



Review article

Alexithymia in children with medically unexplained symptoms: a systematic review

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ABSTRACT

Objective: Adult research investigating the link between alexithymia and medically unexplained symptoms (MUS) has found a significant relationship between increased alexithymia and MUS. This difficulty in expressing emotions is likely to begin in childhood so the objective of this paper is to present a quantitative review of studies focussing on the association between MUS and alexithymia in children.

Methods: Databases were searched with predefined terms relating to alexithymia and MUS in children (0–17 years). Two reviewers independently assessed abstracts, extracted data and undertook quality analyses. Systematic review methods were used in accordance with Cochrane guidelines.

Results: Ten studies met the criteria for inclusion in the review. Seven of the eight studies which focused on a comparison between children with MUS and healthy controls, found higher levels of self-reported alexithymia in the children with MUS. However, in the two studies where children were asked to complete tasks that objectively measure alexithymia, significant differences were not found. Results of studies comparing alexithymia in children with MUS and children with medical/psychiatric controls were inconsistent; there was some evidence of increased anxiety and depression in young people with alexithymia and MUS but inconsistency of measures across studies makes drawing conclusions difficult.

Conclusion: There is preliminary evidence that children with MUS have significantly higher levels of alexithymia than controls based on self-report measures; however, this finding was not replicated in objective tasks of alexithymia. Future studies should include validated tasks that objectively measure emotion recognition abilities and focus on possible mediating factors such as neurodevelopmental and mental health difficulties.

1. Introduction

Alexithymia derives from the Greek words $\lambda\epsilon\gamma\iota\varsigma$ and $\theta\upsilon\mu\omicron\varsigma$ which mean “without words for emotions” [1]. Although alexithymia was traditionally defined as a difficulty in recognizing and describing one's own emotions, evidence in adult populations suggests that alexithymia is also associated with interoceptive difficulties, such as difficulty counting one's own heart beats [2], reduced awareness of physical arousal [3], reduced accuracy on tasks of muscular effort and reduced taste sensitivity [4]. Cognitive characteristics associated with MUS have also been suggested including: difficulty in discriminating between one emotion and another and in distinguishing somatic states from emotions; difficulty in communicating one's own emotions to others; difficulties in flexible thinking; difficulties in social understanding; and an externally oriented cognitive style with an avoidance

of focus on inner experiences [5,6]. It has been hypothesised that alexithymia, particularly impairment in interoception, may be a risk factor for, or precipitate/maintain medically unexplained symptoms (MUS) [7,8].

Medically unexplained symptoms (MUS) are a heterogeneous group of physical symptoms not fully explained by a well-defined medical, psychiatric or somatic illness [9]. DSM-5 classification has helpfully moved away from the need to have no medical explanation, to incorporating cases where distress and impairment is more severe than expected even when there is a defined underlying disease. However, nomenclature and classification in this field continues to be imperfect. Conversion disorder is also under this broad category; some individuals may have – for example – both epileptic and non-epileptic seizures, others are more entirely dissociative and fit less well under the somatic symptom disorder umbrella. MUS are common in childhood and can be

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persistent and disabling [10,11]. Prevalence rates varying depending on definition measurement and age range. Domènech-Llberia found that 20% of Spanish preschoolers were affected [12] whilst Berntsson et al. found that MUS affected 20% of 7-12 year olds [13]. Synonyms or related phenomena include 'somatoform', 'psychogenic' and 'functional' symptoms. Symptoms commonly experienced by children include headache, functional neurological symptoms/conversion disorder (including non-epileptic events, sensory phenomena, motor phenomena), cognitive/psychiatric problems, fatigue, pain and gastro-intestinal complaints [11,14–18].

MUS often remain undetected and misunderstood and can lead to unnecessary and even harmful investigations being carried out [19]. In children as in adults, they are associated with significant symptoms of depression and anxiety [20,21]. MUS can also have a detrimental effect on school attendance [22] and impact on family functioning with increased anxiety, depression and decreased quality of life among parents of affected children [23]. A systematic review of the paediatric literature identified that psychological interventions are associated with a significant reduction in symptom load, disability and school absence and should be the treatment of choice for many of the young people experiencing medically unexplained symptoms [9].

