Review

What solutions exist for cognitive, emotional and developmental delays facing Indigenous children globally? A co-designed systematic review

Rona Macniven 1, 2, 3*, Thomas Lee Jeffries Jr. 2, David Meharg 2, Folau Talbot 2, Boe Rambaldini 2, Elaine Edwards 4, Ian B. Hickie 5, Margaret Sloan 4 Kylie Gwynne 2, 3

1 School of Population Health, The University of New South Wales, New South Wales, Australia r.macniven@unsw.edu.au
2 Poche Centre for Indigenous Health, Faculty of Health and Medicine, The University of Sydney, New South Wales, Australia tljeffri@gmail.com; david.meharg@sydney.edu.au; folau.talbot@sydney.edu.au; boe.rambaldini@sydney.edu.au
3 Faculty of Medicine, Health and Human Sciences, Macquarie University, New South Wales, Australia kylie.gwynne@mq.edu.au
4 Toomelah Public School, Bruxner Hwy, Boggabilla NSW 2409, New South Wales, Australia margaret.cobb@det.nsw.edu.au
5 Brain and Mind Centre, The University of Sydney, New South Wales, Australia ian.hickie@sydney.edu.au

* Correspondence: r.macniven@unsw.edu.au

Abstract: Early childhood is important for future cognitive and educational outcomes. Programs overcoming barriers to engagement in early education for Indigenous children must address family cultural needs and target developmental delays. This systematic review identifies culturally adapted programs to improve cognitive, emotional and developmental delays among young children, in response to an identified priority of a remote Indigenous community. Five databases (the Cochrane Library, Embase, Medline, Scopus and CINAHL) were searched for English language papers in January 2018. Study quality was assessed and findings analysed thematically. Findings were presented to the community at an event with key stakeholders, to determine their inclusion and face validity. Seven relevant studies, published between 1997-2013, were identified by the researchers and each study was supported by the community for inclusion. Two studies focused on Native American children and five studies included children from non-Indigenous disadvantaged backgrounds. Findings were reported narratively across four themes: Storytelling to improve educational outcomes; Family involvement improved development; Culturally adapted Cognitive Behavioral Therapy to reduce trauma; Rewards-based teaching to improve child attention. Limited published research on culturally adapted and safe interventions for children with cognitive, emotional and developmental delay exists but these four themes from seven studies identify useful components.

Keywords: Child, Preschool, Language, Reward, Attention

1. Introduction

Educational outcomes are correlated with broad and optimal health and social outcomes. Individuals who complete secondary school are more likely to gain employment, have stable housing, are less likely to have risky behaviors such as substance misuse or tobacco, and as such have improved health throughout the lifespan [1, 2]. Indigenous young people are less likely than non-Indigenous young people to complete secondary education [3] due to a range of factors as wide ranging as access to transport through to entrenched racism. In Australia, the Closing the Gap strategy is a whole of government policy intended to address the disparities in health, education and other key life outcomes of Indigenous people. Secondary school completion rates are a priority and whilst year 12 completion rates for Indigenous people have improved over the past decade, there
remains a significant gap of 24% between Indigenous and non-Indigenous students (65.3% versus 89.1%) [4].

The importance of early childhood education for future cognitive and educational outcomes is well established, particularly for children from low socioeconomic backgrounds [5] who are overrepresented among Indigenous populations [6]. Critical to successful school completion is establishing the foundations of learning including literacy, numeracy and self-regulation but there are several barriers to establishing these foundations. These include negative past experiences in education of parents and care givers which have made them distrustful of the school environment and subsequently, reluctant to engage children in education [7]. The curriculum often does not reflect Indigenous culture and experience of Indigenous children and families and is therefore less relevant [8]. Further, the cost of purchasing uniforms and other requirements for school may be a barrier for low income families; low incomes are more common among Indigenous families [6]. These barriers combine to impact on enrollment, attendance, participation and success in education [8].

In Australia and internationally, programs have been developed to address the barriers to engagement in early education for Indigenous children and families [9]. These programs have found success when they address the cultural needs of the child and their family in the curriculum. However, an additional barrier to engagement in early education and subsequent educational outcomes is developmental delay. Developmental delay, or vulnerability, is 2-3 times more likely to be experienced by Indigenous children [10]. Traumatic experience in childhood is a strong determinant of developmental delay [11]. Trauma-related behaviours are more common in disadvantaged and disengaged communities and for Indigenous people, the combined effects of colonisation and subsequent harmful government policies and practices such as child removal have contributed to higher population levels of trauma [12]. While trauma is a contributor to developmental delay, the high rates in Indigenous communities are likely to be the consequence of a range of environmental exposures in pregnancy and early childhood.

