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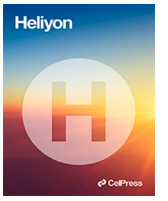

Research article

Development of the Addenbrooke's MSK screening tool (AMST) for children and adolescents with cystic fibrosis

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ABSTRACT

Background: Musculoskeletal (MSK) issues are common in the paediatric population and in people with CF. There is currently no MSK screening tool for children and adolescents with CF.
Methods: The protocol to develop the pGALS (paediatric Gait, Arms, Legs, Spine) was followed to create this screening tool. Paediatric respiratory and MSK physiotherapists, and paediatric respiratory doctors at a large teaching hospital were involved in the creation of this tool. This was then reviewed by paediatric respiratory doctors and physiotherapists at a second large teaching hospital with amendments added. One year data of this screening tool used on children over 7 was collected and analysed as a secondary outcome of this project.
Results: There were 81.8 % more positive screens for MSK issues in the newly developed screening tool compared to the year using Manchester MSK screening tool; there were also 6 more referrals all deemed as appropriate by the services referred to. Almost half of the population screened using the developed tool were positive for an MSK issue, this was commonly related to poor posture or correctable kyphosis. Kyphosis, as measured by plumb line, appeared to be associated with negative health outcomes in this population; pectoralis major length was also associated with kyphosis and these negative health outcomes.
Conclusion: The development of this tool has followed the protocol set out by the PGALs and has shown some promise with interesting initial results. This tool will need further validation before it is used in the wider paediatric CF population.

1. Introduction

Due to a variety of factors such as growth, over training and an immature skeletal system Musculoskeletal (MSK) difficulties are commonly found in childhood with approximately a third of adolescents reporting pain monthly [1]. Urinary incontinence (UI) is also commonly found in children, especially girls, with up to 47 % of them suffering it at one time or another [2], and is screened for in the Manchester Musculoskeletal Screening tool (MMST). MSK difficulties are known to increase the burden of disease in people with Cystic Fibrosis (CF), effecting up to 15 % of the population [3]. CF is the most common lethal condition in the Caucasian population effecting over 10,000 people in the United Kingdom. People with CF have stickier sputum and require regular physiotherapy to clear the airways

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[4].

The most impactful MSK issues are considered to be: Pain, postural changes, and urinary incontinence [5]. Poor posture is common in people with CF [6], with kyphosis of the spine being one of the most commonly considered MSK difficulties for people with CF. This change in spine shape, often driven by shortness in Pectoralis Major [7], can have a significant effect on lung function - a Cobb Angle increase of 0.26° associated with a 1 % drop in Forced Vital Capacity - so is often seen as significant [8]. Historically this was thought to be wide-spread, and though it is a significant issue, recent data has shown that it appears to be becoming less common [8].

Having pain is associated with reduced physical activity levels in children [9]. Physical activity is a key component of care for children with CF with it's effects on improving cardiovascular endurance, whilst decreasing the number of pulmonary exacerbations and hospitalisations [10,11]. There have been suggestions that exercise, especially in the modulator era, could be used as a replacement for PEP devices for airway clearance, with Dwyer et al. [12], reporting no significant difference in mucus clearance from the intermediate and peripheral lung regions when using exercise in comparison to PEP devices.

To address these challenges the Association of Chartered Physiotherapists in Cystic Fibrosis recommend that MSK screening is carried out in all children from the age of 7 years [13]. The MMST is validated for adults with CF [5]. However, there is no validated screening tool for MSK problems in children with CF. The pGALS (paediatric Gait, Arms, Legs, Spine) [14] is commonly used to screen

