on PFM therapy in adults from relevant literature which has progressed since the inception of the International Continence Society, and during which time there has been less work undertaken in this area in children.

It seems that there is an opportunity to learn from adult research with a view to thinking more deeply about the investigation of whether PFM dysfunction is a factor in childhood incontinence. To prescribe an effective muscle training programme the muscle dysfunction needs to be evaluated. Recipe approaches are not best practice. What is the problem? Is it lack of strength, lack of endurance, a problem of timing of contraction or relaxation, or fascial laxity? For example, one could hypothesise that the muscle dysfunction/s, if any, in children with generalised joint hypermobility would be different from those with giggle incontinence, and therefore the prescription of exercise also different.

This paper presents some of the information that is known about PFM function/dysfunction; how it may be measured; and some suggestions as to how research in this area in children may be undertaken to

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maximise the usefulness of the results, and determine if this is an area of paediatric practice that requires more attention.

The anatomic structures that prevent incontinence are smooth and striated muscle sphincters, endopelvic fascia, connective tissue and ligaments, mucosal and vascular tissues and the supportive hammock of the levator ani and the more superficial perineal muscles. In adults, pelvic floor dysfunction refers to any disruption of function in any aspect of these structures potentially leading to pelvic floor disorders of urinary and faecal incontinence, defaecation difficulty, pelvic organ prolapse, sensory and emptying abnormalities, sexual dysfunction, and chronic pelvic pain [3].

This paper is directed at PFMs, meaning the striated muscle component of the pelvic floor which is under voluntary control and therefore able to be exercised or trained.

In adults the anatomy, neurophysiology, function (and dysfunction) of the levator ani, superficial perineal muscles, and anal and urethral sphincters have been the subject of much research. The support, postural, sexual, and continence roles of these muscles are now better understood. These muscles are activated synergistically with non-PFM during functional tasks. PFMs are active during activation of abdominal muscles [4,5], gluteal muscles [6], hip adductors [6], during voluntary shoulder flexion and extension [7], and breathing [7]. There is a postural role of PFMs, in which the interaction of these, the diaphragm, and the deep abdominal and spinal muscles, work to stabilise the spine and allow an efficient base from which limb movements arise. It appears that as well as tonic activity, PFMs have reflex activity, such as the response to a sharp rise in intra-abdominal pressure. PFM activity is also modulated by "pattern generators" in the central nervous system, including the brainstem, that co-ordinate the response to smooth muscle behaviour and groups of striated muscle. Recently, it is has been suggested that feed forward loop synergistic patterns such as the recruitment of PFMs before an event may be mediated at a cortical level [8]. For a detailed understanding of PFM functional anatomy, neuroanatomy, and neurophysiology the reader is directed to relevant chapters in Bo et al. [2]. However a few points of interest are the following.

A correct conscious contraction of the levator ani results in occlusion (squeeze) and a cranioventral shift of the pelvic organs. This "lift" reinforces urethral and anal closure pressure and also plays an important role in protecting the pelvic tissues from excess load. Prolonged downward tensile force has the potential to stretch ligaments and fascia in the pelvis. Besides pregnancy and birth, medical issues such as chronic cough and lifestyle factors such as obesity are relevant. An increase in waist circumference has been shown to be significantly associated with an increase in both abdominal and intravesical pressure [9]. Genetic factors such as poor collagen quality may result in a "lower" pelvic floor so the time for muscles to reach an optimal contraction to prevent descent and augment urethral closure maybe too slow to counteract the rise in intra-abdominal pressure that occurs with coughing, sneezing, or impact, so urinary leakage occurs.

At least 30% of healthy woman cannot readily perform a contraction on command despite individual instruction [10–12]. Instead they may contract abdominals, gluteals, adductors, breath-hold, or increase inspiration or strain down. In an ultrasound study done on healthy children a similar third of children showed inappropriate displacement direction on a contractile effort [13]. A study of levator plate (LP) movement in women with incontinence identified three different patterns of movement of the LP, with 38% of subjects elevating and 43% of subjects depressing the LP and 19% having no change in LP movement. In the stress incontinence group, there was a higher than expected number that elevated the LP. In the urgency and prolapse groups there was a higher than expected number of subjects that depressed the LP [14]. In another study bladder neck depression during attempts to perform an elevating PFM contraction occurred in 17% of continent and 30% of incontinent women. The urge urinary incontinence group had the highest proportion of women who depressed the bladder neck (40%) [15].