There are several potential pathways by which alexithymia might influence symptom severity and treatment outcome for patients with MUS. These include limited ability of individuals with a high level of alexithymia to cope adaptively with stressful situations [24,25], which may contribute to high levels of psychological distress [26,27]. Additionally, individuals with high levels of alexithymia may become prone to functional symptoms because of a tendency to amplify, focus on and misinterpret the somatic sensations that accompany states of emotional arousal as well as other normal bodily sensations [27,28]. A meta-analysis of 18 studies found a significant positive correlation between somatisation and alexithymia in adults and a significant increase in the prevalence of alexithymia in those with MUS compared to healthy controls [29].

In summary, research has demonstrated a possible association between high levels of alexithymia and increased medically unexplained symptoms in adults. There is also some evidence to suggest that alexithymia is associated with poorer treatment outcome for both MUS and commonly co-occurring mental health disorders in adults. However, despite the high prevalence of MUS in children and young people, there have been no systematic reviews which have examined the relationship between alexithymia and MUS in this age group. The aim of this review was therefore to investigate associations between MUS and alexithymia in children and young people. Specific objectives were to determine how the relationship between MUS and alexithymia has been studied to date in the paediatric population, establish whether there is a consistent relationship between alexithymia and MUS in this group and to investigate the association between alexithymia and measures of psychological functioning.

2. Method

Systematic review methods were used in accordance with Cochrane guidelines [30].

2.1. Electronic searches

EMBASE, MEDLINE, PsycINFO and CINAHL databases were searched from inception to 25th April 2018. Independent literature searches were conducted by MHM & EM. Broadly, the search terms were categorized into three primary areas; (1) Medically Unexplained Symptoms (2) Alexithymia (3) Children. See Appendix A for full list of search terms. Search terms were derived from those used in previous studies exploring the association between medically unexplained symptoms and alexithymia in the adult systematic review [29].

2.2. Other search resources

Reference lists and citations of identified papers were also searched for relevant papers

2.3. Inclusion criteria

Studies were included where the focus was on the association between alexithymia and MUS in paediatric populations. Studies were included irrespective of whether MUS/alexithymia were defined categorically (i.e. the presence/absence of a disorder) or dimensionally (i.e. number/severity of symptoms). Papers had to be published in English and all participants in the studies were aged between 0 and 17 years. With respect to alexithymia studies were included if the scales or tasks referred directly to alexithymia or alternatively emotion awareness.

2.4. Exclusion criteria

Papers were excluded where a mixed somatoform-organic group (e.g. epilepsy together with non-epileptic seizures) was present or where it was not clear whether the sample was a mixed organic-somatoform group, which was the case for the majority of studies on chronic pain.

2.5. Data extraction

A data extraction form was developed (Appendix B) which focussed on extracting the main study characteristics such as sample characteristic, alexithymia measures, MUS measures/diagnosis, measures of psychosocial functioning & main findings and results. Data was independently extracted by two reviewers (MHM & EM).

2.6. Methodological quality assessment

Study quality was independently assessed by two reviewers (MHM and EM) using the Effective Public Health Practice Project Quality Assessment Tool (EPHPP – 31,32). This tool was chosen for its suitability in assessing a range of study designs within the area of public health research. Studies are rated as strong, moderate or weak, using predefined criteria, on a range of areas: selection bias, study design, confounders, blinding, data collection methods, withdrawals and dropouts. Total sample size is not considered. An overall total for study quality is also calculated by assessing the number of areas rated weakly (strong studies have no weak ratings, moderate ones have one weak rating and weak studies have two or more weak ratings).

3. Results

The initial search identified 52 unique papers (see Fig. 1 for search process). After screening, a total of 10 papers were found to meet eligibility criteria (see Table 1).

Eight studies compared alexithymia in groups of children who had been diagnosed with MUS versus control groups without MUS. MUS groups included children and young people experiencing: pain [33], tension-type headache [34,35], functional abdominal pain [36,37], headache and stomach-ache [38] and Chronic Fatigue Syndrome (CFS) [39]. The control groups included healthy controls in all studies but in some cases also included children with other medical/psychological difficulties or children who scored highly on a measure of somatic symptoms but without a specific diagnosis. Three studies used non-clinical samples of school children [40–42]. All studies employed a case-control design with the exception of Meade et al. [41] who used a cohort design to explore the relationship between alexithymia and child/parent reported health.

