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Effectiveness of Art Therapy with Pediatric Populations Affected by Medical Health Conditions:
a Systematic Review

Laura A Clapp, MSc, BSc(Hons)

Emily P. Taylor, DClinPsychol, MA(Hons)*

Simona Di Folco, PhD, MSc, BSc

Victoria L Mackinnon, MSc, BSc(Hons)

*Corresponding author

all authors: Centre for Applied Developmental Psychology, Department of Clinical and Health
Psychology, School of Health in Social Science, University of Edinburgh, Teviot Place,
Edinburgh, UK, EH8 9AG.

E-mail: Emily.Taylor@ed.ac.uk

Telephone: +0044(0)131 650 3892

Abstract

Background: Visual art therapy has been widely used with children with medical health conditions within various settings. However, less is known about its effectiveness. The scope of this systematic review is to shed light on what benefit art therapy may have for ill health adjustment in youth populations.

Methods: Electronic databases (Psychinfo, Medline, CINAHL, ERIC, Scopus, Embase, and Child and Adolescent Studies) were searched (1806-2017). Systematic methods for study selection and data extraction were used.

Results: Twelve studies (N = 404 participants, 318 in the control group; aged between 2 and 19 years old, with various medical conditions) were included. Ten studies reported significant improvements concerning at least one outcome with an overall inconclusive trend towards effectiveness.

Conclusions: Visual art therapy interventions were found weak in evidence quality. However, future evidence-based research designs could show the effectiveness of these interventions in healthcare settings.

Keywords: art therapy, adolescents, children, pediatric, chronic illness

153 words

Background

Art therapy is defined as “a form of psychotherapy that uses art media as its primary mode of expression and communication” (British Association of Art Therapists, 2016). Art is frequently used as a means of exploring and assessing the experience of children’s illness - and subsequent pain - and hospitalization (Wennstrom, Tornhage, Hedelin, Nasic & Bergh, 2013; Dolidze, Smith & Tchanturia, 2013; Chesson, Good, & Hart, 2001; Kortessluoma, Punamaki, & Nikkonen, 2008; Rollins, 2005). Findings from previous research support the use of art therapy interventions as a beneficial practice (Styles-Turbyfill, Rogers, Zink, & Kwiatkowski, 2017). Creative art therapy is the umbrella term for a range of art based modalities increasingly used in health care services to support both adults and children (e.g. drama theatre, music, writing, dance, and visual arts). The scope of what constitutes art therapy is diverse and both the approaches and the aims of delivery can vary extensively from setting to setting (Archibald, Scott, & Hartling, 2014). The theoretical framework of art in a pediatric medical setting suggests that art-based interventions can promote a sense of control and mastery (Waller, 2006) in children in unfamiliar clinical settings (Rollin, Drescher, & Kellehr, 2012). This may lead to positive outcomes such as increased coping or lowered levels of anxiety (Wallace et al., 2014).

The use of visual art therapy interventions within health care and clinical settings serve to promote psychological health for young patients in an environment which may indirectly affect and cause mental distress. Art therapy interventions rely on establishing a sense of normalcy (Malchiodi, 1999) and promote distraction and relaxation (Nainis, 2008). The key component of art therapy interventions is identified in distraction, which is further related to pain management and diversion of attention. Pain management related to medical treatments and procedures

represents a primary concern in health care settings for children, young people, parents, and health care professionals. In children and young people pain and fear of pain associated with medical diseases and treatments or therapy can further increase fear and anxiety (Stinley et al., 2015). Anxiety and fear contribute to decrease the threshold for physical pain leading to a vicious cycle, known as “the acute pain phenomenon” (Figure 1). Expressing feelings can help children and young people better cope by providing the opportunity to reflect and to find a meaning in their experiences, thus decreasing the influence of negative emotions such as fear and uncertainty. Emotional expression has been found to be one of the key strategies children and young people use to cope with stress, as it relieves tension and anxiety (Ryan-Wenger, 1992). Visual arts and creative processes linked to visual perception and image making, such as drawing, painting, and craft (Daykin & Joss, 2016), are developmentally appropriate interventions, as they engage young people in a learn-by-doing perspective and indirect expression of their feelings. A recent scoping review concluded that within paediatric populations, art therapy interventions are used in different ways and for various purposes to encourage young people to rely on art works as a communication tool for healing. The primary goal of such interventions is the intentional use of the arts to promote psychological change and as a form of therapy within a therapeutic context where the process of creating as a whole has its own meaning and communication value, taking priority over a completed art or craft product.

(See Figure 1)

A review of supportive treatments for children with chronic illness concluded that interventions focused predominantly on physical outcomes fail to address the potentially adverse psychological sequelae of ill health (Fonagy, Cottrell, Phillips, Bevington, Glaser, & Allison,

2015). A systematic investigation of therapies tackling these outcomes and ensuring appropriate targeted interventions implemented to reduce stress, enhance coping, and protect the psychological wellbeing of the young vulnerable population is essential (Verhoof, Maurice-Stam, Heymans, & Grootenhuis, 2014). However, in providing this overview, some challenges should be highlighted.

