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RESEARCH

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A novel child-centred core palliative care outcome measure for use in clinical practice and research: findings from a multinational validation study

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Abstract

Background Outcome measurement is pivotal to person-centred assessment, quality improvement and research. Children and young people with life-limiting and -threatening illness have high needs and service use, yet there is a lack of evidence for interventions and care models. Efforts to strengthen paediatric palliative care (PPC) services has been hampered by the lack of an appropriate outcome measure.

Objective To determine the validity, reliability, measurement invariance, responsiveness, acceptability, and interpretability properties of the novel Children's Palliative care Outcome Scale (C-POS).

Methods We recruited children (0–17 years) with life-limiting/life threatening conditions and their families in Kenya, Uganda and South Africa. Using C-POS repeated measurement using over four timepoints. We assessed: 1) construct validity (structural properties, discriminant validity, known groups validity, measurement invariance, differential item functioning by country), 2) reliability (internal consistency and test re-test), 3) responsiveness, 4) acceptability (time to complete) and 5) interpretability.

Results We recruited a cohort of 434 children (response rate 94%). Of these, 302 participated in the repeated measures component and 279 (92%) completed four datapoints.

We found evidence for face and content validity as the C-POS items mapped on to themes developed from qualitative interviews, including: pain and other symptoms, pyscho-social well-being, and family wellbeing that matter to children and their families. We confirmed: 1) the two-factor structure (child and family subscales). We also confirmed discriminant and known groups validity, as well as construct equivalence for the child self-report and proxy versions. Controlling for age, we found no differential item functioning by country setting. 2)The sub-scale internal consistency was moderate, given the multi-dimensional nature of the C-POS self and proxy report versions omega scores (0.67 and 0.73, respectively). The test characteristic curve information confirmed the moderate internal subscale consistency scores between 0.3- 0.9 for the proxy version and 0.3–0.5 for the self-report version. Test–retest reliability was acceptable for all items, with weighted kappa range for scores: self-report (0.43–0.57) and proxy version

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(0.35–0.64) and family items (0.51–0.71). 3)Responsiveness was demonstrated, except for the feeding item. 4)Median completion time at the last visit was 10 min for both versions with minimal missing data. 5)The minimum important difference was 3 for the self and proxy report versions on a scale of 0–30 and 4 for the child and family scale on a scale of 0–55.

Conclusions and relevance The C-POS has good psychometric properties. To further improve the measure, we identified items for potential removal, conceptual gaps that should be addressed and domains for which developmental age-appropriate items are needed. C-POS has potential to evaluate and improve person-centred children's palliative care in research and routine clinical practice.

Keywords Paediatric palliative care, Outcome measurement, Children, Africa

Background

Worldwide, there are approximately 21 million children and young people (hereafter "children") aged 0–19 years with life-limiting and life-threatening conditions (LLCs) [1]. There are almost 400 conditions that affect children for which palliative care could be beneficial [2, 3].

Due to medical advances, children are living longer with life-limiting conditions [2, 4]. Children with LLCs face a high burden of multidimensional symptoms and concerns, spanning physical (e.g. pain) psychological (e.g. anxiety), social (e.g. education, isolation) and spiritual (e.g. meaning and uncertainty) [5, 6]. Although referral to specialist paediatric palliative care may reduce hospital admissions [7], the impact on their person-centred outcomes (i.e. their symptoms and concerns) is largely unknown.

Palliative care for adults is effective and cost-effective, reducing unplanned admissions and futile treatments [8–10], while improving quality of life, care quality and survival [11–13]. However, evidence for effectiveness of children's palliative care is limited in part due to a lack of a valid and reliable person-centred outcome measure [14, 15]. Systematic reviews report that there are no valid patient-centred outcome measures (PCOMs) for paediatric palliative care [16–18]. Development of such a measure has repeatedly been identified as a research priority [16, 19, 20].

Person-centredness is holistic healthcare and a core commitment of the World Health Organisation [21–24]. To deliver child-centred care, it is essential to understand what is important to children and their families [25, 26]. The United Nations Convention on the Rights of the Child emphasises the importance of children being involved in matters that affect them [27].

Patient-reported information is central to improving care and quality of life, and evidence demonstrates that children can reliably self-report [26, 28]. However, their voices have not been prioritised in clinical care or research [29]. Person-centred, ideally patient-reported, outcome measures are key to assessment, problem measurement, demonstrating the effectiveness of care, and driving healthcare quality and equity [30]. Minimal evidence has been published on the outcomes of children with life-limiting or threatening conditions [17]. This evidence is affected by the use of methods which majorly use retrospective and proxy methods, and lacking a validated tool to collect outcome data [31, 32]. Progress on documenting children's palliative care outcomes is therefore limited compared to advances in adult patients outcome measurement [33].

The Palliative care Outcome Scale (POS) was developed to measure adult patient-reported outcomes in palliative care [34, 35] and is now used worldwide in multiple versions with widely reported clinical and research use [36–39]. An adult patient African version was developed and validated [40, 41] and has been used widely to measure and improve care quality for adults [39].