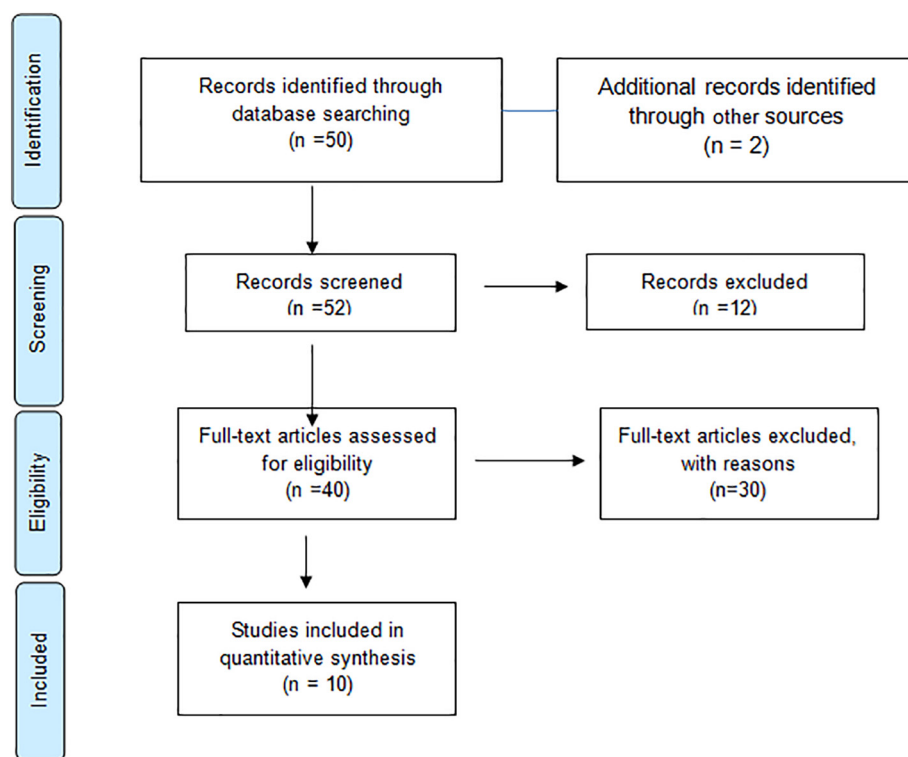


Fig. 1. Search process for studies focussing on alexithymia in children with medically unexplained symptoms (MUS).

3.1. Quality assessment

Nine out of 10 studies were rated 'moderate' with respect to quality (see Appendix B). All studies had sample sizes that were representative of the population being studied, with the exception of one study [37], which was rated weakly due to poor take up of participants in the study and the high number of withdrawals/dropouts. Six of ten studies used well validated and reliable measures [36,40,33,38,37,41] and so were rated strongly for data collection methods, whereas the four remaining used reliable measures but which have not yet been validated in child populations and therefore rated moderately [34,35,39,42].

3.2. Measures

3.2.1. Alexithymia

Four studies used the Toronto Alexithymia Scale (TAS; 43, e.g. 33,41,38,39), two studies used the Alexithymia Questionnaire for Children (ACQ; 44, e.g. 34,35) and two studies used the Emotion Awareness Questionnaire [EAQ; 36, 51,37]. All these measures are completed by the child.

There is strong support for the generalizability of the three-factor structure of TAS across languages and cultures [45]. In addition, the full-scale TAS-20 and the first two factors show adequate to good internal reliability for most of the translations when used with children [e.g., 46]. Validation studies of the AQC [44] have identified the three-factor structure of alexithymia, although the factor Externally-Oriented Thinking showed low factor loadings and a low reliability. The predictive value of the questionnaire was also satisfactory. The measure has also been found to have good cross-cultural validity [47,48]. The EAQ has also been found to have also has good predictive validity with respect to self-reported somatic complaints in children (in samples from two different countries, the UK and the Netherlands. This questionnaire has also been validated cross culturally [49].