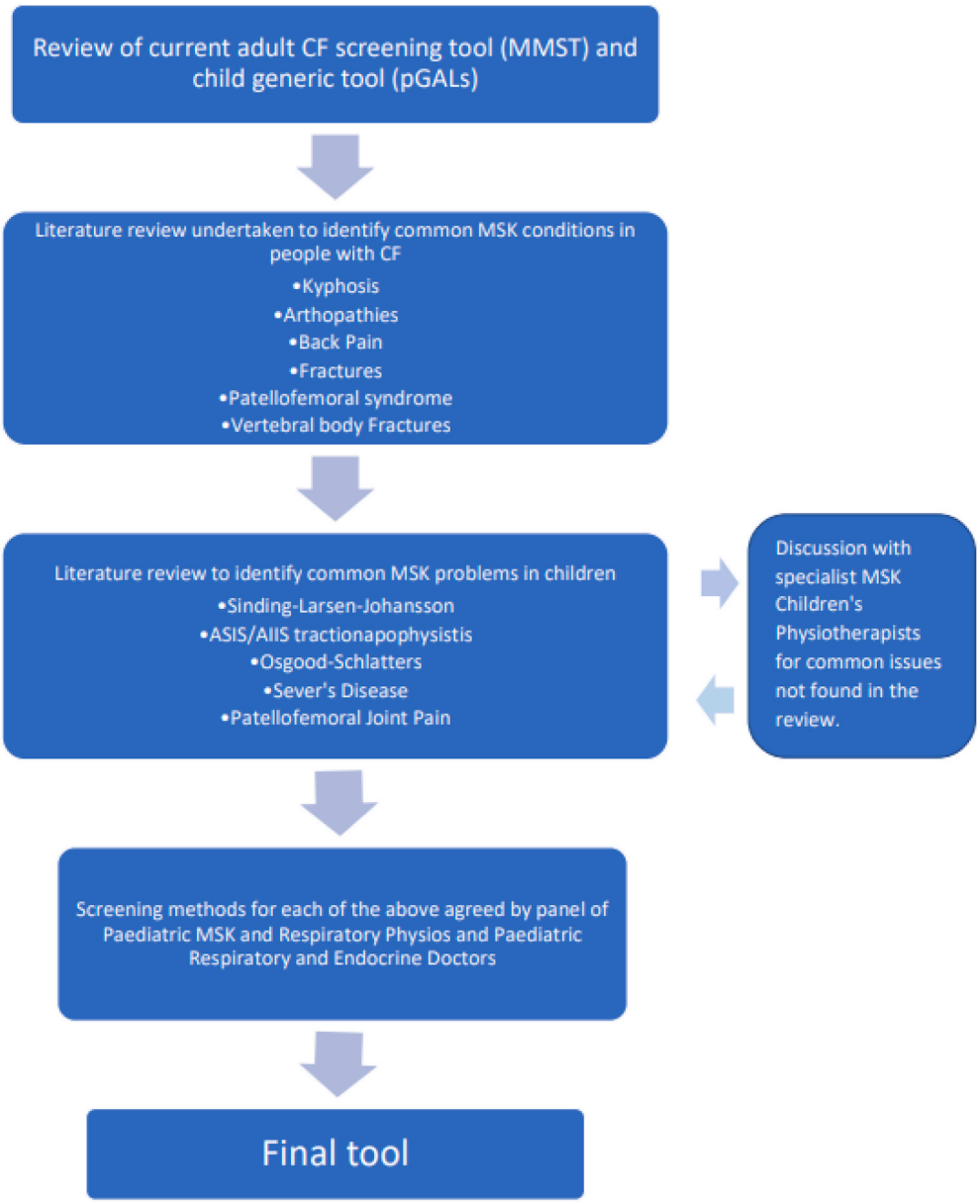

Fig. 1. Flowchart to illustrate the development of the AMST.

MSK issues in children, though is not specific for children with CF. Without a standardised tool, practice can vary and issues can be missed [15]. This paper describes the development and preliminary data of the Addenbrooke's Musculoskeletal Screening Tool (AMST).

2. Method

2.1. Development

The tool was produced following the protocol discussed by Foster et al. [14], in the development of the pGALs and is illustrated in Fig. 1. The current screening tools for both adult CF (MMST) and children (pGALs) were reviewed and appropriate assessments were identified.

A review of the literature was undertaken to identify common MSK conditions in individuals with CF which included: Kyphosis, arthropathies (generally accepted to be most frequent in children [16]), back pain, fractures, patellofemoral syndrome [17] and vertebral body fractures – which can present in children with CF and are associated with reduced bone mineral density (BMD) [18,19].

A second literature review looking into common MSK problems in children was performed. This was combined with discussions with paediatric MSK physiotherapists about the most common conditions they see. The most common relevant MSK conditions identified included: Sinding-Larsen-Johansson, ASIS/AIIS traction apophysitis, Osgood-Schlatters, Sever's disease and patellofemoral joint pain [20].

A screening method for the above conditions was collated into one document. This document was reviewed by a panel of paediatric MSK physiotherapists, paediatric respiratory physiotherapists and paediatric respiratory and endocrine doctors at a large teaching hospital, and paediatric respiratory physiotherapists and paediatric respiratory doctors at a second large teaching hospital. The panel review led to necessary minor changes to the tool (Appendix 1).

2.2. How to use the AMST

The screening tool starts with a series of questions as seen in appendix 1. The first question establishes concerns regarding posture. The second question discusses joints and is used to screen for CF arthropathies [16]. Question three screens for stress/urinary incontinence. Question four asks around back pain and is followed in objective assessment by palpating vertebral bodies of the spine to assess for increased pain which may indicate a need for DEXA or lateral thoracic x-rays to determine if there are spinal fractures. Asking about difficulty going up and down stairs reviews patellofemoral pain syndrome [17].

