International Journal of Osteopathic Medicine 57 (2025) 100763

Contents lists available at ScienceDirect

International Journal of Osteopathic Medicine

journal homepage: www.elsevier.com/locate/ijosm

Potential impact of an osteopathic intervention for internationally adopted children with fetal alcohol spectrum disorder (FASD); a prospective case series

Ramon Cases-Solé [a,b,c], David Varillas-Delgado [b,d,*], Marta Astals-Vizcaino [e], Victoria Aldecoa-Bilbao [e], Óscar García-Algar [b,c,e]

[a] Centre Osteopatia La Seu, La Seu D'Urgell, Lleida, Spain
[b] Grup de Recerca en Osteopatia Pediàtrica (GROP), Barcelona, Spain
[c] Department of Surgery and Medical-Surgical Specialties, University of Barcelona, Barcelona, Spain
[d] Faculty of Health Sciences, Universidad Francisco de Vitoria, Pozuelo de Alarcón, Madrid, Spain
[e] Department of Neonatology, Hospital Clínic-Maternitat, ICGON, BCNatal, Barcelona, Spain

ARTICLE INFO

Keywords:
Fetal alcohol spectrum disorders (FASD)
Osteopathic manipulative treatment (OMT)
Health-related quality of life (HRQoL)
Neurocognitive disorders

ABSTRACT

Objective: To explore the potential impact of Osteopathic Manipulative Treatment (OMT) on Health-Related Quality of Life (HRQoL) on internationally adopted children with fetal alcohol spectrum disorder (FASD).
Methods: Twelve children with FASD adopted from Eastern European countries were recruited in this prospective observational study. HRQoL was collected using the Kidscreen-52 questionnaire. The participants followed a six-month intervention plan that included 4 bi-weekly OMT sessions and 4 monthly OMT sessions.
Results: Significant differences were detected in the pre-post intervention values ($p = 0.001$), interaction by group ($p = 0.048$), in the results of four dimensions (Moods & Emotions, $p = 0.008$, Self-Perception, $p = 0.016$, School Environment, $p = 0.008$ and Social Acceptance, $p = 0.017$) and in the overall assessment for parents' version ($p = 0.002$). A mild adverse effect (pain/discomfort) was reported by one participant (8.3 % of the total sample) after one session.
Conclusion: This study shows that a six-month OMT intervention is a feasible personalized intervention for children and adolescents with FASD coming from international adoption. The study suggests that measuring instruments can detect changes in HRQoL over time, pointing out the need to develop a specific HRQoL assessment instrument for children with FASD to be applied in future studies. Even though promising significant changes were observed, these were most likely not only due to OMT, and further studies are required before assuming these could be due to osteopathic care.

Implications for practice

- A six-month OMT intervention may have beneficial effects on the HRQoL of internationally adopted children and adolescents with FASD, as observed primarily in the parent-reported version of the questionnaire
- OMT appears to be a safe adjuvant therapeutic approach for children with FASD in the improvement of the HRQoL
- OMT is a feasible personalized intervention for FASDs.

1. Introduction

Fetal Alcohol Spectrum Disorder (FASD) and its most obvious presentation, Fetal Alcohol Syndrome (FAS) is the result of alcohol consumption during pregnancy and, therefore, of Prenatal Alcohol Exposure (PAE) of the embryo or fetus. PAE can cause physical, mental, cognitive, behavioral, and learning disabilities, with repercussions in the affected person's adult life, in the family environment and in society [1,2]. Individuals with FAS exhibit the FAS facial phenotype, impaired growth, and cognitive and behavioral abnormalities. Individuals with partial FAS (pFAS) exhibit FAS without growth deficiency, or exhibit FAS with most, but not all, of the facial features [1]. A global prevalence of FASD

* Corresponding author. Ctra. Pozuelo-Majadahonda Km 1,800, 28223, Madrid, Pozuelo de Alarcón, Spain.
E-mail address: david.varillas@ufv.es (D. Varillas-Delgado).

https://doi.org/10.1016/j.ijosm.2025.100763
Received 8 October 2023; Received in revised form 9 January 2025; Accepted 30 April 2025
Available online 14 May 2025

among children and young people in the general population has been estimated at 7.7 per 1000 inhabitants [3]. There is currently no treatment that reverses the effect of PAE, so treatments are aimed at improving the secondary disabilities [4,5]. Only abstinence from alcohol consumption during pregnancy can prevent FASD [6]. At the same time, as a result of increased morbidity and the lack of effective therapeutic interventions, FASD is currently a complex international public health problem [7].

On the other hand, there is evidence that those affected by FASD live with a high tolerance to pain and with somatic problems [8]. Significantly lower levels of Health-Related Quality of Life (HRQoL) were found in children [9] and young adults with FASD [10].

Moreover, after the United States, Spain is the country with the highest rate of children adopted from Eastern Europe, with Catalonia being the region with the largest population - 5120 adoptions from Russia and Ukraine during the period from 1998 to 2015. It has been calculated that 50 % of children adopted from Eastern Europe present FASD in some of its types [11].