Differences in the amount of bladder neck and bladder base movement were more marked between incontinent and continent women during functional activities than during the voluntary PFM contractions. This highlights the importance of including an assessment of the PFM during functional activities. The ability to elevate the pelvic floor may not in itself be evidence of good PFM function and should be interpreted in combination with data obtained from a full clinical assessment including functional manoeuvres known to generate intra-abdominal pressure to assess the ability of the PFM to resist depression under load [16].

If PFM contraction is being taught, the "lift" needs to be verified. Methods to do this are discussed below, as electromyography (EMG) does not necessarily do this.

Pelvic floor muscle function in children

Little is known about PFM interaction and control in children and at what ages these particular functions develop, and despite extensive searching of all relevant databases no new work seems to be forthcoming. Wendell-Smith [17] postulated that the pelvis of the neonate is funnel shaped so that when the baby cries or is held upright pressure is directly transmitted to the visceral mass with the tendency for the organs to enter the pelvic cavity and the PFMs contract to counteract this rise in pressure. When the child begins to stand and walk the diaphragm becomes more muscular and the effect of sudden contractions of the diaphragm is to direct pressure towards the anterior abdominal wall, and in turn is directed downwards and posteriorly to the sacrum and causes its concavity by puberty. Hence the pelvic floor is protected from full visceral thrusts [17]. It was suggested by Lansman and Robertson [18] that it is not until this time that pubococcygeus and puborectalis truly separate in their function.

Kolar says that "at birth the diaphragm is still in an oblique position while the pelvic floor has no postural function, nor is there any postural synergy between the intrinsic back and the abdominal muscles. As the intrinsic back musculature comes into play, the spinal column straightens. The thorax with the ribs is stabilised by the caudal pull of the abdominal muscles. The ribs are steeper than in the newborn stage. Straightening up of the spine in concert with stabilizing activity in the abdominal muscles alters the fixation point for the attachments of the diaphragm, thus bringing the diaphragm into a horizontal position. This is as specific for humans as is erect posture. Increased intra-abdominal pressure and decreased pelvic anteversion through the pull of the abdominal and gluteal muscles enable the pelvic floor to perform its postural function" [19]. Functionally, PFMs cannot be considered in isolation. Little work has been done on the relationship between posture and PFM dysfunction in children. The authors only know of several small studies in children. Wennergren et al. [20] demonstrated differences in degree of PFM activity in different toilet sitting postures in girls. A difference between abdominal and spinal musculoskeletal function in children with slow transit constipation compared with controls has been demonstrated [21].

It is likely that both internal and external factors affect PFM function. Looking at the well-known model of potential aetiological factors (Table 1), many of these listed external factors may well be relevant to children. Others internal behavioural factors are present — for example children who have experienced distress around defecation are predisposed to withholding of stool, PFM non-relaxation during defecation, and functional constipation. Such a model in children would be worth developing.

More recently magnetic resonance imaging (MRI) has been used to identify PFM disruption in children with anorectal malformations, and also in children with no anomalies. Such details of PFM, in this case puborectalis and the striated anal sphincter, are not usually seen in paediatric papers [22—24]. It would seem that in specialist centres MRI (perhaps already available and done for reasons other than PFM dysfunction) and 3D or 4D ultrasound offer opportunities to better understand the development of PFM structure and function over a range of age groups.

Reporting of studies in children

The evidence for PFM training in adult women with dysfunction is level 1A [25]. Underlying this evidence are well-designed studies with investigators skilled in muscle physiology, assessment of function and dysfunction, exercise prescription and the measurement of muscle outcomes. In the paediatric arena this has not happened to date, and one of the frustrations for clinicians is the poor reporting of methodologies.

The authors know of only one study that reports the exercise regime for children with dysfunctional voiding. In this study the children practised a 3-second submaximal PFM contraction followed by 30 s of relaxation [26]. This regime would not achieve any gains in strength or endurance, so improvements could be attributed to increased awareness and/or ability to contract and relax. Measurement of muscle parameters seems not to exist at all.