The plumb line is used to assess if there is evidence of excessive kyphosis. The patient is then asked to retract shoulders to see if this is correctable, if there is still rounding they are asked to shoulder flex. If there is a non-correctable kyphosis they are referred to MSK, if it is correctable then exercises are provided. Palpation of tibial tuberosity, inferior pole patella, ASIS/AIIS and calcaneal squeeze test assessed for apophysitis'.

Length of the pectoralis major is assessed with the patient in supine with the hands clasped together behind the head; the patient relaxes their shoulders, the tip of the olecranon to surface of bed is measured. Length of the pectoralis minor is assessed with the patient in supine and arms by their side. With forearms supinated, shoulders are externally rotated. The participant relaxes their shoulders, and the distance from the posterior border of the acromion to surface of bed is measured [21].

Table 1
Whole group description.

Gender (22 Female)		
Modulator (46)		
Non-Modulator (12)		
	Mean	SD
Age (58)(Years)	11.38	2.78
Pectoralis minor (57) (cm)	5.97	1.47
Pectoralis major (57) (cm)	5.56	2.80
FEV1 (%) (57)	97.83	18.23
FEV1 (L) (57)	2.49	0.86
FEV1 (Z score) (57)	−0.149	1.54
Height (cm) (58)	147.66	15.84
Weight (kg) (58)	42.30	13.1
Dexa (Z score) (25)	−0.98	1.12
	Median	Interquartile Range
FVC (%) (57)	103.50	16.70
FVC (L) (57)	2.93	1.68
FVC (Z score) (57)	0.29	1.36
Number of Intravenous Antibiotics (IVs) in past year (56)	0	1
No. Additional Orals in past year (56)	1	2

2.3. Data collection

The MSK screening tool was used at CF annual review for children aged seven or greater over a one-year period. This age cut-off was chosen as per the recommendations by the ACPCF [13]. The three CF physiotherapists that undertake annual reviews at Addenbrooke's hospital performed the MSK screening tool, results were documented in the annual review report and this data was extracted for analysis.

This project was accepted as a service evaluation project by the Cambridge University Hospitals NHS Trust's REC team and the following information was collected from electronic notes of children with cystic fibrosis aged 7 and over. Patient age, sex at birth, ethnicity, height, weight, number of antibiotics, CFTR modulator, sputum or cough swab sampling results, DEXA scan results and pulmonary function test results were taken. For comparison, the previous years' MSK screening data (if present) was also collected.

3. Results

Pilot data was collected and is presented below. As this study was not devised with data analysis as a primary outcome, "p" values are presented but should be analysed with caution as no attempt is made to control for p hacking. It is the hope that this data will provide suggestions for future research rather than readers taking firm conclusions from this.

In total 58 children with CF were screened using the developed tool. Those whose screen had a MSK issue identified where described as "Screen positive", those who didn't were described as "Screen negative". In total there were 29 children who screened positive for a MSK issue on the AMST and 29 who screened negatively. Whole group data can be seen in Table 1:.

In total nine (15.5 %) people were screened positive for kyphosis and 49 were not. While this is only pilot data tests of comparison were run between the Kyphosis + ve and the Kyphosis -ve groups as this is known to impact on lung function [8]. Independent T tests were used for parametric data and Wilcoxon used for non-parametric data (Table 3). Differences that the authors feel may warrant further investigation are highlighted bold. These include a lower mean FEV1 % and Z-score, and a longer mean Pectoralis Major in the group with a Kyphosis. There were also significantly more IV Abx and microbial growths. There was a higher proportion of males in the kyphosis group (77.8 % vs 40.8 %). There were a similar number of children and young people on modulators between the kyphosis (88.9 %) and non-kyphosis (82.2 %) groups.

There were 163.6 % more positive screen results in the year using the AMST [29] compared to the year prior using the MMST [11]. Referrals to paediatric MSK outpatients increased from zero with MMST to six with AMST. All referrals were deemed as appropriate by the services referred to, however one suspected scoliosis had self-resolved and was likely secondary to pain. Advice was given around MSK issues to 83 % more children in the year using AMST [11] compared to the year using MMST [6] – this was generally postural advice. Two patients screened positive for urinary Incontinence in the AMST, with one screening positive the year before with the MMST; none of these patients were referred onwards due to lack of service provision.