The experiences lived prior to adoption are a predictive factor of later behavior. Exposure to Early Life Stress (ELS) increases the risk of psychiatric disorders throughout life [12]. Although little is still known about their relationship, it is considered that ELS would cause a cumulative effect on children born with FASD [12,13]. In this way, worse HRQoL indices have been observed in a sample of young adults with FASD coming from international adoption [10]. The application of individualized and adapted therapeutic interventions could improve these factors and, consequently, their HRQoL levels [9].

Research to date indicates that Osteopathic Manipulative Treatment (OMT) has an anti-inflammatory effect [14] and can increase parasympathetic nervous system function [15]. Thus, in recent years the impact of osteopathic treatment on psychosocial, psychological and quality of life factors has been evaluated [16]. Post-OMT improvements were observed in neurocognitive symptoms associated with FASD, as well as positive effects on learning processes and child neurological development [17].

Thus, we hypothesized that a planned OMT intervention aimed at correcting somatic dysfunctions (SDs) detected through individualized osteopathic treatment could improve HRQoL of internationally adopted children with FASD, as assessed by the Kidscreen-52 questionnaire. Kidscreen-52 includes 2 versions: a self-administered version for children and adolescents, as well as a version for parents or caregivers [18]. Therefore, the main objective of this study was to evaluate the feasibility of a six-month OMT intervention performed by an osteopath qualified in pediatric osteopathy. This would be assessed through drop-out rates, adverse events, and the time needed for the interventions; and the potential impact on the HRQoL variable assessed through the self-administered and parental versions, to evaluate on what components of the Kidscreen-52 changes are observed to focus future outcomes on these components. The secondary objectives were to evaluate the application of the Kidscreen-52 questionnaire in the measurement of HRQoL before and after an OMT intervention in children and adolescents with FASD and to evaluate the concordance of the results between the self-administered versions and that of the parents. The utility of assessing concordance between versions for children and parents is based on the low levels of HRQoL reported in previous studies with children with FASD, combined with the potential presence of assessment biases in this variable by external observers. There is evidence that there may be significant differences between parent-reported and self-reported versions regarding parents' perceptions, with parents potentially overestimating or underestimating levels of HRQoL [19].

2. Materials and methods

2.1. Study design

A prospective case series study was assessed.

2.2. Participants

Children and adolescents adopted from Eastern European countries with FAS and pFAS between the ages of 8 and 16, supervised by the GRIE (Grup de Recerca Infància i Entorn), Hospital Clínic-Maternitat (Barcelona), were recruited between January 2 and March 1, 2018. During the recruitment period, the GRIE evaluated potential candidates, preselecting a total of 31 cases. Cases in which the medication and/or base neuropsychological treatment were modified during the study period were excluded, as were cases that had received osteopathic treatment during the 6 months prior to the start of the intervention. Fifteen were diagnosed with FAS, and 9 with pFAS. Seven were not diagnosed with FAS or pFAS. Families were informed about the study's characteristics, and a Participant Information Sheet was provided. Interested families could choose to participate, read, and sign an Informed Consent Form for participants. Some families read the information, asked questions to the research team, and requested a few days to consider their participation. Ultimately, a total of 12 participants diagnosed with FASD - 7 with FAS and 5 with pFAS - agreed to participate and were selected. All parents of children and adolescents' participants signed the Informed Consent Form before participation in the study. The families that chose not to participate cited personal reasons. The protocol of the study was approved by the Research Ethics Committee "Parc de Salut MAR" (Barcelona, Spain) (IRB 2016/7052/I), carried out according to the guidelines of the Declaration of Helsinki for Human Research 1964 (last modification in 2013).

2.3. Interventions and planning

All the children and adolescents (n = 12) followed a six-month OMT intervention plan that included 4 bi-weekly OMT sessions and 4 monthly OMT sessions. At the same time, all the children continued with their psychosocial and pharmacological treatment. Therefore, the study sample received the OMT intervention and the base neuropsychiatric treatment.

2.4. Implementation

In the first OMT intervention, a protocolized anamnesis and osteopathic physical examination was performed, based on the SOAP anamnesis and examination forms - Subjective, Objective, Assessment, Plan, - for each participant [20,21]. These procedures were performed in each of the sessions with the aim of increasing the safety of the intervention. Obtaining relevant clinical information allowed us to rule out possible warning signs or contraindications to the application of OMT.

The physical examination performed in each intervention was performed using procedures adapted to the clinical characteristics of each participant. Once the exploration phase was completed, the individualized application of the OMT was carried out with the aim of correcting the detected SDs.

Using OMT techniques, the SDs detected throughout the body were corrected one by one. The following techniques were used: Balanced Ligamentous Tension; Balanced Membranous Tension/Osteopathy in the Cranial Field, Facilitated Positional Release, and Myofascial Release [22]. In each session, an osteopathic physical examination and an OMT intervention were performed to evaluate and correct the SDs. A time of 1 h was assigned for the first session and 50 min for the following seven.