A further study used only two of the subscales from the EAQ as part of their study together with children's performance on an experimental task [40]. Two studies utilized non-standardized tests tapping into the

emotional abilities of the child using the spontaneous attention to emotion task, identification of own emotions task or identification of an emotion in a mixed emotion situation.

3.2.2. Medically unexplained symptoms

A number of validated measures of medically unexplained symptoms were used (see Table 1) including the Children's Somatization Inventory [41], the Somatic Complaint List [36,40], the Abdominal Pain Index [37] and the Checklist Individual Strength [39]. Five studies used clinician reported diagnosis in accordance with ICD-10 or ICHD (International Classification of Headache Disorders) as their measure of MUS and referred to four conditions (tension- type headaches, migraine, somatoform pain disorder and functional abdominal pain; [33–36,38]. Psychological factors play a contributory role in all conditions mentioned above including headache/migraine onset and maintenance. For example, the migraine patient may be over focussed on their symptoms in a way that is functionally impairing, and/or their symptoms may be triggered or exacerbated by psychological factors. Psychological interventions may be useful.

3.2.3. Comorbid mental health conditions

There was significant variability in the utilisation of measures of mental health. Three studies did not use any mental health measures in children with MUS [34,35,40]. Three studies used the Children's Depression Inventory or State- Trait Anxiety Inventory for Children [e.g. 36,38,39] and one study used the Hospital Anxiety and Depression Scale [33] whilst Rieffe et al. [42] used the MOOD Questionnaire.

3.3. Synthesis of results

3.3.1. Alexithymia in children, with MUS, and healthy and clinical controls

In studies where children with MUS were compared with healthy controls, the children with MUS had significantly higher scores on at least some aspects of survey measures of alexithymia in seven of eight studies, the exception being Sayin et al. [38]. In the two studies, which involved experimental measures of alexithymia, differences between

Table 1
Studies which have focussed on alexithymia in children with Medically Unexplained Symptoms (MUS).

Year	Author	Study design	Sample N (females, age in years)	Alexithymia measures	MUS measures/diagnosis	Mental health measures	Main findings
2015	Gatta et al.	Case control	-47 (36 females, aged 8–17 yrs) with tension type headache (TTH) -42 (24 females, aged 8–17 yrs) with migraine (M) -32 (26 females, aged 8–17 yrs) Healthy Controls (HC)	ACQ	Diagnosis for TTH & M based on ICHD	None	-TTH- Alexithymic (38%)/Borderline (24%) -M – Alexithymic (12%)/ Borderline (24%) -Cont – Alexithymic (9%)/Borderline (41%) -TTH group had significantly higher scores than other two groups on measures of alexithymia
2012	Van der Veek et al.	Case control	-114 (78 female) with Functional Abdominal pain (FAP) -407 (277 female) schoolchildren with some abdominal pain (SAP) -255 (84 female) schoolchildren without abdominal pain (no AP)	EAQ	API	None	-Children with FAP scored significantly lower on most aspects of emotion awareness than children without AP although differences were small -Children with FAP scored lower than children with AP on verbal sharing of emotions
2011	Gatta et al.	Case control	All children aged 7–18 yrs -32 (26 females, aged 8–15 yrs.) with tension-type headache (TTH)	ACQ	Diagnosis for TTH based on ICHD	None	-Significantly Higher rates of alexithymia were observed in TTH group compared to controls on total score
2009	Jellesma et al.	Case control	-32 healthy controls (26 females, aged 8–15 yrs) 34 (13 females, aged 8–13 yrs) with few somatic complaints # 35 (23 females, aged 8–13) children with many somatic complaints =	- Spontaneous attention for emotions task ^a - Identification of own emotions ^a - Emotion Identification in mixed emotion situations ^a - 2 Subscales EAQ	Based on scores on SCL	None	-Compared to children with few somatic complaints, children with many somatic complaints had significantly higher self-reports of alexithymia on the differentiating emotions subscale but not verbal sharing of emotions subscale of EAQ
2007	Van de Putte et al.	Case Control	-40 adolescents (31 female aged 12–18 yrs.) with Chronic Fatigue Syndrome (CFS) -36 adolescent healthy controls (12–18 yrs)	TAS-20	Clinical diagnosis of CFS CIS-20 CIS	CDI STAIC	-Groups did not differ on a task measuring spontaneous attention for emotion -Children with many somatic complaints reported higher intensities of fear (and sadness compared to children with children with few somatic complaints - 12 (30%) CFS adolescents fulfilled the criteria for alexithymia - CFS adolescents scored significantly higher only on the total score and the subscale identifying feelings of the TAS-20 - Those with CFS and alexithymia had higher scores for depression and anxiety but similar equal scores for fatigue & somatic complaints -Children with non-organic pain were not significantly more alexithymic than depressed patients and controls
2007	Sayin et al.	Case Control	-15 children (7 female, aged 7–17 yrs) with depression -21 patients with complaints of pain (14 female, aged 7 to 17 yrs) -15 healthy controls	TAS-20	Clinical Complaints of pain without organic aetiology	BDI STAI	-In the non-organic pain group, alexithymia scores were significantly and positively correlated with STAI scores but not BDI scores -Rate of alexithymia (i.e. scores above cut off on TAS-20) in adolescents with somatoform pain disorder were significantly higher than that in healthy control subjects -Anxiety but not depression was significantly higher in somatoform pain group (62%) compared to control subjects
2006	Burba et al.	Case control	-120 children (84 females aged 12–17 yrs.) with somatoform pain disorder -60 healthy control children aged 12–17 yrs	TAS-20	ICD-10 Somatoform pain disorder	HADS	(continued on next page)