Table 2
Correlations between measured variables in AMST and health related outcomes. Correlations between measured scores were undertaken (Table 2). Spearman's rank test was used to review Age (years), height (cm), Dexa (Z-score), number of IV, Pectoralis major (Olecranon to bed in cm) and minor (Acromion process to bed in cm) length and FEV1 Z-score. Significant correlations authors suggest warrant further investigation are highlighted in this table in bold. These particularly focus on the correlations with Pectoralis Major length and FVC z-score, DEXA z-score and number of IVs.

	Age	Pectoralis Minor	Pectoralis Major	FEV1 Z	Height	No of Intravenous Antibiotics (IV Abx)	DEXA
Pectoralis Minor	r=0.276 p=0.040						
Pectoralis Major	r=0.425 p=0.001	r=0.448 p<0.001					
FEV1 Z	r=-0.024 p=0.860	r=-0.047 p=0.736	r=-0.253 p=0.063				
Height	r=0.886 p<0.001	r=0.283 p=0.035	r=0.346 p=0.009	r=-0.023 p=0.868			
No of IVS	r=-0.029 p=0.830	r=-0.034 p=0.804	**r=0.312*** **p=0.019**	r=-0.230 p=0.089	r=-0.053 p=0.693		
DEXA	r=-0.030 p=0.883	r=-0.200 p=0.308	**r=-0.410*** **p=0.038**	r=-0.019 p=0.924	r=0.236 p=0.235	r=-0.585 p=0.001	
FVC Z	r=-0.044 p=0.750	r=-0.075 p=0.586	**r=-0.339*** **p=0.011**	r=0.912 p<0.001	r=-0.090 p=0.510	r=-0.210 p=0.120	r=0.127 p=0.527

Table 3
Kyphosis and it's relation to health related outcomes.

		Kyphosis + ve		Kyphosis -ve
N		9		49
Gender Female		2		20
Asian or Asian British		2		1
On Modulator		2		37
	Kyphosis Mean (S.D)	**Non-Kyphosis Mean (S.D)**	**Independent *t*-test**	**p=**
Age (years)	12.27(1.68)	11.17(2.96)	t(56) = -1.187	0.240
FEV1 %	**86.02(26.98)**	**100.66(14.48)**	**t(55)=2.50**	**0.008**
FEV1 (Z-score)	**−1.11(2.27)**	**0.08(1.23)**	**t(55)=2.42**	**0.009**
FEV1 (L)	2.45(1.06)	2.50(0.82)	t(55) = 0.17	0.432
Pectoralis Minor (Acromion to bed in cm)	6.27(1.56)	5.90(1.56)	t(55) = -0.75	0.229
Pectoralis Major (olecranon to bedin cm)	**6.91(2.15)**	**5.24(1.57)**	**t(55)=-2.94**	**0.002**
Height (cm)	154.31(10.62)	146.11(16.54)	t(56) = -1.56	0.062
Weight (kg)	47.70(16.27)	41.04(12.19)	t(56) = -1.53	0.066
	Kyphosis Median (IQR)	**Non-Kyphosis Median (IQR)**	**Mann-Whitney U**	**p=**
FVC %	84.70(29.80)	104.95(15.00)	U = 148.00	0.034
FVC (Z-score)	−0.89(3.36)	0.29(1.56)	U = 150.50	0.038
FVC (L)	3.24(1.65)	3.81(0.91)	U = 250.00	0.952
No. of IV Abx	**2(2)**	**0(1)**	**U=49.00**	**0.035**
No of additional oral Abx	0(1.5)	1(2)	U = 84.50	0.636
DEXA (Z-score)	−1.15(1)	−0.80(1)	U = 71.50	0.541
Microbial growths in last year	**1(1)**	**0(1)**	**U=46.50**	**0.024**

4. Discussion

This is the first description of a paediatric specific MSK screening tool in CF. The tool is not validated so sensitivity and repeatability have not yet been reviewed. Pilot data has been presented with the aim that it will provide initial data to guide and power future studies but caution should be used in any interpretation of this.

The creation of the AMST shows the importance of looking outside of the traditional scope of one's practice (respiratory to MSK) for the benefit of the patient. It demonstrates respiratory specialist therapists working in a holistic approach to understand the best ways to assess and treat, with the aim of improving health outcomes for their patients.

The way this tool was developed shows the strength of multidisciplinary team working and we believe has been created in the most effective way possible. The development took a similar approach to that of the PGALS [14]:

1."Audit of adult GALS applied in school-age children." Similarly AMST was built by reviewing the current literature and based on respiratory specialist physiotherapists' experience in this area.

2."Derivation of amended version of adult GALS." Involving a paediatric MSK specialist then allows for a deeper understanding of common MSK conditions in children and the ways that these can be screened for. Involving respiratory specialist physiotherapists from other centres enabled a review of the usability of this tool and an ability to make changes collaboratively on the tool.