The evaluation by means of the Kidscreen-52 questionnaire lasted between 15 and 40 min. In cases where the clinical presentation related to intellectual disability required qualified professionals were required to assist in the completion of the questionnaires. The members of the research team in charge of the application of the OMT intervention were blinded to the evaluation procedures of the participants, that is: the osteopath who carried out the OMT intervention was blinded to the evaluation of the HRQoL variable through the Kidscreen-52 questionnaire. The information was stored in sealed envelopes at the

headquarters of the Hospital Clínic-Maternitat.

The OMT interventions were performed by a pediatric osteopath with a master's degree in Osteopathy - following the recommendations of the European Standard UNE-EN 16686 (16686:2015) - and a post-graduate specialization in Pediatric Osteopathy. A qualified psychologist and a resident physician administered pre and post OMT intervention questionnaires to all participants at the start and end of the intervention. The specific and general recommendations of each questionnaire were followed.

2.5. Study variables and measurement instruments

Adverse events were documented per session through a question asking the patient and the parents generically for any adverse events observed. Drop-out rates were evaluated through the measure of patients that complete the OMT intervention plan. The HRQoL variable was evaluated using the Kidscreen-52 questionnaire [18]. The Kidscreen-52 is a HRQoL measurement instrument for children and adolescents, ranging from 8 to 18 years of age. It contains the self-administered version for children and adolescents, and the version for parents or caregivers. It is a generic, multidimensional quality of life instrument that allows information on the physical, emotional, cognitive, and social health profile to be obtained. The Kidscreen-52 questionnaire is validated for healthy children and adolescents and those with chronic diseases and has 52 items. The 10 dimensions that make up the questionnaire have been extracted from the Kidscreen-52 Manual; 1. Physical Well-being; 2. Psychological Well-being; 3. Moods & Emotions; 4. Self-Perception; 5. Autonomy; 6. Parent Relations & Home Life; 7. Financial Resources; 8. Social Support & Peers; 9. School Environment and; 10. Social Acceptance (Bullying) [23]. A 5-point Likert Scale was used to record each item in the questionnaire on a scale from 1 to 5, where 1 represents 'never' and 5 represents 'always' or 'a lot'. Therefore, the total score for the Kidscreen-52 questionnaire, which consists of 52 items, was transformed to a standardized range from 1 to 100. As the total score increases, perceived HRQoL is considered higher. Regarding reliability, the Kidscreen-52 questionnaire has been shown to be reliable across various populations and contexts, although studies may vary in specific results. The minimal clinically important difference for the Kidscreen-52 is unavailable because it has not been definitively established or universally agreed upon [24]. During the anamnesis details of the neuropsychiatric base treatment and family context (known co-founders) were documented. Reliability: KIDSCREEN-52: Internal consistency values (Cronbach's Alpha) range satisfactorily between 0.76 (Social Acceptance) and 0.89 (Financial Support) for the different dimensions for the self-report version, test-retest reliability at a 2-week interval varies between 0.56 and 0.77. Item intraclass correlation (ICC) between self-reported scores and scores from parents filling out the KIDSCREEN-52 proxy-version ranging from 0.45 (Moods & Emotions) and 0.62 (Physical Wellbeing, School Environment) [24].

2.6. Statistical analysis

All statistical analysis was carried out using SPSS (Statistical Package for the Social Sciences) v.21.0 for Windows (IBM Corp. Released 2012. IBM SPSS Statistics for Windows, Version 21.0. Armonk, N: IBM Corp.). Categorical variables were evaluated through frequencies and percentages and quantitative variables with means and standard deviations, which included maximum and minimum values (range) and medians and interquartile ranges. The Shapiro-Wilk test was used to check the normality of all variables. Since all variables were non-normally distributed, the Wilcoxon test was applied to compare the pre-post results of each dimension and Mann-Whitney *U* test was used to compare pre and post periods in each dimension between groups. When comparing scores between children and parents, if one version (children or parent) was missing, the corresponding comparison for that individual was excluded. However, overall group-level analyses were performed using available data, ensuring sufficient representation for statistical validity. This strategy was chosen to balance retaining as much data as possible while minimizing bias introduced by imputation or exclusion of incomplete cases. Inconsistencies in the Kidscreen-52 responses, defined as repetitive or incoherent answers within at least one dimension, were noted but not excluded from the analysis to preserve the dataset's completeness. These inconsistencies were assumed to reflect cognitive or attentional challenges related to the participants' condition.

Two-sided p values < 0.05 were considered statistically significant.