"Without a complete published description of interventions, clinicians and patients cannot reliably implement interventions that are shown to be useful, and other researchers cannot replicate or build on research findings". For this reason the Template for Intervention Description and Replication (TIDieR) checklist and guide has been developed and future studies should adhere to the 12 items [27]. When reporting results from muscle testing the equipment used needs to be specified, position during testing reported, the instruction and motivation given outlined, and definition of the parameters tested supplied e.g. the ability to contract, strength, endurance etc. Even the words used for instruction are important [28].

Measurement of interventions

There is no one way of measuring the actions of PFM. Measuring PFM function in children is difficult because of the potential invasiveness of doing so, but it is an issue that now needs more thought. Clinicians will already have ways of assessing PFM function but not actually measuring it (see Table 2). To progress our understanding, research needs to measure before and after interventions. We need to be clear what we are measuring. If we are training to improve defaecation, then it is the anorectal angle and function of puborectalis and anal sphincters that need close attention. This is not effectively measured by perineal electrodes. If it is the effect of PFM at the bladder neck that is the issue, then measuring anal sphincter function is not sufficient. These methods give little information about the direction, the "lift" or the occlusion of PFM activity. We need to be more precise when addressing specificity of muscle action, measurement and rehabilitation.

Confounders of measurement abound. An EMG reading alone is not comparable on two different occasions. PFM EMG and pressure measurements can be affected by posture, talking, breathing, and especially "cross-talk" from surrounding muscles. Muscle measurement needs to be done in a standardised position before and after intervention and confounders controlled for. The length of the exercise session and muscle fatigue alters results.

Leaving urodynamic measurements aside, in adults PFM can be measured by visual observation, vaginal or anal palpation, EMG, vaginal pressure measurement, pelvic floor dynamometry, transabdominal or transperineal ultrasound.

Table 1 Potential aetiological factors leading to pelvic floor dysfunction in women.

Predisposing factors — gender, genetic, neurological, anatomical, collagen, muscular, cultural, environmental
Inciting factors — childbirth, nerve damage, muscle damage, radiation, tissue disruption, radical surgery
Promoting factors — constipation, occupation, recreation, obesity, surgery, lung disease, smoking, menstrual cycle, infection, medication, menopause
Decompensating factors — ageing, dementia, debility, disease, environment, medication

From Delancey et al. [1].

Table 2 Initial non-invasive PFM and postural assessment of children in the continence clinical setting.

Observe	Habitual standing posture		
	Habitual sitting posture		
	Breathing — upper chest, diaphragmatic		
	Overall "tone"		
	Habitual toilet posture	Buttocks	
		Thoracic spine	flexed/neutral
		Lumbar spine	flexed/neutral/extended
		Abdominal wall	lax/normal/tight
		Hips	abducted/adducted
			flexed to 90°
			flexed to <90°
			flexed to >90°
		Feet	supported/not supported
		♂ standing	pants well down/lumbar spine flexed/extended/neutral
		Abdominal relaxation	Yes/No
		Abdomen	retract/bulge/neutral brace/bulge then retract
	Defaecation pattern	Breathing	hold on inspiration
			upper chest
			diaphragmatic
		Awareness of anal contraction/relaxation	Yes/No
Palpate (after age-appropriate education)	On voluntary PFM contraction	Co-contraction	Transversus abdominis
			deep multifidis
		Perineal body	lift/descent/no movement
		Medial to ischial tuberosities	lift/descent
		Global abdominals	relaxed/overactive
		Gluteals	relaxed/active
		Adductors	relaxed/active
		Breathing	continuous/held
		Awareness lying	Yes/No
		Awareness sitting	Yes/No
		Awareness standing	Yes/No
		Awareness 4 foot kneel	Yes/No
	Overall sensorimotor awareness	Comment	

Note. PFM = pelvic floor muscle.

For detailed information about each of these in regard to responsiveness, reliability and validity the reader is directed again to Bo et al. [2].