Table 1 (continued)

Year	Author	Study design	Sample N (females, age in years)	Alexithymia measures	MUS measures/diagnosis	Mental health measures	Main findings
2006	Jellesma et al.	Case control	- 33 children (16 females aged 8–13 yrs.) with Functional Abdominal Pain (FAP) - High SCL scores. 61 (31 females, 8–12 years) - Low SCL scores. 59 that scored in the (26 female, 8–12 years)	EAQ	Clinical diagnosis of FAP or constipation SCL	CDI MQ	- Children with FAP and children with more somatic complaints reported more difficulty differentiating their emotions and communicating them (on the EAQ) and a lower sense of coherence compared to children with few complaints - These two groups also reported more negative moods on Anger, Sadness and Fear scale of the MQ and on the CDI - There was no sig difference between the groups with respect to happiness - No differences between low/high SCL groups in being able to identify emotions on vignettes - Children in the low SCL group reported more anger than in the high somatic complaints groups whereas this pattern was reversed for fear - Children with more somatic complaints reported significantly more negative and less positive moods than those who reported less somatic complaints - Children who have higher levels of alexithymia have lower self-reported health but higher parent-reported health
2004	Rieffe et al.	Case control	- 282 children (141 female aged 8–13 yrs.) sampled 26 with the highest SCL scores (high SCL) and 26 with no or almost no somatic complaints (low SCL) subsequently compared	Emotional Identification Task*	SCL	MQ	
2001	Meade et al.,	Cohort Study	92 children (49 male) aged 10–13 years	TAS-20	CSI (Child and Parent)	None	

^a Spontaneous attention for emotions: - picture card shown to child and asked 'tell me something about this picture', answer rated on whether they referred to an emotion/cause of the emotion; Identification of own emotions - 4 questions regarding 4 basic emotions [59]; Emotion identification in mixed emotion situations Story with the potential of evoking multiple emotions, after the story child asked if they would feel happy/angry/sad/afraid, why they would feel this & the intensity [60], * scoring in the lowest 10% on the Somatic Complaint List, [†] i.e. the children scoring in the highest 10% on the Somatic Complaint List, [‡] Task created by researchers- presenting children with 16 emotion-evoking vignettes with tape recordings of children's responses for the q 'how would you feel?' & the intensity of this emotion, ACO - Alexithymia Questionnaire for children [44], API - Abdominal Pain Index (API); [61], EAO - Emotion Awareness Questionnaire (EAO); [50;51], CCSC-R1 - Children's Coping Strategies Checklist-Revision 1 [62], TAS-20 - Toronto Alexithymia Scale [43], BDI- Beck Depression Inventory [63], STAI - State- Trait Anxiety Inventory [64], CIS-20 - The Checklist Individual Strength [65], CSI- Children's Somatization Inventory [66], CDI - Children's Depression Inventory [67,72], STAI-C - State- Trait Anxiety Inventory for Children (STAI-C; Spielberger, 1986), LEC - Life Events Checklist [68], ICHD - International Classification of Headache Disorders (2013), SCL- The Somatic Complaint List [69], HADS - Hospital Anxiety and Depression Scale [70], SOC-13 - Sense of Coherence Scale [71], MQ-The Mood Questionnaire [44].