3. Results

Twelve participants (n = 12) diagnosed with FASD were included in this study, 7 with FAS and 5 with pFAS. The gender ratio (male:female) was 8:4.

a. Safety and adverse events

No serious adverse effects were reported in any of the interventions. A mild adverse effect (pain/discomfort) was reported by one participant (8.3 % of the total sample) after one session (0.96 % of the total sessions).

b. Retention rate

No participant left the study before its completion.

c. Changes in parent and children's questionnaires

The change over time in the results of parent and children's Kidscreen 52-questionnaire showed differences in the parent version between pre- and post-questionnaire (48.7 (44.3–52.3) vs. 53.9 (50.2–55.6) respectively; p = 0.023), not showed in children version between pre- and post-questionnaire (52.0 (44.4–53.7) vs. 54.1 46.1–56.2) respectively; p = 0.353) as depicted in Fig. 1.

d. Changes in sub-scales

In the analysis by dimensions, statistically significant differences were observed in the results pre- and post-questionnaire of Moods &

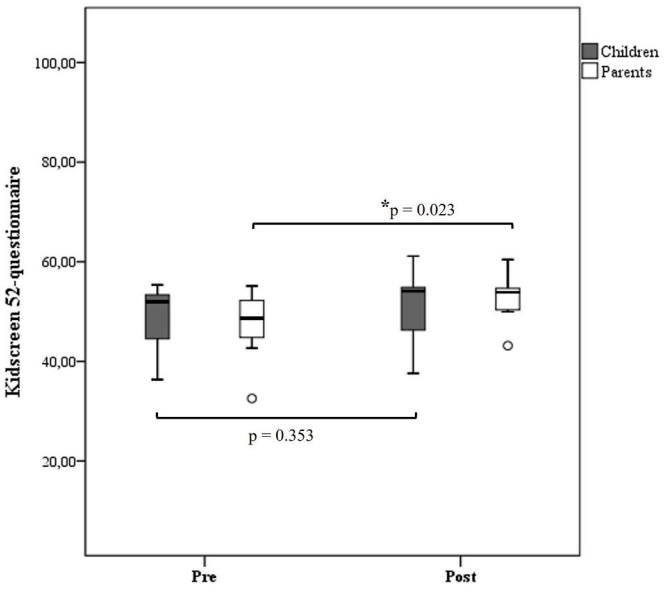

Fig. 1. Temporal development of Kidscreen-52 overall scores questionnaire. Data are given as median and interquartile range.

Emotions (42.0 (36.6–51.3) vs. 51.4 (42.3–70.8) respectively; p = 0.008); Self-Perception (45.4 (42.3–52.3) vs. 52.3 (47.1–60.1) respectively; p = 0.016); School Environment (45.5 (43.8–49.2) vs. 50.9 (45.9–62.5) respectively; p = 0.008); Social Acceptance (30.9 (27.6–42.3) vs. 44.8 (34.6–50.5) respectively; p = 0.017) and in the overall assessment (48.1 (45.6–52.0) vs. 54.9 (50.5–55.2) respectively; p = 0.023) of the parent version (Table 1).

e. Response rate

The response rate of the self-administered version was a total of 96 %, being 93.6 % in the pre-questionnaire and 98.5 % in the post-questionnaire. The response rate in the version for parents was 98.3 %, with 98.5 % completing the pre-questionnaire and 98.1 % the post-questionnaire. Three parents in the pre-questionnaire and four in the post-questionnaire did not respond to the dimension Financial Resources (3 items), citing personal reasons.

f. Variability in measures

Inconsistent or repetitive responses were detected in 66.6 % of the questionnaires for children (85.7 % FAS; 40 % pFAS), in at least one dimension. Inconsistent and repetitive responses refer to participants providing answers in a clearly and unreflectively repetitive manner or without coherence, considering the meaning of each of the Kidscreen-52 questionnaire's dimensions [18].

g. Concordance of responses between parents and children

In relation to the concordance of the results between the groups of children and parents, significant differences were found in the dimension Financial Resources (p = 0.017) in the pre-questionnaires and in Moods & Emotions (p = 0.023), School Environment (p = 0.015) and Social Acceptance (p = 0.017) in the questionnaires after the intervention (Table 2).

h. Duration of interventions

The mean duration (minutes ± standard deviation) of each part of the intervention was as follows (in minutes): anamnesis -1st session- 20.7 min ± 1; exploration: 12.4 min ±1; treatment: 13.3 min ±1.6.

i. Somatic disfunctions and techniques

The mean number of SDs found per participant at the beginning of the intervention was 5.5 ± 1.1 in the first session, decreasing to 2.9 ± 0.7 in the last session; the most prevalent areas being: skull (27.8 %); diaphragm (13.0 %) and T12-L1 (10.0 %). The proportion of techniques used was as follows: balanced ligamentous tension (50.9 %); balanced membrane tension/osteopathy in the cranial field (28.8 %); myofascial release (10.0 %) and facilitated positional release (10.3 %).