Understanding of development of motor control in children

It is necessary to acknowledge the distinct age-related differences in children to make informed decisions about optimal clinical interventions; we should consider the developmental maturation of the child. Little is documented in the literature about the developmental, biomechanical, and musculoskeletal approaches to providing a part or whole solution for children with continence difficulties. It is important when contemplating research or intervention in this age group to remember that not only are children physically different and changing over time, they are also under the influence of rapidly altering cognitive function and ability. Cognitive ability and motor performance go hand in hand to develop functional outcomes in children.

The Dynamic Systems Theory is frequently used to drive current paediatric research and intervention strategies, to keep them focused on the relationship between child development and the overall desired outcome/s [29,30]. The theory considers how internal components, their rates of maturation, and their interaction constrain or affect the outcome of intervention, but also the external context of the task. In the paediatric population the maturation of particular systems (subsystems) will affect motor performance. An example of this might be achieving continence when there has been a prolonged dysfunctional voiding pattern. This will involve organization, and in some areas maturation, of the internal components of strength, coordination, body composition, balance, postural control, tonal equilibrium, sensorimotor influences, and motivation.

Table 3 Factors to be considered in training pelvic floor muscle function in children.

Area	General information	Paediatric specific information
Posture	In the adult population the ability to maintain appropriate spinal position and use anticipatory postural adjustments is critical to pelvic floor control. Children are still experiencing changes in their posture over time. Postural control is integral to the execution of goal-directed action. Trunk control, breathing, and internal functions are dependent on the ability to regulate pressure in the thoracic and abdominal chamber, the control of which extends from the vocal chords to the pelvic floor.	Sagittal alignment changes Increasing pelvic incidence angle Increasing thoracic kyphosis angle Increasing lumbar lordosis angle increasing until adulthood Postulated age norms Development of adult like postural control including Reactive Postural Adjustments (RPA) taking into consideration visual, somatosensory and vestibular factors, 7—10 years RPA better controlled at the age of four than at six due to the changing reliance on vision in childhood development at the age of six Anticipatory Postural Adjustments (APA) are much slower to achieve adult like status, $>$11 years of age. It is postulated that children are not efficient at regulating positive expiratory pressure in comparison with adults.
Sensorimotor	It has been shown that adults rely more on somatosensory inputs and children more on visual input. Sensory development significantly affects a child's ability to use mature motor strategies in movement.	Young children are considered to be: Predominantly dependent on vision to maintain upright posture in sitting and standing $<$6 years Transition to somatosensory reliance 6—7 years Growth rate or rate of change affects many systems and periods of rapid growth as periods of readjustment in the sensory motor system must be taken into account.
Musculoskeletal	Musculoskeletal development causes significant changes to occur over the first six years of life with $\sim$90% of adult structure being achieved at this age.	Pelvic cavity Relatively small and increased vertical diameter Fundus of bladder lies above the pubic symphysis even when empty with the anterior surface lying in contact with the lower abdominal wall. Approaching maturity at 9 years Ribcage Geometry affects the function of the diaphragm Minimal changes occur after the age of 2 Medical diagnoses, e.g. cerebral palsy and cystic fibrosis, severely affect ribcage geometry

Cognitive	Piaget's classification, although considered by many to be outdated in that is does not consider individual differences, culture and other environmental impacts, continues to be a useful tool to direct thought processes to what is possible in the paediatric population. Gardiner [30] comments that while he continues to see Piaget as a leader in the field of psychology — "I see development as permeated from the first by contingent forces pervading the time and place of origin."	The area of cognitive development is important when considering pelvic floor training in children. If we use Piaget's classification as a basic guide: the preoperational stage of 2—6 year olds makes them unlikely to respond to intervention that is not logical to them, is not able to be visualised, and that they are not really motivated to change due to the inability to extrapolate the consequences; the logical thinking of seven to eleven year olds makes it appear that we will be much more successful in the 7+ age range; but even more primed for intervention are the over 12s when children have the ability to think abstractly and to systematically plan and use deductive reasoning to direct outcomes. We must bear in mind however that some of our client groups do not get past the concrete operational stage and occasionally even the preoperational stage, e.g. clients with an autism spectrum disorder.

The external context of the task might consider the authenticity of the environment and other factors, for example the perception of parental frustration, or retributions that the child has or does receive. Subsystems that might affect the ability to train the PFMs in children, unlike adults, include the biomechanical and musculoskeletal development of the child. Consideration needs to be given to the factors described in Table 3.