groups were not found. Jellesma et al. [40] found that groups reporting few or many somatic complaints did not differ on a task measuring spontaneous attention for emotion. No differences were found between those who had low/high scores on the Somatic Complaint List (SCL) regarding the ability to identify emotions on vignettes in Rieffe et al. [42].

Regarding controls who had another health condition, Gatta et al. [35] reported that children with Tension-type headache had significantly higher scores than children with Migraine and Van der Veek et al. [37] reported that children with Functional Abdominal Pain (FAP) scored lower than children with Abdominal Pain but only on one aspect of emotion awareness - verbal sharing of emotions. However, Sayin et al. [38] reported that children with non-organic pain were not significantly more alexithymic than depressed patients and Jellesma et al. [36] did not find differences in emotional functioning between children with functional abdominal pain and children with many somatic complaints.

3.3.2. Association between measures of mental health and alexithymia

Regarding measures of emotional symptoms Van de Putte et al. [39] found that those with CFS and alexithymia had higher scores for depression and anxiety compared with those with CFS without alexithymia. Burba et al. [33] reported that anxiety but not depression were significantly higher in somatoform pain group compared to control participants whilst Sayin et al. [38], reported that in a non-organic pain group, alexithymia scores were significantly and positively correlated with symptoms of anxiety but not symptoms of depression. Rieffe et al., [42] found that children with more somatic complaints reported significantly more negative and less positive moods than those who reported less somatic complaints on a MOOD Questionnaire.

Jellesma et al. [36] found that Children with FAP and children with more somatic complaints reported more negative moods on Anger, Sadness and Fear scale of the MQ and on the CDI compared to the control group with few somatic complaints. There was not a significant difference between the groups with respect to happiness.

4. Discussion

MUS in the paediatric population are distressing and impairing for children and their families, and represent a significant challenge to health service providers. Both patients and supporting health professionals often express frustration regarding diagnosis and treatment. Understanding more about the association between alexithymia and the development and maintenance of MUS and co-occurring conditions may help improve the assessment and treatment of children experiencing these difficulties. In this systematic review, we synthesized evidence from studies examining the relationship between alexithymia and medically unexplained symptoms in children. Higher levels of self-reported alexithymia in the children with MUS compared to healthy controls were found in the majority of studies but this finding was not replicated in studies where objective tasks measuring alexithymia were used. Results of studies comparing alexithymia in children with MUS and children with medical/psychiatric controls were inconsistent. There is thus some evidence of increased emotional symptoms in children with MUS and alexithymia compared to controls but studies have employed a diverse range of methods of measuring alexithymia and emotional functioning making comparisons across studies difficult. The current limited evidence does not allow firm conclusions to be drawn about the relationship between MUS and alexithymia in children and young people.

The difference in results regarding the relationship between MUS and alexithymia between self-reported measures and task-based approaches noted in this review is important. The efficiency of self-report measures and the long-standing belief that self-report provided optimal access to one's own psychological processes has kept self-report measures at the forefront in research on psychological functioning. Yet, self-

report may be limiting particularly with respect to assessing alexithymia. There is the conceptual difficulty regarding the reporting of characteristics that by definition involve limited or impaired introspection, thus raising questions about the validity of this approach [52,53]. Studies adopting task based methodology have not found a consistent association between MUS and difficulty in understanding one's own emotions and these studies suggest that it is not a difficulty in *understanding* emotions per se that might be different between children with MUS and controls, but differences in emotion processing. It is possible that children have learned what emotional experience they or another person would/should have in a specific circumstance (emotional empathy), but not spontaneously recognise when they are feeling bodily symptoms associated with the specific emotion.