4. Discussion

This work, focused on measures of feasibility and safety for a future clinical trial, demonstrates that a 6-month OMT plan, administered by a pediatric-qualified osteopath, is a viable therapeutic approach for children and adolescents with FASD adopted from Eastern European countries, and that changes can be measured over time using the HRQoL Kidscreen-52. The primary disabilities of FASD are those that most directly reflect the underlying central nervous system damage caused by PAE. Secondary disabilities are those that do not present at birth but occur later in life as a result of the primary disabilities associated with FASD [1]. Evidence has been found about the improvement of secondary FASD disabilities through therapeutic interventions [4,5] and its potential positive effect on HRQoL [9]. No serious adverse effects were reported during the study. A mild adverse effect (pain/discomfort) was reported by one participant (8.3 % of the total sample) after one session (0.96 % of the total sessions), lasting less than 24 h. These data are in line with other publications and confirms the theory put forward by different authors on the safety of OMT [25,26]. The HRQoL Kidscreen-52 is able to detect changes over time, particularly in the dimensions of Moods & Emotions, Self-Perception, School Environment, and Social Acceptance, as well as the total score for the parent version. However, it is important to note that the children's version did not demonstrate this ability in the present study. Previous research has also reported discrepancies between the parent and children's versions in terms of score evaluation and sensitivity to change over time, suggesting the need for further exploration of these differences in diverse populations [18,24,27,28].

Although the total response completion rates meet the application objective: self-administered questionnaire 96.0 %, parents 98.3 %, in the self-administered version, inconsistencies or repetitive responses were detected in at least one dimension in 66.6 % of participants. These results call into question the use of questionnaires in this population, especially in cases of FAS, where this proportion rose to 85.7 %, compared to 40.0 % of the participants with pFAS. Although this data was to be expected, since the rates of intellectual disability among the population with FAS are very high, we decided not to exclude any participant from the self-administered evaluation. To avoid possible performance bias as much as possible, professionals with experience assisted in the completion of the questionnaires for the evaluation of children and adolescents with FASD [29–31].

In our opinion, the parent version provides a complementary measure to the child version. According to Varni et al. [32], parent versions should be included as a secondary measure but considered primary when the children or adolescent has a disability that prevents self-reporting. Offering the completion of questionnaires to all children

Table 1
Analysis of the Kidscreen-52 questionnaire dimensions between the pre- and post-measurement for children and parent's groups.

	Children (n = 12)			Parents (n = 12)		
	Pre, median (IQR)	Post, median (IQR)	p value	Pre, median (IQR)	Post, median (IQR)	p value
1. Physical Well-Being	54.0 (42.5–71.0)	57.5 (52.4–73.2)	0.161	55.9 (52.7–59.4)	57.6 (52.7–71.2)	0.373
2. Psychological Well-Being	53.1 (41.9–59.8)	51.9 (45.1–68.5)	0.799	52.1 (44.1–58.2)	50.5 (45.9–67.7)	0.167
3. Moods & Emotions	40.6 (37.2–54.4)	42.5 (37.8–50.3)	0.722	42.0 (36.6–51.3)	51.4 (42.3–70.8)	**0.008**
4. Self-Perception	46.1 (43.2–69.8)	53.8 (44.3–69.8)	0.176	45.4 (42.3–52.3)	52.3 (47.1–60.1)	**0.016**
5. Autonomy	46.8 (43.6–68.7)	54,7 (41,7–66,7)	0.859	48.2 (40.0–50.9)	48.2 (41.4–60.0)	0.333
6. Parent relations & Home Life	54.6 (49.5–65.9)	47.6 (43.3–63.1)	0.260	50.7 (46.9–61.4)	53.6 (47.5–61.4)	0.534
7. Financial Resources	44.2 (38.6–50.8)	49.3 (30.8–62.9)	0.484	51.9 (46.5–57.3)	64.2 (53.7–65.0)	0.080
8. Social Support & Peers	50.2 (43.6–58.1)	52.4 (46.4–61.5)	0.114	53.0 (36.2–61.2)	49.6 (42.4–63.1)	0.646
9. School Environment	54.2 (45.3–61.9)	67.8 (47.3–73.8)	0.074	45.5 (43.8–49.2)	50.9 (45.9–62.5)	**0.008**
10. Social Acceptance	35.4 (22.4–48.1)	31.1 (25.0–56.1)	0.507	30.9 (27.6–42.3)	44.8 (34.6–50.5)	**0.017**
Overall assessment	52.2 (45.2–54.1)	54.3 (46.8–55.3)	0.353	48.1 (45.6–52.0)	54.9 (50.5–55.2)	**0.023**

IQR; interquartile range.

Table 2
Analysis of the Kidscreen-52 questionnaire concordance dimensions between the pre- and post-measurement for children and parent's groups.