Art therapy and therapeutic art making are often grouped together, making the process and impact of art therapy difficult to understand (Angheluta & Lee, 2011). A scoping review (Archibald et al., 2014) of 16 studies investigating the use of art therapy with children living with medical conditions identified substantial variation among the studies it included, in terms of the descriptions of art therapy, the methods and designs employed, and measurement and outcomes reported. Overall, this review suggested that the use of art therapy with chronically ill children fell into two categories: 1) art therapy to foster understanding and 2) art therapy to enhance or reduce attributes. Whereas the first category comprises art therapy interventions aimed at exploring the inner world of children experiencing chronically ill health, the second one is meant to investigate the effectiveness of art therapy interventions in reducing the negative psychological outcomes of a chronic ill-health condition.

A second challenge in this process can be identified in the available art-based therapy literature, which largely consists of descriptive, qualitative evaluations and professional expertise. There is a lack of rigorous research on the contribution these therapies make specifically to healthcare outcomes (Staricoff, 2004). The breadth and value of this descriptive, exploratory research is acknowledged - within art therapy literature there is debate regarding the appropriate methodology (Daykin, Orme, Evans et al., 2008) and the evidence of effectiveness

(Edwards, 1999; Boydell, Gladstone, Volpe, Allemang, & Stasiulis, 2012). It is argued that the unstructured, person specific and qualitative nature of art therapy does not fit easily into a structured, quantitative framework (Eaton et al., 2007). However, for art therapy to be justifiably included in the commissioning of children's services, there must be robust evidence of its effectiveness and impact (Daykin & Joss, 2016). Outcome research is also required to develop an understanding of the therapeutic process and active ingredients in therapeutic art-making practices (Kapitan, 2012). Furthermore, it has to be acknowledged that previous conclusions on the outcomes were also derived from studies with adult or mixed populations with a broad spectrum of difficulties (Burleigh & Beutler, 1997; Reynolds, Nabors, & Quinlan, 2000; Slayton, D'Archer, & Kaplan, 2010; Maujean, Pepping, & Kendall, 2014). However, it is not appropriate to assume the knowledge gained from these studies can be applied to younger populations therefore more population specific research is needed. It is reasonable to hold art therapy to the same tests of effectiveness as other psychological therapies, using standard healthcare evaluation methods (Daykin & Joss, 2016), to ensure that recommended interventions meet ethical requirements (they improve health rather than having a neutral or negative effect) and that they represent the best use of limited economic resources (they are of at least equal benefit as equivalent interventions). Some psychosocial interventions lend themselves better to this evaluation framework, such as cognitive-behavior therapy, than others, which can lead to the efficacy being conflated with measurability. Evaluation of art therapy interventions are challenged by the lack of a standardised framework for intervention and outcome measurement, which makes it difficult to compare interventions and their outcomes with each other and with alternative interventions.

A systematic review to investigate the evidence and the effectiveness of visual art therapy, while identifying strengths and weaknesses, is a necessary step in recommending its use in pediatric medical settings. Based on our current research findings, no systematic evidence such as this has been provided to date. Following on from Archibald et al.'s review (2014), we focus on visual arts specifically as the most frequently used in pediatric settings; programs that are defined as play, drama, dance or music modalities are therefore excluded. Thus, the aim of this review is to identify the evidence for the effectiveness of visual art therapy in supporting positive psychological outcomes for children and young people in a pediatric context. The following research questions will be addressed: is there any evidence of the effectiveness of visual art therapy interventions on psychological and health-related conditions of children and young people in medical care settings? In drawing upon the strengths and weaknesses within this evidence, recommendations for future clinical research and practice are proposed. Taking into account Archibald et al.'s (2014) scoping work, aimed at providing a map of available literature in the field, the present review analyses the quality of the evidence base and of the effectiveness of art therapy interventions within the young population affected by medical health conditions. The present review differs from previous reviews by focusing specifically on pediatric populations with physical health diseases, excluding psychiatric disorders. This distinction has been made in attempt to reduce the diversity between studies and reported variegated outcomes, allowing a clearer synthesis of the findings to date.

Method

The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA; Liberati et al., 2009) guidelines were followed for the review. Search results were related to the period to October 10th, 2017.

The following electronic databases were searched: Psychinfo 1806-March 2017; Medline 1966 to date; Cinahl 1937 to date; Eric 1966 to date, Scopus 1960 to present date; Embase 1980 to date and Child and Adolescent Studies 1933-2017.

Keywords from three categories (intervention, population and setting) were agreed by the authors and a colleague and used to search the databases and journals: [“art therap*” OR “creative therap*” OR art-based OR “visual arts”] AND [child* OR “young people” OR adolescen* OR youth] AND [paediatric OR paediatric OR hospital* OR “chronic illness” OR pain*].

A secondary search was made of journals identified as relevant and included. Key texts and studies listed in the references of relevant articles were also searched. Key journals and websites were also searched to ensure all potential articles were retrieved.

Inclusion/Exclusion Criteria

Duplicates of the studies identified were removed (see Table 1 for details). The titles and abstracts of the remaining papers were screened according to the following inclusion criteria: a sample of children and adolescents up to 19 years old living with a medical (physical) health condition of any degree of severity and from both inpatient services and outpatient services were included; the therapeutic intervention was clearly defined as visual ‘art therapy’ and not in combination with other standalone treatments (drawing, painting, craft, digital media), facilitated

by an art therapist or a suitably trained practitioner; control groups matched for different variables were included, due to wide variations in study designs; the intervention was aimed at producing a range of measured outcomes (due to wide variation in studies).