3."Testing of pGALS in school-age children." The AMST was then trialled in one clinic using this screening tool.

There were almost double the number of positive screens using the AMST, compared to the year prior with the MMST; while this project was not set up to comment on this, further work looking into the sensitivity and specificity of the tool is warranted. As all of these referrals were deemed appropriate by the MSK specialists receiving them, it is less likely this increase is due to oversensitivity of the tool. With this possible increase in MSK issues identified from the previous year, when using the AMST, and the changing role of the physiotherapist in the modulator era, it may indicate a necessity for physiotherapists working in CF to have a good basic level of MSK knowledge. Developing in this area may allow the practitioner to be able to provide basic advice and understand when to refer on to specialist physiotherapists. The frequency of urinary incontinence may be an under representation of the issue, with 2 of 58 being considerably lower than is documented in the literature [2], though this evidence has not been recently reviewed. However, there was only one reported in the year prior; pelvic health and incontinence can be a difficult area for children to discuss [2] which may be a potential reason for this reduced prevalence. The few patients who did screen positive for urinary incontinence were provided pelvic floor exercises by CF physiotherapist as there was no local service for these patients to be referred to for specialist care.

Pectoralis major length was significantly different between those who screened positive and those who screened negative for MSK issues. Interestingly there were no significant differences in FEV1, often described as a proxy for disease progression [22], of children with CF who screened positive for MSK issues. The high incidence of MSK symptoms in children and adolescents may be a contributing factor to these numbers rather than CF disease in itself [1].

Within this cohort, children who screened positive for kyphosis were associated with three negative health outcomes. FEV1 z-score was lower in children with kyphosis, in keeping with previous work in this area [8], with number of IVs and bacterial growths were

also significantly higher. Kyphosis was defined by plumb line rather than X-ray. The group with kyphosis also had a higher number of oral antibiotics compared to those without kyphosis. Kyphosis is likely as a consequence of worsening lung health as described by Barker et al. [8], therefore continuing to monitor this is important for children with CF. It may also be pertinent to monitor any changes in pectoralis major length during and prior to pulmonary exacerbation to see if there is a predictive value, though this is more likely due to chronic pulmonary and MSK changes.

Kyphosis and shoulder rounding was associated with pectoralis major length, in keeping with the current literature [7]. Pectoralis major length had a number of associations in this study with a significant negative correlation to DEXA z-score and FVC z-score, and positive correlation to number of IV courses. Pectoralis major length also has a positive correlation with height and age; this would be in keeping with growth of skeletal structures. Reviewing normal values of pectoralis major and minor lengths for age may be useful moving forwards.

5. Limitations

There are noteable limitations to this project. It is a presentation of the tool for the first time and it is not meant to be a validation; all data presented should therefore be reviewed with this in mind. The data collected was from one site and a small convenience sample was used. Due to the convenience sample used, we cannot say that this population was representative. The aim of this project was to create a screening tool; review of musculoskeletal outcomes was a secondary aim.

6. Recommendations

Further psychometric evaluation of this tool should be undertaken, particularly with regards validation.
A multicentre study utilising this tool would enable more representative and generalisable data.
Control values for pectoral length could be reviewed, with particular interest on specific predicting values.

7. Data availability statement

The participants of this study did not give written consent for their data to be shared publicly, so due to the sensitive nature of the research supporting data is not available.

8. Conclusion

A screening tool specific for children with CF is important in identifying conditions specific to this population, any tool used will need to be validated. Tighter pectoralis major in this population appeared to be associated with worse health outcomes. Urinary incontinence appears to be under reported in this population when comparing to other studies; there was no service to refer these patients to, which may indicate a need for physiotherapists in CF to upskill in this area. Overall, developing the MSK skills of physiotherapists working in CF will likely be important in the coming years, especially with the introduction of modulator therapy.

CRediT authorship contribution statement

Kieren Lock: Writing – review & editing, Writing – original draft, Visualization, Methodology, Conceptualization. **Sarah Nethercott:** Writing – review & editing, Writing – original draft, Formal analysis. **Colin Hamilton:** Writing – review & editing, Writing – original draft, Formal analysis. **Faye Grace:** Writing – review & editing, Methodology. **Nicholas Hall:** Methodology. **Laura Lowndes:** Writing – review & editing. **Monica Musgrave:** Writing – review & editing, Conceptualization. **Theofilos Polychronakis:** Writing – review & editing, Supervision, Formal analysis.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper. No funding or grants were received for this work.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.heliyon.2024.e25403.

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