	Pre			Post		
	Children, median (IQR)	Parents, median (IQR)	p value	Children, median (IQR)	Parents, median (IQR)	p value
1. Physical Well-Being	54.0 (42.5–71.0)	55.9 (52.7–59.4)	0.308	57.5 (52.4–73.2)	57.6 (52.7–71.2)	0.209
2. Psychological Well-Being	53.1 (41.9–59.8)	52.1 (44.1–58.2)	0.480	51.9 (45.1–68.5)	50.5 (45.9–67.7)	0.272
3. Moods & Emotions	40.6 (37.2–54.4)	42.0 (36.6–51.3)	0.237	42.5 (37.8–50.3)	51.5 (42.3–70.8)	**0.023**
4. Self-Perception	46.1 (43.2–69.8)	45.4 (42.3–52.3)	0.859	53.8 (44.3–69.8)	52.3 (47.1–60.1)	0.408
5. Autonomy	46.8 (43.6–68.7)	48.2 (40.0–50.9)	0.929	54.7 (41.7–66.7)	48.2 (41.4–60.0)	0.308
6. Parent relations & Home Life	54.6 (49.5–65.9)	50.7 (46.9–61.4)	0.667	47.6 (43.3–63.1)	53.6 (47.5–61.4)	0.066
7. Financial Resources	44.2 (38.6–50.8	51.9 (46.5–57.3)	**0.017**	49.3 (30.8–62.9)	64.2 (53.7–65.0)	0.090
8. Social Support & Peers	50.2 (43.6–58.1)	53.0 (36.2–61.2)	0.657	52.4 (46.4–61.5)	49.6 (42.4–63.1)	0.875
9. School Environment	54.2 (45.3–61.9)	45.5 (43.8–49.2)	0.182	67.8 (47.3–73.8)	50.9 (45.9–62.5)	**0.015**
10. Social Acceptance	35.4 (22.4–48.1)	30.9 (27.6–42.3)	0.929	31.1 (25.0–56.1)	44.8 (34.6–50.5)	**0.012**
Overall assessment	49.5 (45.9–57.8)	48.1 (45.2–50.0)	0.182	52.3 (45.1–57.1)	50.9 (47.2–61.6)	0.071

IQR; interquartile range.

and adolescents under professional care proved to be a successful approach. The decision to administer the questionnaires to all children in the sample, regardless their FAS or pFAS diagnosis and whether their parents completed the parent version, led to a higher response rate for the child version after the intervention (93.6 % pre-intervention vs. 98.5 % post-intervention) compared to the parent version (98.5 % pre-intervention vs. 98.1 % post-intervention). A possible recall bias in subsequent results should be considered. The lower proportion of participants with inconsistent responses in cases of pFAS (40.0 %) allows a more favorable assessment of the use of this tool in less severe cases, although this argument requires additional studies.

The characteristics of this study do not allow us to draw more conclusions in this regard, and a more in-depth analysis would be recommended, particularly regarding the extent to which the responses of children and adolescents with neurocognitive disorders are useful and how to integrate the results of the parent and self-administered versions [28]. Additionally, it is important to question the validity of the questionnaire if such discrepancies exist between the child and parent versions.

Moreover, we selected a generic HRQoL assessment tool due to its widespread use in children with neurodevelopmental disabilities [19] and due to the absence of a specific HRQoL assessment tool in children and adolescents with FASD. In our opinion, the use of a specific HRQoL assessment tool that assesses specific aspects that may be important for children and adolescents with disabilities - for example, pain and discomfort [29] should be evaluated in future studies with children and adolescents with FASD. Thus, we point out the need to develop a specific HRQoL assessment instrument for children and adolescents with FASD.

Regarding the concordance between the versions of parents and children and adolescents, we can affirm that there is correspondence between the results of both versions, except for the Financial Resources dimension in the pre-intervention questionnaires, as well as the Moods & Emotions, School Environment, and Social Acceptance dimensions in the post-intervention questionnaires. The total scores for the pre- and post-intervention questionnaires present favorable correlation results.

On the other hand, the positive results obtained should be taken with caution, since it is a study without a control group that allows comparison with standard conventional treatment, or a third therapeutic intervention or placebo intervention. Due to the characteristics of the sample, we could not select and evaluate a similar group to include it as a control group, not exposed to the intervention. We must take into account potential candidates for explaining changes over time other than those due to OMT, such as regression towards the mean, natural and spontaneous remission, social desirability, lack of blinding, and non-specific effects.

Despite the strength of the study, we present several limitations of the study, including lack of blinding for measures taken before and after treatments, lack of standardization and description of the intervention, a small sample size which has limited the use of statistical normality tests to interpret the results and the magnitude of the effect, the limitation in generalizability of results given treatments were provided by a single practitioner, an absence of control group and the short duration of the follow-up period.

TART parameters are used to diagnose the SDs. The techniques applied were directed to correct the SDs diagnosed during the physical exploration. The characteristics of the FASD population, with a high rate of somatic problems and traumatic incidents could lead to a higher rate of SDs and a higher risk of their relapse [8]. The impact of the OMT techniques in the correction of the SDs in internationally adopted children and adolescents with FASD, and their evolution in time, deserves to be studied in future studies.