Studies were excluded according to the following criteria: if no measured child outcomes were included, if the outcomes did not relate directly to the pediatric patient (i.e. reported only sibling/parent outcomes), if studies were evaluating art as part of a 'life program' or mixed modality creative arts program, if the art was only evaluated as an assessment tool and if participants were not actively engaging.

Due to the significant heterogeneity in the categories listed above it was not possible to integrate the data and a meta-analysis was deemed inappropriate. For this reason, a narrative review is presented. The studies included demonstrated numerous inconsistencies in the reporting of data collected and statistical analyses carried out, thus, effect sizes were included when possible (Table 2). Where studies have failed to report an effect size but have included sufficient data (means, standard deviations, and numbers in each group), Cohen's d was calculated (Cohen, 1992: $d = 0.20$, small; $d = 0.50$, medium; $d = 0.80$, large, and $d = 0.00$, no effect).

Data Extraction and data synthesis

Data extraction was carried out on the papers identified for final selection. Data extraction followed a systematic approach using a standard form developed in agreement between the authors. The extraction of data from each study was carried out by one author and checked for accuracy by another. Extracted data categories were presented in Table 1.

[Insert Table 1 here]

Quality Assessment

Quality assessment was carried out in accordance with the Effective Public Health Practice Project Quality Assessment Tool (Jackson & Waters, 2005), a reliable tool designed to evaluate heterogeneity in study design and lack of studies at randomized control trial level (RCT) (Thomas, Ciliska, Dobbins & Micucci, 2004). This tool considers the components of design was deemed more appropriate than a checklist format because the aim was to identify strength and limitation within the literature. The quality of each study for the individual components and the overall global rating was classified as either 'weak', 'moderate' or 'strong'. The tool reports upon quality across six domains: selection bias, study design, confounders, blinding, data collection methods, withdrawals and drop-outs. Component ratings and global ratings for each study are presented in Table 2. Following the EPHPP guidance any discrepancy was discussed and agreed upon between the authors.

[Insert Table 2 here]

Results

Following the application of the inclusion/exclusion criteria, 58 papers were selected for potential inclusion. These articles were reviewed in detail and 46 studies were rejected because they did not meet the inclusion criteria set (see Table 1), leaving 12 studies for inclusion in the review (see Figure 2).

[Insert Figure 2. PRISMA Flow Diagram here]

Population

As shown in Table 3, the 12 studies identified for this review reported a combined sample of 722 children and young people with chronic conditions or who were hospitalized; 404 of the children received visual art therapy and 318 were included in comparison groups. The age of participants ranged from 2 to 19 years, with four out of the twelve studies (Strafstrom, Havlena, & Krezinski, 2012; Bordonaro & Gaelynn, 2003; Broome, Maikler, Kelber, Bailey, & Lea, 2001; Favara-Sacro, Smirne, Schiliro & Di Cataldo, 2001) explicitly stating that attempts were made to account for child development by splitting children into age groups during the activities. The studies included samples with various medical conditions (Siegel, Iida, Rachlin, & Yount, 2016; Stinley, Norris & Hinds, 2015; Rollins, Drescher, & Kelleher 2012; Colwell, Davis, & Schroeder, 2005) but more frequently the research was condition specific and included children affected by HIV (Mueller, Alie, Jonas, Brown & Sherr, 2011), medical trauma (Chapman, Morabito, Ladakakos, Schreier & Knudson, 2001), Epilepsy (Strafstrom et al., 2012), Leukemia (Favara-Scacco et al., 2001), Asthma (Beebe, Gelfand & Bender, 2010), Cerebral Palsy (Wilk, Pachalska, Lipowska, Herman-Sucharska, Makarowski, Mirski & Jastrzebowska, 2010) and Sickle Cell Disease (Bordonaro & Gaelynn, 2003; Broome et al., 2001).

[Insert Table 3]

Study Design and Comparison Groups

The included studies employed a variety of study designs and demonstrated high levels of heterogeneity with regards to the control group. Five studies were described as Randomized Control Trials (RCTs) and of these, three involved non-active controls (Stinley et al., 2015;

Beebe et al., 2010, Siegel et al., 2016), one compared an active control (Colwell et al., 2005), and one study included a three-arm design with active and non-active control conditions (Broome et al., 2001). The other seven studies employed a variety of designs, including three within group studies with no comparators (Strafstrom et al., 2012; Rollins et al., 2012; Wilk et al., 2010) and a single subject design (Bordonaro & Gaelynn, 2003).