This review aims to identify culturally adapted interventions to improve cognitive, emotional and developmental delay among young children. The review is undertaken in response to an identified priority of a remote Indigenous community to develop a program to enhance educational outcomes for young children living in this community.

2. Materials and Methods

The researchers were approached by a small (<300 population) Indigenous community in central northern New South Wales, Australia with whom the researchers had a prior relationship to conduct co-designed program delivery and evaluation. The focus of this program was to identify culturally relevant solutions to improve trauma-related developmental delay in Indigenous preschool children. Through a series of community meetings between the researchers and community representatives, the need to undertake a systematic review of international evidence to inform culturally relevant solutions was determined.

2.1 Protocol and registration

This systematic review was registered with the International Prospective Register of Systematic Reviews (PROSPERO; Registration no. CRD42018088752), and was conducted and reported following the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement Search Strategy [13].

2.2 Search strategy and inclusion criteria

Five peer-reviewed, relevant online databases were searched; the Cochrane Library, Embase, Medline, Scopus and CINAHL. The search was conducted between January and May 2018. A panel comprising four of the authors gathered to determine the inclusion and exclusion criteria. Search terms to identify global Indigenous groups were used including Oceanic ancestry group, Indigenous, Aboriginal, First Nation people, Native, Inuit, Sami, Indian, American native, African
and European continental ancestry group. Search terms further stratifying the condition or domain being studied were added; neurological and developmental delay, social-emotional development, physiological development, chronic stress, emotional regulation, social competence, hyperarousal, language delay, and stress disorder. We added pre-school aged children and finally terms around intervention to include; therapeutics, early intervention, solution, service, program. All types of study design were considered for inclusion. There were no date restrictions and our search was limited to English language publications. A full search strategy for MEDLINE is provided in Supplementary file 1.

2.3 Data extraction

The panel met to examine the abstracts of all studies that met the inclusion criteria and conduct a risk of bias (quality) assessment. The articles were assessed using the Critical Appraisal Skills Programme (CASP) tool [14] for qualitative research. Data on the study population, type of developmental delay, aim, intervention, findings and main themes were extracted. Data were independently extracted data from each article by one author and were then independently assessed by another author. The studies were then discussed by the panel to determine final inclusion or exclusion of studies with any doubts resolved by discussion.

2.4 Data synthesis and interpretation

Upon completion of the database search and quality assessment we conducted a content analysis [15] of the findings from the relevant publications to identify common themes. The findings of the potential studies for inclusion were presented to the community at a face to face event with key community stakeholders in order to determine their final inclusion and face validity. Following this meeting, the findings were finalized by the researchers.

3. Results

Our search yielded 326 publications (after exclusion of duplicates). The titles of the 326 publications were reviewed by the primary researcher. A total of 301 publications were initially excluded because they did not fall within inclusion criteria. The abstracts of 25 publications were reviewed by a panel of four of the authors against inclusion criteria and 10 articles were excluded. The panel separately examined the 15 full-text studies and met together and determined that while none of the studies met each of the inclusion criteria, seven studies were considered to have relevant learnings for the development of the community program. The PRISMA flow diagram for the study is shown in Figure 1.
Records identified through database searching (n = 374)

Additional records identified through other sources (n = 0)

Records after duplicates removed (n = 326)

Records screened (n = 25)

Records excluded (n = 10)

Full-text articles assessed for eligibility (n = 15)

Full-text articles excluded, with reasons (n = 8)

Studies included in qualitative synthesis (n = 7)

Figure 1. PRISMA Flow Diagram

Two studies focused on Indigenous children [16] [17] and five studies included children from non-Indigenous disadvantaged backgrounds, specifically Hispanic [18, 19], African American [20], rural Indian [21] and refugee [22] children. These study findings were presented to the community for their views on their relevance and inclusion in the program design and all seven studies were considered relevant for inclusion by the community and were therefore included in the review. The CASP Qualitative Analysis of the included studies is presented in Supplementary file 2.