5. Conclusion

This study shows that a 6-month OMT intervention is a feasible personalized intervention for FASD children and adolescents with FASD coming from international adoption. The study suggests that measuring instruments can detect changes in HRQoL over time. The findings highlight significant limitations in the children's version of the Kidscreen-52, including its reduced sensitivity to change in small sample sizes, inconsistencies in responses, and lack of a minimal clinically important difference. These challenges call into question its use as a primary outcome measure for children with FASD, pointing out the importance of the version for parents as a primary outcome, especially in cases of FAS. Future studies should focus on developing and validating HRQoL instruments tailored specifically to FASD population, ensuring better accuracy and consistency across assessments.

CRediT authorship contribution statement

Ramon Cases-Solé: Writing – review & editing, Writing – original draft, Visualization, Validation, Supervision, Resources, Project administration, Methodology, Investigation, Formal analysis, Data curation, Conceptualization. **David Varillas-Delgado:** Writing – review & editing, Writing – original draft, Visualization, Software, Methodology, Investigation, Formal analysis, Data curation. **Marta Astals-Vizcaino:** Writing – review & editing, Visualization, Methodology, Investigation, Data curation. **Victoria Aldecoa-Bilbao:** Writing – review & editing, Methodology, Investigation, Data curation. **Óscar García-Algar:** Writing – review & editing, Writing – original draft, Visualization, Validation, Supervision, Methodology, Investigation, Data curation, Conceptualization.

Ethical approval details

All parents of children and adolescents' participants signed a written inform consent before participation in the study. The protocol of the study was approved by the Research Ethics Committee "Parc de Salut

MAR" (Barcelona, Spain) approved the study protocol (IRB 2016/7052/I), carried out according to the guidelines of the Declaration of Helsinki for Human Research 1964 (last modification in 2013).

Funding sources

None.

Declaration of competing interest

None.

Acknowledgements

The authors wish to thank the participants for their invaluable contribution to the study.