Outcomes and Measurements

High levels of heterogeneity were found across the studies with regards to the outcomes they investigated. However, to provide a systematization of identified outcomes, the effects of visual art therapy interventions on psychological and medical conditions were clustered in two areas (Archibald et al., 2014): 1) interventions aimed at enhancing an attribute or a construct associated with targeted medical diseases; and 2) interventions aimed at reducing an attribute or a health-related condition. Within the former area, positive outcomes included measurements of self-esteem (Mueller et al., 2011), self-efficacy (Mueller et al., 2011), self-concept (Colwell et al., 2005; Strafstrom et al., 2012), quality of life (Rollins et al., 2012; Beebe et al., 2010), perceived locus of control (Bordonaro & Gaelynn, 2003), coping skills (Favarras-Scacco et al., 2001; Broome et al., 2001), emotional response to hospitalization (Siegel et al., 2016), and speech production (Wilk et al., 2010). Within the second area, the benefits of visual art therapy were found with respect to depression (Mueller et al., 2011), emotional/behavioral problems (Mueller et al., 2011), PTSD symptoms (Chapman et al., 2001), pain experience (Stinley et al., 2015), and anxiety (Favarras-Scacco et al., 2001; Bordonaro & Gaelynn, 2003). Measurement predominantly relied upon child-self report, with nine studies collecting data using standardized and/or ad hoc developed questionnaires and scales. This produced a split in the quality of data

collection methods, with standardized tools demonstrating validity and reliability, whereas ad hoc developed techniques are open to interpretation bias. Further data collection methods included observation, categorization of child behaviors, physiological observations and ad hoc developed tasks.

Quality of Research Assessment

Quality of research assessment is presented in Table 2 and demonstrates considerable weakness in the methodological rigor of the studies included. Two studies were assessed as having a strong global rating for methodological quality (Stinley et al., 2015; Mueller et al., 2011) and ten studies were rated as weak (Siegel et al., 2016; Rollins et al., 2012; Strafstrom et al., 2012; Beebe et al., 2010; Wilk et al., 2010; Colwell et al., 2005; Bordonaro & Gaelynn, 2003; Favara-Scacco et al., 2001; Chapman et al., 2001; Broome et al., 2001). Common methodological areas of weakness were found in the reporting of and controlling for confounders, with all the studies (except Mueller, 2011 and Bordonaro & Gaelynn, 2003) failing to address baseline group imbalances. Detection bias was demonstrated as a common area of weakness, as all the studies failed to report if assessor blinding had been carried out. Limited reporting regarding the information children had been given as to the purpose of the interventions and/or the research questions was an area of weakness in 8 of the studies included. Among studies included, a common area of strength was the study design as only one study was rated as weak, due to its single subject design (Bordonaro & Gaelynn, 2003). Data collection methods were predominantly rated as strong as all considered studies employed standardized measurement tools reported as valid and reliable. Withdrawals and drop outs were a limitation in

the high intensity programs with poor reporting and missing data creating issues with evaluating long term outcomes.

Intervention

Definition and descriptions of visual art therapy implementation differed across the twelve studies. Given the diversity of visual art-therapy interventions, for the purpose of this systematic review, an attempt to identify a pattern or a trend within available interventions was proposed. The duration of intervention programs was selected to differentiate interventions presented in the literature. This allows fairer comparison, as it would be reasonable to expect greater effectiveness in programs of longer duration (up to a certain point). With regards to duration of program, the studies fell into two categories: 1) low intensity, single session programs and 2) high intensity, consisting of more than one session. High levels of heterogeneity were demonstrated within and across these categories with regards to: the descriptions of the visual art therapy implementation, the materials used, the activity setting and the background of the practitioner facilitating the therapy. Full details of the art therapy descriptions for each study are presented in Table 4 (supplementary).

[Insert Table 4 here]

Evidence of effectiveness

Ten out of the twelve studies reported significant improvements relating to at least one stated child outcome. Significance was reported in comparison to care as usual controls (Stinley et al., 2015; Mueller et al., 2011; Favara-Scacco et al., 2001), wait-list controls (Siegel et al., 2016; Beebe et al., 2010), active controls (Colwell et al., 2005; Broome et al., 2001) and within group

design with no comparators (Rollins et al., 2012; Wilk et al., 2010; Bordonaro & Gaelynn, 2003). Effect sizes were infrequently reported and relevant data was often missing or inconsistent. All obtainable quantitative results are presented in Table 1. Heterogeneity was high within the descriptions and definitions of the visual art therapy interventions implementation. To simplify synthesis of the findings, categories of high intensity and low intensity are presented. Methodological weaknesses that limit the validity or reliability of findings are highlighted.

Low Intensity Interventions. Low intensity interventions involved a single session of visual art therapy and the duration of these sessions varied with the longest being 90 minutes (Siegel et al., 2016). Low intensity programs predominantly involved mixed health conditions: out of the 6 low intensity studies only Favara-Scacco et al. (2001) was specifically related to Leukemia, whereas three studies (Siegel et al., 2016; Stinley et al., 2015; Rollins et al., 2012) did not provide any details. Interventions were frequently carried out on a one-to-one basis with the participant. The therapeutic aims of the low intensity programs were varied but typically related to reducing stress caused by hospitalization (Siegel et al., 2016; Rollins et al., 2012; Colwell et al., 2005) and invasive medical procedures (Stinley et al., 2015; Favara-Scacco et al., 2001). The studies related to invasive procedures and hospitalization commonly reported significant improvements related to various child outcomes, including a reduction in physical and psychological symptoms of stress during needle procedures (Stinley et al., 2015), a larger proportion of positive behaviors during invasive medical procedures (Favara-Scacco et al., 2001) and improved emotional response to hospital admission following visual art therapy (Siegel et al., 2016). All three studies used care as usual as the control condition. Hospitalized children reported significant improvements in a single area of the development, namely self-concept,

when compared to a music therapy control group (Colwell et al., 2005). Improvement in quality of life assessed by self-reports were found in hospitalized children in the absence of a control group (Rollins et al., 2012). Chapman et al. (2001) reported no significant reduction in PTSD symptoms for children following a structured art program. However, in this study art therapy was also used with the control group, thus minimizing the comparative effect directly attributable to the program. A qualitative reduction in acute stress was reported but not quantified.