In future research, consideration needs to be given not only to the types of incontinence, but how best to intervene in different age groups.

Where to from here?

The problem is how to measure PFM in children without being invasive, and to be specific about what we are measuring. To date it seems that secondary measures have been used, for example uroflow, post-void residual, severity of incontinence, and these are important. The challenge is to measure muscle function directly. It seems that the first task is to test measurement methods in the paediatric population.

Ultrasound allows structures within the pelvis to be visualised repeatedly without the concerns associated with multiple exposure to X-rays. Transabdominal ultrasound (TAUS) images movement of the PFMs and also look at the function of the rectum. It can teach the child about their problem and be used a form of biofeedback.

In adults TAUS imaging is primarily used to assess the lifting aspect of a PFM contraction by observation of movement of the bladder base as a marker for PFM activity during voluntary PFM contraction. Good intrarater and inter-rater reliability for measurement of bladder base displacement (transverse and sagittal views) during a PFM contraction (intraclass correlation coefficient [ICC], 0.81—0.88), as well as good intrarater reliability (transverse view) during functional, active, straight leg raise testing (ICC, 0.98), have been reported in adults [31].

Using transperineal ultrasound (TPUS), the position and mobility of the bladder neck have been reported to be reliable, as has change in the anorectal angle. The reference points used to generate this measurement are either the central axis or inferior margin of the symphysis pubis and the junction of the proximal urethra with the bladder. This methodology has been shown to have good intra- and inter-rater reliability (ICC, 0.76—0.98) for the measurement of bladder neck movement during a PFM contraction and Valsalva manoeuvre, and the assessment of levator function by TPUS correlates strongly with vaginal palpation of muscle strength and perineometry [32].

Using ultrasound and repeating responsiveness, validity and reliability studies in children would be a good start to establishing a gold standard for PFM assessment in children.

PFM function needs to be researched in children with particular problems: bladder overactivity, Generalised Joint Hypermobility (GJH), giggle incontinence, chronic lung disease, obesity, to name a few. Research may well need to be tailored to specific age groups, potentially 7 years and under, 7—11 years, and 12 and above, because of the developmental differences. It is expected that if PFM function is to change in children, the retrained muscle needs to be integrated into posture and other aspects of motor development and lifestyle.

Reporting of studies must conform to Template for Intervention Description and Replication (TIDieR) checklist if they are to be reproducible and of any use to inform clinicians.

Conclusion

It is hoped that more thought and informed discussion around this topic will lead to more productive research. We suggest that the research on the paediatric PFM be undertaken by those with an understanding of muscle function and dysfunction and a child's sensorimotor development. It would appear that research needs to be more directed at specific age or developmental groups and parameters more rigorously defined as using the same interventions across such a diverse population cannot be recommended. Gold standard measurements for PFM function in children need to be developed and reporting done in accordance with TDieR guidelines.

Conflicts of interest

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Multiple-choice questions

Q1. In regard to pelvic floor muscle (PFM) function, which one of the following statements is not correct:
1. PFM contribute to spinal stability
2. PFM respond to increases in intra-abdominal pressure
3. PFM work alone to augment bladder control
4. Under- or over-activity of PFM is likely to involve dysfunction of posture and breathing

Q2. The aim is to effectively measure muscle function on two separate occasions. Which one of the following statements is not correct:
1. Testing position and instructions should be standardised
2. EMG mV can be used to measure muscle function
3. The tester needs to control for action of agonists and antagonists
4. Central or peripheral fatigue can influence results

Q3. Which statement is incorrect regarding depression of the pelvic floor on voluntary activation
1. It occurs in one third of healthy woman
2. It occurs more often in incontinent woman than continent woman
3. Women with urge incontinence are the highest proportion to depress the pelvic floor
4. Children don't do this

Q4. Which statement is not correct
In childhood development, the age of six is significant because -
1. Fundamental motor skills are attained
2. There is a transition to somatosensory reliance
3. It is a time of rapid growth
4. Only minor changes occur in biomechanics after this age

Q5. Which is not a core concept of the Dynamic Systems Theory
1. Development is dynamic
2. Development occurs within a system
3. Development is non linear
4. Development occurs in discrete stages

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