Children with alexithymia would appear to be at significant risk of mental health symptoms in particular symptoms of anxiety based on results of the current review and several other studies previously conducted [54]. However, in the current review, not all studies included measure of emotional symptoms and measures varied across studies. Another factor not considered in the studies concerns the validity of measures of self-reported mental health symptoms in this population. These measures may not be useful in detecting mental health problems in children with MUS [55] and thus professional clinical diagnoses are likely to be the gold standard in the MUS population.

4.1.1. Future research and clinical implications

The higher rates of alexithymia in children with MUS compared to healthy controls based on questionnaire measures found in this review may mean that evidence based psychological interventions to treat both the MUS and comorbid mental health difficulties may need to be adapted. Children with alexithymia may benefit from specific interventions such as body awareness training/ interoceptive training [56] to enhance engagement with, and response to, cognitive behavioural therapy, but studies are needed to examine this.

Given that individuals with alexithymia may have difficulty recognizing and reporting on their own emotions the use of experimental tasks for assessing alexithymia should be a priority in future research studies. Additionally as well as self and parent report of behavioural-emotional functioning the need for professional clinical assessment and diagnoses of mental health conditions should be a priority in research. All studies to date are cross-sectional and there is a need for longitudinal data to better understand the relationship between alexithymia and MUS over time. Studies of psychological interventions for MUS in children need to include measures of alexithymia to better understand factors which might contribute to outcome in this group. It will be important to assess whether levels of alexithymia change after treatment and what this might mean for prognosis. In adults there is evidence in patients with eating disorders that alexithymia improved following treatment [56] and it may be that psychological interventions change core symptoms but also impact on alexithymia. To date studies that have included measures of behavioural and emotional functioning have included only measures of depression and anxiety. There is also a need for future studies to include measures of autism spectrum disorder [57], attention deficit hyperactivity disorder and also neuropsychological assessment data which may be related to alexithymia. Finally, little is known about the developmental course of MUS and alexithymia; this requires more research to establish whether difficulties in childhood also play out in adulthood.

5. Conclusion

There is some evidence that children with MUS have significantly higher levels of alexithymia than healthy controls based on self-report measures however, this finding was not replicated in objective tests of alexithymia and measures have varied significantly across studies.

Children with alexithymia have elevated rates of anxiety. Future studies which employ both self-report and task based measures of alexithymia and include measures of comorbid psychiatric and neuropsychological functioning are needed to better understand the possible role of alexithymia in paediatric MUS.

Declarations of interest

None

Appendix A. Search terms

Adolescent* boy* child* girl* infant* juvenile* preadolescent* pre-adolescent* preschool* pre-school* schoolchild* toddler* teen* young youth* Children* alexithymia* emotional awareness *somatisation* medically-unexplained symptom(s) somatic symptom(s)* functional somatic symptom(s)* chronic fatigue syndrome* headaches* somatoform disorder(s)* and non-epileptic seizures*

Appendix B. Ratings by reviewers using the Effective Public Health Practical Project Quality

Authors	Selection bias	Study design	Confounders	Blinding	Data collection methods	Withdrawal & dropouts	Global rating of paper
Van De Putte et al., (2007)	3	2	2	2	2	NA	2
Gatta et al., (2011)	2	2	3	2	2	NA	2
Gatta et al., (2015)	2	2	3	2	2	NA	2
Rieffe et al., (2004)	2	2	3	2	2	2	2
Sayin et al., (2007)	2	2	2	3	1	2	2
Van der Veek (2012)	3	2	3	2	1	3	3
Burba et al., (2006)	2	2	3	2	1	NA	2
Jellesma et al., (2006)	2	2	3	2	1	NA	2
Jellesma et al., (2009)	2	2	3	2	1	NA	2
Meade et al., (2001)	3	2	2	2	2	NA	2

Assessment Tool (EPHPP).

1 = Strong, 2 = Moderate, 3 = Weak.

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