High Intensity Interventions. High intensity interventions involved therapy consisting of more than one session and duration of these programs varied widely within this category. The longest program took place over six months and consisted of approximately sixty sessions (Mueller et al., 2011), whereas the shortest involved 3 hour-long sessions (Bordonaro & Gaelynn, 2003). In contrast to the low intensity interventions, these studies included samples that were condition specific (Mueller et al., 2011; Beebe et al., 2010; Wilk et al., 2003; Bordonaro & Gaelynn, 2003; Broome et al., 2001; Strafstrom et al., 2012). These interventions were predominantly carried out using a common group based approach, and varied in both therapeutic aims and the outcomes investigated. However, the outcomes were all broadly related to positive adjustment regarding chronic ill health. Significant findings in relation to this adjustment included increased self-efficacy in children affected by HIV when compared to routine care (Mueller et al., 2011); reduction in anxiety and increase in positive self-concept reports in children with Asthma when compared to wait-list controls (Beebe et al., 2010); improvements in speech production in children with Cerebral Palsy (Wilk et al., 2010), improvements in outcomes for children with Sickle Cell Disease; reduction of anxiety in single case studies (Bordonaro & Gaelynn, 2003), and changes in children's coping mechanisms, when compared to Cognitive

Behavior Therapy (CBT) and attention controls (Broome et al., 2001). Strafstrom et al. (2012) reported no significant difference in attitude towards illness related to epilepsy in a one-group design, whereas a qualitative benefit from group experience was addressed.

Discussion

This review presents the first systematic attempt to synthesize the results of outcome studies investigating the effectiveness of art therapy within a pediatric population. Overall this review cannot provide any conclusive evidence for art therapies effectiveness as an intervention to increase a variety of positive psychological outcomes associated with health conditions, management of diseases, and compliance to medical treatments within a pediatric population. The twelve studies identified for review provided inconclusive evidence of the feasibility of visual art therapy interventions within pediatric settings to support and enhance positive psychological outcomes associated with the management of chronic health conditions and compliance to medical treatments. The heterogeneity of study design, intervention model, outcomes definition, and capture reflects the breadth of impact art therapy aspires to have in the pediatric population. One of the two studies rated as strong in terms of quality, supported preliminary evidence of visual art therapy's effectiveness on psychological adjustment of children and relied on the strengths of a pre-post design, the inclusion of a control group, and a large sample size (Mueller, 2011). The second strongly rated study (Stinley, 2015), a randomized controlled trial, provided evidence of the effectiveness of visual art therapy intervention in pain and anxiety management through the collection of physiological data, in addition to self-reports. Despite considering a smaller sample ($N = 40$), findings from this study are valid, as the

physiological data provide reliable and accurate information of participants' physiological activation following the intervention.

Demonstrating robust evidence of effectiveness and impact of art therapy is critical to its inclusion in the commissioning of children's health care services (Daykin & Joss, 2016), as enhancing children and young people's compliance with routine medical treatments and interventions and positive adjustment to ill health. Overall, the systematic search and rigorous quality assessment applied by this review presents findings, challenges, and limitations that may represent an initial step towards this objective.

High/Low intensity intervention

Despite the diversity within categories of low and high intensity interventions, common themes are applicable with regards to approach and purpose. In the low intensity category, four out of the six studies had similar purpose and aimed to support children with hospital admission and/or medical procedures. However, the outcomes stated for investigation varied from study to study, including self-report of quality of life, PTSD symptoms, self-concept, mood, pain, anxiety and coping skills. In presenting the variation within this small range of studies, this review demonstrated the need within visual art therapy research to explicitly report the purpose of the art and define the quantifiable outcomes.

The high intensity category included broad themes of positive adjustment to specific chronic conditions. However, variation both in the outcomes and the tools limited the attempts to combine reports of effectiveness. The studies involved a wide range of measurement tools and predominantly relied upon measures of child self-report with nine studies collecting data using

standardized and/or ad hoc developed questionnaires and scales. This heterogeneity demonstrates the need for standardized methods of data collection that will increase validity and reliability of measurement and will enable effective synthesis of data (Deighton, Croudace, Fonagy, Brown, Patalay, & Wolpen, 2014).

Measurement- Measurements were predominantly made immediately after the therapy and any follow up measurement was hampered by attrition. This is a particular issue, which could be addressed with larger initial sampling. Data on longer-term outcomes is essential in the high intensity group where long-term adjustment to illness appears to be the key objective.