References

[1] Hoyme HE, Kalberg WO, Elliott AJ, Blankenship J, Buckley D, Marais AS, et al. Updated clinical guidelines for diagnosing fetal alcohol spectrum disorders. Pediatrics 2016 Aug;138(2).
[2] McQuire C, Paranjothy S, Hurt L, Mann M, Farewell D, Kemp A. Objective measures of prenatal alcohol exposure: a systematic review. Pediatrics 2016 Sep;138(3).
[3] Lange S, Probst C, Gmel G, Rehm J, Burd L, Popova S. Global prevalence of fetal alcohol spectrum disorder among children and youth: a systematic review and meta-analysis. JAMA Pediatr 2017 Oct 1;171(10):948–56.
[4] Ordenewitz LK, Weinmann T, Schlüter JA, Moder JE, Jung J, Kerber K, et al. Evidence-based interventions for children and adolescents with fetal alcohol spectrum disorders - a systematic review. Eur J Paediatr Neurol 2021 Jul;33:50–60.
[5] Hilly C, Wilson PH, Lucas B, McGuckian TB, Swanton R, Froude EH. Effectiveness of interventions for school-aged-children and adolescents with fetal alcohol spectrum disorder: a systematic review and meta-analysis. Disabil Rehabil 2023 May;9:1–26.
[6] Pichini S, Busardò FP, Garcia-Algar O. Only total abstinence from alcohol consumption during pregnancy guarantees absolute absence of any deleterious effect in the fetus and child. J Stud Alcohol Drugs 2020 Mar;81(2):220–1.
[7] Mattson SN, Bernes GA, Doyle LR. Fetal alcohol spectrum disorders: a review of the neurobehavioral deficits associated with prenatal alcohol exposure. Alcohol Clin Exp Res 2019 Jun;43(6):1046–62.
[8] Domeij H, Fahlström G, Bertilsson G, Hultcrantz M, Munthe-Kaas H, Gordh CN, et al. Experiences of living with fetal alcohol spectrum disorders: a systematic review and synthesis of qualitative data. Dev Med Child Neurol 2018 Aug;60(8):741–52.
[9] Stade BC, Stevens B, Ungar WJ, Beyene J, Koren G. Health-related quality of life of Canadian children and youth prenatally exposed to alcohol. Health Qual Life Outcome 2006 Oct 13;4:81.
[10] Gyllencreutz E, Aring E, Landgren V, Landgren M, Grönlund MA. Visual perception problems and quality of life in young adults with foetal alcohol spectrum disorders. Acta Ophthalmol 2022 Feb;100(1):e115–21.
[11] Colom J, Segura-García L, Bastons-Compta A, Astals M, Andreu-Fernandez V, Barcons N, et al. Prevalence of fetal alcohol spectrum disorders (FASD) among children adopted from eastern European countries: Russia and Ukraine. Int J Environ Res Publ Health 2021 Feb 3;18(4).
[12] Alberry B, Laufer BI, Chater-Diehl E, Singh SM. Epigenetic impacts of early life stress in fetal alcohol spectrum disorders shape the neurodevelopmental continuum. Front Mol Neurosci 2021;14:671891.
[13] McLachlan K, Flannigan K, Temple V, Unsworth K, Cook JL. Difficulties in daily living experienced by adolescents, transition-aged youth, and adults with fetal alcohol spectrum disorder. Alcohol Clin Exp Res 2020 Aug;44(8):1609–24.
[14] Licciardone JC, Kearns CM, Hodge LM, Bergamini MV. Associations of cytokine concentrations with key osteopathic lesions and clinical outcomes in patients with nonspecific chronic low back pain: results from the OSTEOPATHIC Trial. J Am Osteopath Assoc 2012 Sep;112(9):596–605.
[15] Ruffini N, D'Alessandro G, Mariani N, Pollastrelli A, Cardinali L, Cerritelli F. Variations of high frequency parameter of heart rate variability following osteopathic manipulative treatment in healthy subjects compared to control group and sham therapy: randomized controlled trial. Front Neurosci 2015;9:272.
[16] Saracutu M, Rance J, Davies H, Edwards DJ. The effects of osteopathic treatment on psychosocial factors in people with persistent pain: a systematic review. Int J Osteopath Med 2018;27:23–33. 2018/03/01/.
[17] Frymann VM, Carney RE, Springall P. Effect of osteopathic medical management on neurologic development in children. J Am Osteopath Assoc 1992 Jun;92(6):729–44.
[18] Ravens-Sieberer U, Gosch A, Rajmil L, Erhart M, Bruil J, Duer W, et al. KIDSCREEN-52 quality-of-life measure for children and adolescents. Expert Rev Pharmacoecon Outcomes Res 2005 Jun;5(3):353–64.
[19] Lemmon ME, Huffstetler HE, Reeve BB. Measuring health-related quality of life in pediatric neurology. J Child Neurol 2020 Sep;35(10):681–9.
[20] Sleszynski SL, Glonek T. Outpatient Osteopathic SOAP Note Form: preliminary results in osteopathic outcomes-based research. J Am Osteopath Assoc 2005 Apr;105(4):181–205.
[21] Sleszynski SL, Glonek T, Kuchera WA. Standardized medical record: a new outpatient osteopathic SOAP note form: validation of a standardized office form against physician's progress notes. J Am Osteopath Assoc 1999 Oct;99(10):516–29.
[22] Degenhardt B, van Dun PLS, Jacobson E, Fritz S, Mettler P, Kettner N, et al. Profession-based manual therapy nomenclature: exploring history, limitations, and opportunities. J Man Manip Ther 2024 Feb;32(1):96–110.
[23] Befus EG, Helseth S, Mølland E, Westergren T, Fegran L, Haraldstad K. Use of KIDSCREEN health-related quality of life instruments in the general population of children and adolescents: a scoping review. Health Qual Life Outcome 2023 Jan 20;21(1):6.
[24] Ravens-Sieberer U, Gosch A, Rajmil L, Erhart M, Bruil J, Power M, et al. The KIDSCREEN-52 quality of life measure for children and adolescents: psychometric results from a cross-cultural survey in 13 European countries. Value Health 2008 Jul-Aug;11(4):645–58.
[25] Hayes NM, Bezilla TA. Incidence of iatrogenesis associated with osteopathic manipulative treatment of pediatric patients. J Am Osteopath Assoc 2006 Oct;106(10):605–8.
[26] Degenhardt BF, Johnson JC, Brooks WJ, Norman L. Characterizing adverse events reported immediately after osteopathic manipulative treatment. J Am Osteopath Assoc 2018 Mar 1;118(3):141–9.
[27] Haraldstad K, Christophersen KA, Eide H, Nativg GK, Helseth S. Health related quality of life in children and adolescents: reliability and validity of the Norwegian version of KIDSCREEN-52 questionnaire, a cross sectional study. Int J Nurs Stud 2011 May;48(5):573–81.
[28] Robitail S, Siméoni MC, Ravens-Sieberer U, Bruil J, Auquie P. Children proxies' quality-of-life agreement depended on the country using the European KIDSCREEN-52 questionnaire. J Clin Epidemiol 2007 May;60(5):469–78.
[29] Eiser C, Morse R. Quality-of-life measures in chronic diseases of childhood. Health Technol Assess 2001;5(4):1–157.
[30] Riley AW. Evidence that school-age children can self-report on their health. Ambul Pediatr 2004 Jul-Aug;4(4 Suppl):371–6.
[31] Young B, Rice H, Dixon-Woods M, Colver AF, Parkinson KN. A qualitative study of the health-related quality of life of disabled children. Dev Med Child Neurol 2007 Sep;49(9):660–5.
[32] Varni JW, Limbers CA, Burwinkle TM. How young can children reliably and validly self-report their health-related quality of life?: an analysis of 8,591 children across age subgroups with the PedsQL 4.0 Generic Core Scales. Health Qual Life Outcome 2007 Jan 3;5:1.