Due to the heterogeneity of study designs, a meta-analysis could not be performed, and the findings are reported narratively around key themes. The seven studies were published between 1997 and 2013. Sample sizes varied considerably; five studies took place in the United States [16-20], one took place in the UK [22] and one took place in India [21]. Of the seven studies, two were case studies [16, 18], two were cross-sectional or exploratory studies [21, 22] and the remaining three were intervention studies [17, 19, 20]. The citation, study population, type of developmental delay, study aim, design & methodology, intervention, findings and main theme for each study is presented in Table 1.
<table>
<thead>
<tr>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Population (N; race, age)</td>
<td>48 Native American, 6th grade (11-12 years)</td>
<td>1 Native American, 14 years</td>
<td>1196 Urban African</td>
<td>3668 Rural Indian, 0-6 years</td>
<td>2 Hispanic, 6 and 8 years</td>
<td>47 Refugee, 4-19 years</td>
<td>5 Hispanic (4 years)</td>
</tr>
<tr>
<td>Type of Developmental Delay</td>
<td>Psychological symptoms</td>
<td>Trauma exposure related</td>
<td>Behavioural disturbance</td>
<td>Psychosocial and physical delay</td>
<td>Psychosocial reactions to single event trauma</td>
<td>Psychosocial</td>
<td>Developmental disabilities</td>
</tr>
<tr>
<td>Study aim</td>
<td>Decrease psychological trauma or PTSD symptoms</td>
<td>Propose a form of culturally adapted trauma focused therapy</td>
<td>Improve positive classroom outcomes</td>
<td>Assess the psychosocial development of well-nourished and malnourished children</td>
<td>Determine whether a novel type of therapy be adapted to a cultural context</td>
<td>Assess intervention effects of for poorly adjusting children</td>
<td>Improve the storytelling ability of learning delayed children</td>
</tr>
<tr>
<td>Design</td>
<td>Pre-post study</td>
<td>Pre-post study</td>
<td>Cluster randomised controlled study</td>
<td>Cross-sectional study</td>
<td>Case study</td>
<td>Exploratory study</td>
<td>Pre-post study</td>
</tr>
<tr>
<td>Intervention</td>
<td>Trauma based, culturally adapted</td>
<td>Trauma based, culturally adapted</td>
<td>Good Behavior</td>
<td>Game</td>
<td>N/A</td>
<td>Storytelling to guide therapy</td>
<td>Use of storytelling computer software to improve</td>
</tr>
<tr>
<td>Study</td>
<td>Control group</td>
<td>Treatment group</td>
<td>Cluster randomised controlled study</td>
<td>Cross-sectional study</td>
<td>Case study</td>
<td>Exploratory study</td>
<td>Pre-post study</td>
</tr>
<tr>
<td>Findings</td>
<td>Improvement in psychological symptoms</td>
<td>Improvement in classroom outcomes</td>
<td>Improvement in psychosocial development</td>
<td>Improvement in storytelling ability</td>
<td>Improvement in mental health outcomes</td>
<td>Improvement in school; support from parents at home</td>
<td>Improvement in storytelling computer software to improve</td>
</tr>
<tr>
<td>Conclusion</td>
<td>Treatment was effective</td>
<td>Treatment was effective</td>
<td>Treatment was effective</td>
<td>Treatment was effective</td>
<td>Treatment was effective</td>
<td>Treatment was effective</td>
<td>Treatment was effective</td>
</tr>
<tr>
<td>Findings</td>
<td>Therapy</td>
<td>Therapy</td>
<td>Storytelling ability</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>----------</td>
<td>---------</td>
<td>---------</td>
<td>---------------------</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Improved life equivalent scores and decrease child Post-Traumatic Stress Disorder symptom score</td>
<td>Paternal childcare involvement especially, spending time, telling stories, taking child for outings important for positive psychological sequelae of trauma</td>
<td>Can provide a culturally appropriate intervention to treat the from mental health services improved in peer problem scoring</td>
<td>Children receiving direct intervention Improved storytelling with Story Champs software</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Findings</td>
<td>Therapy</td>
<td>Therapy</td>
<td>Storytelling ability</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>----------</td>
<td>---------</td>
<td>---------</td>
<td>---------------------</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Culturally adapted CBT may ease psychological distress in adolescents exposed to trauma</td>
<td>Physically and psychosocially well-nourished children do be effective therapy in overcoming trauma in children</td>
<td>Play therapy, drawing, storytelling may be effective</td>
<td>Mental health services and family support may benefit poorly adapted children in the school setting</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Main Theme</td>
<td>CBT may be a good form of treatment for adolescents who have been produce long term positive outcomes</td>
<td>Story telling can improve the cognition and learning ability of children if adapted prior to formative education</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Table 1: Characteristics of included studies
Four common themes were identified within the seven publications. Firstly, storytelling for children with a traumatic past can be utilized as a tool for literacy improvement. Secondly, a family unit with a father figure showing interest in his child’s education has been shown to improve child development. Thirdly, culturally adapted cognitive behavioral therapy (CBT) has been successfully utilized in children for whom trauma is a barrier to educational development. Finally, a rewards-based teaching method may be an effective way to improve individuals’ attention in class thus contribute to improving educational outcomes.