Lack of clarity in reporting therapeutic aims and not making explicit the intervention purpose complicates links to theoretical frameworks and hinders investigation of change and process. Uncertainty as to what the art is ‘doing’ and specifically, what the art aims to change, creates difficulties for the effective evaluation of outcomes. It was found that studies frequently attempted to address broad, unspecified outcomes, e.g. Siegel et al. (2016) attempted to improve ‘mood’ and Rollins et al. (2012) to promote a better quality of life. To identify the mechanisms potentially responsible for any improvements to child outcomes (Daykin & Joss, 2016) research must focus on clearly defined outcomes that are theoretically linked to the intervention. Children’s experience of ill health, pain and emotion may be mediated by support that targets stress reduction and/or enhances coping strategies (Robinson & Riley, 1999). In making a distinction between two separate therapeutic aims, relevant theoretical frameworks can be developed, research and practice can be guided by distinct objectives and specified outcomes. Although adopting such a structured framework can be seen as contrasting with the nature of

visual art therapy itself, valuing a person-to-person, developmentally flexible approach (Eaton et al., 2007; Edwards, 1999), we argue that these do not need to be mutually exclusive.

Limitations

This review is limited by the small number of studies included, by their weak quality assessment, and by their small sample sizes spanning a wide age range. Primary and secondary outcomes could not always be distinguished, as well as the setting (inpatient vs. outpatient) and the type and intensity of the intervention. Further limitations relating to controls and weak study design were evident, as well as the limitations of the studies that employed no control group in one group pre/post-test design and measured outcomes immediately following intervention. In the majority of the studies included, effect sizes were not reported.

Implications and future directions

Given the limited evidence of the effectiveness of visual art therapy interventions found in this review, there is an imperative for improvements in research design and measurements. Small and large scale randomized controlled trials, involving population of children and young people with different medical conditions, reliable and standardized measures, and comparisons groups are needed to generate evidence of visual art program's effectiveness in managing medical health conditions within a pediatric setting.

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Table 1 Exclusion Table.

Reasons for Exclusion*	Number
Population	5
Intervention	10
Control	0
Outcomes	19
Study Design	8
Non-English	3
Duplicate	1

*Reasons for rejection based on eligibility criteria categories

Table 2. Quality of Included Studies

First author	Selection bias	Study design	Confounders	Blinding	Data Collect Method	Withdrawals	Global rating
Mueller (2011)	Moderate	Strong	Strong	Moderate	Strong	Moderate	Strong
Beebe (2010)	Weak	Strong	Weak	Moderate	Strong	Weak	Weak
Rollins (2012)	Weak	Moderate	Weak	Weak	Strong	Strong	Weak
Chapman (2011)	Moderate	Moderate	Weak	Weak	Strong	Weak	Weak
Colwell (2012)	Moderate	Moderate	Weak	Weak	Strong	Strong	Weak
Strafstrom (2012)	Moderate	Moderate	Weak	Weak	Strong	Moderate	Weak
Siegel (2016)	Strong	Strong	Weak	Moderate	Weak	Strong	Weak
Stinley (2015)	Strong	Strong	Moderate	Moderate	Strong	Strong	Strong
Broome (2001)	Moderate	Moderate	Weak	Weak	Weak	Weak	Weak
Bordonaro & Gaelynn (2003)	Moderate	Weak	Strong	Weak	Strong	Strong	Weak
Favara-Scacco (2001)	Weak	Moderate	Weak	Weak	Weak	Weak	Weak
Wilk (2010)	Weak	Moderate	Weak	Weak	Weak	Weak	Weak

Rated according to the Effective Public Health Practice Project (EPHPP) Quality Assessment

Tool (Jackson & Waters, 2005).

Table 3. Study Characteristics.

First Author	Study Design	Population/sample	Medical population	Number of sessions Intervention intensity	Outcomes	Measures	Results	Effect sizes
Mueller (2011)	Quasi-experimental cross sectional evaluation	Age: 8-18 (N) 297 2 group AT (177)/CAU (120) pre/post design	HIV affected children in South African community	n: 50 H	Self-esteem Self-efficacy Depression Emotional/behavioural problems	Parent and child self-report: RSES, SEQC, CDI, SDQ, Adapted AIDS Stigma Scale, Non-standard study-specific questions.	No significant association between AT and depression, increased self-esteem or emotional/behavioural problems.	improved self-efficacy $B=3.61$, ($p=0.02$)
Rollins (2012)	Mixed methods	Age: 5-19 (N) 50 1 group pre/post design	Various conditions	n: s L	QOL	Parent and child self-report and observation: PedsQL, Investigator developed parent survey	Significant improvement in QOL.	N.A.

Chapman (2001)	Prospective random cohort design	Age:7-17 (N) 85 3 groups; AT(31)/CAU(27)/No PTSD ¹ pre/post	Pediatric trauma patients	n: s L	PTSD symptoms	Parent and child self-report at 1 wk/1 mnth/6 mnth: PTSD-1	No significant differences between groups.	
Colwell (2005)	Randomised trial	Age: 7-18 (N) 24 2 group AT(12)/MT(12) pre/post	Various conditions	n: s L	Self-Concept	Parent and child self-report: PHCSS	Significant difference for AT in 1 category $F(1, 21 = 6.2, p = .021)$	
Srafstrom (2012)	Mixed methods	Age 7-18y (N) 16 1 group pre/post	Epilepsy	n: 4 H	Self- image	Parent and child self-report, drawing task and assessment: PHCSS, CATIS, ICNDS, SDT, LECATA, FEATS	No significant differences in pre-post attitudes towards illness Reports a qualitative benefit	
Siegel (2016)	Pilot study,	Age 3-17 (N) 25	Various	n: 1 L	Emotional response to hospital	Child self-report by author developed questionnaire,	Improved mood ($p = 0.07$)	N.A.