3.1 Theme 1: Storytelling to improve educational outcomes

Four publications identified storytelling as a method to improve educational outcomes in children with developmental delay. Within these, three were focused on gradual exposure to overcome traumatic experience, two of which were descriptive studies [16, 18]. Both proposed narrative play and storytelling as gradual exposure-style therapy as potential solutions to alleviate emotional stress tied to memories of trauma. The third study was an intervention using storytelling to improve school preparedness in Hispanic preschoolers [19]. With the use of software known as Story Champs the authors conclude that three of the four students improved their story telling in several categories. A further study used storytelling as an intervention for unspecified learning delay aimed to provide evidence that better nutritional status leads to better learning outcomes [21]. An additional finding was that children whose fathers read to them led to better learning outcomes.

3.2 Theme 2: Family involvement improved development

The second theme was based on two papers with findings that family involvement improved emotional and psychosocial development of children during their transition period into school. One study indicated that refugee children living in the UK who were having difficulties with post-traumatic stress disorder (PTSD) and cultural identification at school showed academic improvement when their parents were willing to take part in finding solutions with the teachers at school [22]. The second study revealed that young, rural Indian children showed higher levels of psychosocial development if their parents spent leisure time with them, additionally if their fathers took them on outings and if their fathers read to them [21].

3.3 Theme 3: Culturally adapted Cognitive Behavioral Therapy to reduce trauma

Two of our studies focused their interventional approach on culturally adapted trauma focused cognitive behavioural therapy (CBT). The first, a quantitative study, aimed at decreasing psychological childhood trauma in 6th graders successfully utilized adapted CBT to improve life equivalent scores (LES) and decrease child PTSD symptom score (CPSS) [17]. The second study utilized culturally-adapted trauma-focused CBT to address years of psychological trauma leading to unspecified developmental delay and incorporates elements of Native American spirituality in the treatment [16].

3.4 Theme 4: Rewards-based teaching to improve child attention in an educational setting

One study presented a unique solution for classroom behavior intervention, known as the ‘Good Behavior Game’ (GBG) [23]. This study looked at the long-term outcomes of the intervention on students growing up in high-risk areas during their early formative education [20]. The intervention encouraged a positive learning environment by incentivizing good classroom behavior with peers and teachers. The game was introduced gradually, and rewards were dependent on overall class behavior, encouraging a group-based atmosphere. The study presented results 15 years after students participated and showed lower levels of incarceration and drug-seeking behavior among participants.
4. Discussion

We found seven studies of culturally adapted interventions to improve cognitive, emotional and developmental delay among young children that were captured within four unique themes. While descriptive studies about cognitive, emotional and developmental delay among priority populations are useful in understanding the issue [24], very few studies propose solutions to the extensive and complex issues surrounding developmental delay. Our study highlights the importance of identifying solutions to such a complex issue and was undertaken in response to an identified priority of a remote Indigenous community to develop an evidence-based program to enhance educational outcomes for children.

Of the seven studies, two included Indigenous populations; both of these were American Indian and Alaska Native children and the remaining five studies included culturally diverse population groups; African American, Hispanic, rural Indian and refugee children. The findings from each of these studies were considered by the Australian Indigenous community to be relevant to their community. Each of these population groups experience disparities across many health and educational outcomes [25] and are typically under-represented in research, limiting the evidence base of effective ways to improve health and education equity.

We found that storytelling can improve educational outcomes, with evidence from two studies, both of which focused on Hispanic children, the former having experienced single event trauma and the latter with broader developmental disabilities [18, 19]. The Story Champs storytelling intervention [19] has previously demonstrated clear improvements for children on narrative retell tasks and moderate improvements on personal generation tasks [26]. The present study within this review enhanced these findings with evidence of retell and personal storytelling practice, on story retells, personal stories, and story comprehension [19]. The other study found using super heroes as ‘magical realism’ and a culturally relevant metaphor to overcome trauma [18]. While both studies have small sample sizes which may limit their generalizability, they provide some evidence that storytelling is a culturally relevant method that can benefit children with different forms of developmental delay and may benefit Indigenous children. Further, the oral nature of storytelling culturally relevant and valued within Indigenous communities [27] which further supports its inclusion in program design.

We also found that family involvement improved development, based on two studies with rural Indian [21] and refugee children based in the UK [22]. The Indian study identified story telling as an important part of this family involvement, building on the storytelling theme above. While this study was descriptive rather than an intervention, the study sample size was large with over 3500 children which adds weight to its findings. The association between paternal childcare involvement especially spending time together, storytelling and taking children for outings and positive psychosocial development. The refugee study found that support from parents at home, as well as mental health service help at school, can improve child adjustment. A therapy treatment models that support traditional beliefs and child-centered parenting practices has been adapted for American Indian and Alaska Native children through focusing on engagement, language cadence and other culturally relevant factors [28]. An evaluation of the effects of this adaptation would provide important learnings for future program development. The cost effectiveness of a family-centered program for low socioeconomic and ethnically diverse US children that improves academic, behavioral and health outcomes has also been demonstrated [29].

Our third theme was that culturally adapted CBT can reduce trauma among adolescents, based on two studies, both with American Indian children [16, 17]. While we did not aim to examine specific therapies in this review, these findings indicate that culturally- adapted CBT may be a potential program element. The inclusion of CBT in programs for Indigenous youth is further supported by findings from an adaptation of an established CBT program for American Indian
adolescents [30] and evaluation [31] that found increases in cultural identity, self-esteem, positive coping strategies, quality of life, and social adjustment. However it may not be suitable for young children who are the focus of this program development and CBT’s focus on trauma may also mean that is less relevant for overcoming general developmental delays.

Our final theme was that rewards-based teaching can improve child attention in an educational setting, based on one study of African American children [20]. The 15 year follow up period in this study and its large sample size of over 1000 children gives strength to its findings that a classroom behavior game, GBG [23], reduces long-term incarceration rates and drug-seeking behavior [20]. The GBG is a long-established type of interdependent group-oriented contingency management procedure that has been used in classrooms and other settings, utilizing team competition and peer influence combined with reinforcement procedures. It has been found to be popular, easy-to-use, time-efficient, and widely applicable and versatile [23] and a current trial of an adaptation and enhancement of the GBT in First Nations communities in Canada will give important future understandings of its impact [32].

These review findings support the inclusion of each of these four themes in the design and development of the Indigenous community program to improve cognitive, emotional and developmental problems among young children, although CBT may not be relevant for this age group. We also recommend future high-quality intervention trials and the sharing of their findings to inform future policy and practice to best overcome developmental delay among young children, particularly children from priority population groups such as Indigenous people.

Strengths of our study include the broad, focus of the review and strong community co-design process with community involvement throughout the review. It identifies practical solutions to inform the development of a community-initiated intervention to overcome a community identified issue, developmental delay and subsequent low educational outcomes. Limitations of this review were the small number of participants (≤5) in three of the seven studies and the heterogeneity of study design that did not allow for adequate comparisons across studies. None the less, the thematic findings provide rich contextual information and were considered by community representatives to give useful learnings to inform the development of a locally designed community intervention.

5. Conclusions

There is limited published research focused on culturally sensitive interventions for children with cognitive, emotional and developmental delays. The four themes from seven relevant studies identified in this review provide useful components for a future program for Indigenous young children to improve educational outcomes. The strong co-design element throughout this review means the findings are highly relevant to the Indigenous community and will inform the program design, development and evaluation.

Supplementary Materials: Table S1: Search strategy, Table S2: CASP Assessment

Author Contributions: Conceptualization, B.R and K.G.; methodology, T.L.J., B.R., K.G.; analysis, D.M., F.T., E.E., M.S.; writing—original draft preparation, R.M.; T.L.J.; writing—review and editing, D.M., F.T., I.B.H, M.S.; All authors have read and agreed to the published version of the manuscript.

Funding: This research received no external funding.

Acknowledgments: We would like to acknowledge the community stakeholder partners who contributed their time and expertise to this review.

Conflicts of Interest: The authors declare no conflict of interest.
References