¹ 27 children enrolled showed no PTSD symptoms and were not assigned to active or control groups.

	randomised	2 group (art 13/wait list 12) pre/post	conditions			measures of cortisol in saliva, Parent interviews 3-6 month post discharge	Significance set at >0.1 level	
Stinley (2015)	RCT	Age 7-18 (N) 40 (AT 20/CAU 20)	Children undergoing painful procedures	n: 1 L	Pain Anxiety	Physiological data and Child self-report W-BVAS HFRS.	Significantly reduced anxiety and physiological stress	Anxiety within treatment group $d = 0.809$, ($p = 0.06$); (change in heart rate/blood oxygen) $d = -0.24$ -.051 $p < 0.1$
Favara-Scacco (2001)	Pilot study	Age 2-14 (N) 49 2 group ACT 32, CAU 19 pre/post	Leukemia patients during blood test	n: 1 L	Coping skills Anxiety	Observation and categorization of child behaviours.	AT produced a larger proportion of positive behaviour. No analysis	N.A.
Beebe (2010)	RCT	Age 7-14 (N) 22 2 group AT 11/WL 11 pre/post	Asthma	n: 7 H	Outcomes relating to HRQOL (anxiety, self-concept)	Parent and child self-report pre/post/6mth FEATS, PedsQL (Asthma), HRQOL (Asthma), BYI	Significant decreased anxiety and worry Increased score self-concept Maintenance of some positive	Worry post-test $d = 0.9$, $p = .0142$ / $d = 0.98$ $p = .0279$; Anxiety post-test $d = -1.9$ $p = .0388$ / $d = -0.8$;

							changes at 6 months	Self-concept $d = 1.4, p = .0222$
Wilk (2010)	Retrospective	Age 12-18 (N) 14 1 group pre/post design	Cerebral palsy (Dysarthria)	n: 112 H	Speech production -linked to psych-social adjustment	Verbal fluency test ADS	Significant improvement reported in 9 speech categories ($p = <0.001$) No information on statistical testing.	N.A.
Wolf Bordonaro (2003)	Single subject design	Age 6-9 (N) 3 pre/post	SCD	n: 3 H	Anxiety LOC	Observations and child self-report CHS, ABS, CHLCS	Reports reduction in anxiety No statistical analysis reported.	N.A.
Broome (2001)	Randomised group design	Age 6-12 /13-18 (N)75 ² CBT/AT/ AC Pre/post	SCD	n: 4 H	Coping strategies	Child self-report SCSI, A-COPE	Coping strategies changed	N.A.

² Numbers of children assigned to active or control group not detailed (inconsistent data).

ABBREVIATIONS- ABS: Anxiety Behaviour Scale, AC: Attention control, A-COPE: Adolescent Coping Orientation for problem experiences, ADS: Auditory Dysarthria Scale AT: Art therapy, BYI: Becks Youth Inventory, CATIS: Child Adjustment to Illness Scale, CATTI: The Chapman Art Therapy Treatment Intervention, CAU: Care as usual, CBT: cognitive behavioural therapy, CDI: Child Depression Inventory, CHLCS: Children's Health Locus of Control Scale, CHS: Children's Hope Scale, FEATS: Formal Elements Art Therapy Scale, H: High, HFRS: Hospital Fears Rating Scale, ICNDS: Impact of Child Neurological Disability Scale, L: Low, LECATA: The Levick Emotional and Cognitive Art Therapy Assessment LOC: Locus of Control, MAD: Make A Difference About Art Programme, MT: Music Therapy, n: number, N.A.: Not Available, no effect size reported or calculated due to insufficient data, PedsQL: Present Functioning Visual Analogue Scales, PHCSS: The PIERS-Harris Self-concept, PTSD-1: The Children's Post Traumatic Stress Disorder Index, PTSD: Post Traumatic Stress Disorder, QOL: Quality of Life, RCT: Randomised controlled trial, RSES: Rosenberg Self-esteem Scale, s: single session; SCD: Sickle Cell Disease, SCSi: School-agers Coping Strategy inventory SDQ: Strengths and difficulties Questionnaire, SEQC: Self-efficacy Questionnaire for

children, SDT: Seizure drawing task, W-BVAS: Wong-Baker visual analogue scale pain instrument, WL: Wait List

Table 4. Diversity within pediatric art therapy interventions.

First author	Duration	Activity and Materials	Purpose	Setting	Outcomes
Mueller (2011)	6 months – 50 sessions High intensity	‘Make A Difference’ structured program Including; art with various materials, education activities, making a ‘hero’ book	To build self-worth, empowerment, emotion control	Outpatient: school based project in deprived community in South Africa	Self-esteem Self-efficacy Depression Emotional/behavioral problems
Rollins (2012)	Single session 45-60 mins Low intensity	By proxy drawing by therapist based on ‘The Moon Balloon’ book, therapist draws images selected by child	To encourage children to choose images and color with aims to explore/communicate emotions	Inpatient: Pediatric unit in university-affiliated hospital	General quality of life
Chapman (2011)	Single session 60 mins	Author developed CATTI- structured drawing program specially designed for medical trauma	To encourage expression towards traumatic event in order to validate	Inpatient: Bedside activity in hospital room	Post-traumatic Stress symptoms

	Low intensity		responses and reduce anxiety		
Colwell (2005)	Single session 45-60 mins Low intensity	Free drawing with pencils, markers, oil pastels, narrative description of experience, framing of work	To explore media, therapist available to answer questions and 'brainstorm'	Inpatient: Pediatric unit of teaching hospital	Self-concept
Strafstrom (2012)	4 x 1.5hr sessions over 1 month High intensity	Thematic structured therapy program drawing/painting/col lage/digital media activities with other children	To meet others, share experience/feelings, provides closure, identifies positive aspects of participant and situation, achievement of goal, discuss future goals	Outpatient: Family center	Self-image
Siegel (2016)	Single session 90 mins	Craft, creating sock puppets with buttons, needles, filled with 'magic beans'	To encourage expression of hopes/worries, aim to help children communicate and	Inpatient: pediatric unit	General mood – in response to hospital

	Low intensity		understand hospital experience, written wishes placed inside puppet		
Stinley (2015)	Single session Low intensity	Mandala making on I pad using touch screen	To reduce stress experienced during procedure	Outpatient: Working phlebotomy clinic	Pain Anxiety
Favara-Scacco (2001)	1hr session Low intensity	Drawing/visual imagination/medical role/clinical dialogue, adapting modality to suit individual needs	To prepare/support/Comfort patients during intrusive medical procedures	Inpatient: Treatment room, pediatric unit, university hospital	Coping skills, Anxiety
Beebe (2010)	1hr weekly, 7 weeks	Structure and themes each week including 'feelings related to illness' and 'healthy expressions of	To encourage illness related topics like experiences, anger felt, pain	Outpatient: School based	Health related quality of life

	High intensity	anger' children making art with rang of materials, discussing feelings and art created	management and coping		(component s)
Wilk (2010)	90 mins daily, 5 then 2 days per week, 16 weeks High intensity	Learning about art, using materials and creation using drawing/modelling/p ainting/graphics /sculpture	To encourage conversation, to evoke openness, sense of play, self-expression, provide supportive, positive atmosphere, display work to public	Outpatient: Reintegrati on training center for patients with brain dysfunctio n	Speech production
Wolf (2003)	3x 1hr sessions High intensity	Various art materials, collage making, drawing, games, discussion	To provide opportunity for self-expression and learning, provide safe space to process emotional experience of illness	Inpatient: Activity room of pediatric unit	Anxiety, Locus of control

Broome (2001)	Wily sessions, 4 weeks High intensity	Various materials, African art, drawing, develop a group mural with individual images	To provide opportunity to express feelings of pain experience in supportive sharing environment, develop socials skills, develop coping	Outpatient: Classroom like setting at SCD centers	Coping strategies
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Figure 1 Model of Art Therapy in Management Pediatric Chronic Pain

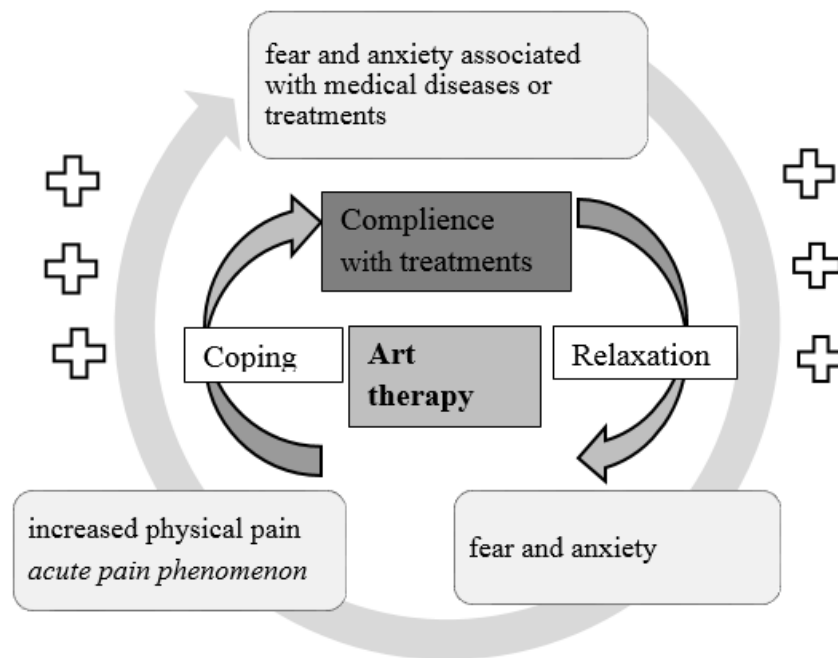


Figure 2. PRISMA Flow Diagram