The vast majority of children with LLCs (98%) live in low- and middle-income countries [42], where late presentation, fewer curative options, and higher prevalence of infectious diseases lead to the urgent need for evidence of optimal and effective care delivery models [43]. A review of the status of children's palliative care in sub-Saharan Africa identified the need for a validated outcome measure [44]. Consequently, the first outcome measure for children with LLCs was developed and piloted: the APCA African Children's Palliative care Outcome Scale (hereafter referred to as C-POS) [45, 46]. This early version was revised by a multidisciplinary cross-national expert panel. This process integrated findings from the pilot with a new systematic review of the evidence for outcomes meaningful to children with life-limiting and life-threatening illness [6]. Further primary qualitative data for children's needs in sub-Saharan Africa generated to improve face and content validity [25]. We previously reported content validation (mapping C-POS items onto an evidence-based framework from prior evidence), comprehensiveness, comprehensibility, acceptability feasibility, and implementability from qualitative in-depth and cognitive interviews with children, families and health care professionals in three sub-Saharan African countries [47]. Assessment of those properties concluded

that C-POS items capture the core symptoms and concerns that matter to children and their families, and that it is feasible, comprehensible, and acceptable for use in clinical settings.

We present here the results from the full quantitative psychometric validation. This validation study aimed to determine the psychometric properties of a novel child- and family-centred palliative care core outcome measure in sub-Saharan Africa for use in research and clinical practice.

Methods

The objectives were to determine: 1) construct validity (structural properties, discriminant validity, known groups validity, measurement invariance, differential item functioning by country), 2) reliability (internal consistency and test re-test), 3) responsiveness, 4) acceptability (time to complete and item completeness) and 5) interpretability.

Design

We applied the COSMIN taxonomy of best practices in tool development and validation [48, 49] and outcome measure guidance in palliative care [40, 50, 51]. The overview of our study methods is show in Fig. 1; we present methods and findings by phase of study. The overall construct being measured was symptoms and concerns among children facing LLCs and their families.

Setting

Three clinical sites delivering palliative care (a children's HIV outpatient service in Uganda, a teaching and referral hospital-one provincial hospital in Kenya, and a national children's hospital in South Africa) in line with the WHO definition of paediatric palliative care [52] and ability to recruit at least 30 new patients per month to allow for timely study completion.

Inclusion/exclusion criteria

Inclusion: aged between birth and 17 years receiving care for a LLC, with parent or legal guardian present to consent to study participation.

Exclusion: We excluded children who were deemed by their clinician to be too ill or had cognitive impairments of a severity that precluded meaningful participation. Families were excluded if their child's clinician felt the family member was either too unwell or distressed to take part. We defined a family caregiver as a family member who took care of the child for at least 50% of the time.

Recruitment and consent

Caregivers were approached by a member of the clinical team and informed of the study objectives and procedures. All participating children aged 8 years and above gave assent, and adult caregivers gave informed written consent for children to participate.

Data collection

Data collection occurred between February 2012 to November 2012. Study instruments, information and consent forms were forward and back translated from English [53] into local languages: Uganda (Luganda, Runyakore-Rukiga); Kenya (Kikuyu, Luo, Kiswahili); South Africa (Xhosa, Zulu, Pedi Sesotho, South Sotho). This was followed by the reconciliation of the two forward translations through dialogue and consultation with content experts.

Due to varying levels of respondent literacy, the research nurses read the questions aloud in all instances and recorded responses from children or their caregivers. We trained all the research nurses on how to administer the study instruments and gave each nurse a copy of the standard operating procedures for administering the study tools. Patients could score the C-POS using a hand or verbal scale. In our earlier phases of developing the C-POS, children were asked about preferred scales and hand and verbal were most preferred and self-reporting children could interpret hand and verbal scales. Children aged 7 years and above were allowed to respond on their own if possible given evidence shows they can self-report on health and wellbeing [54, 55]. For test–retest, the C-POS was re-administered to a sub-cohort of in-patients whose well-being the clinical team did not expect to change significantly in a period of 24 h.

Measures

The C-POS

The C-POS addresses children's symptoms and concerns, drawing on the "total pain" construct that drives assessment and intervention in palliative care (i.e. physical and pyscho-social, spiritual, practical and emotional concerns, and needs of the family) (Appendix in Fig. 3). Scores use Likert scales from 0 to 5. Questions 1–7 of the C-POS are directed at patients/children; these questions can be asked directly to children (either self-report or proxy) or proxies can be observer informants. The tool is self-reported for children aged 7 years and above [56] where literacy was a problem, staff read aloud the items and responses with respondents choosing the best response options, on their own. Proxies can respond where a child is unable because of age (e.g. the 0–6-year-old) or those with advanced disease, although research with adults demonstrated that family members/caregivers and professionals' ratings may differ from the patient [57]. For assessment, the C-POS was administered four times, and each point the completion time was recorded.

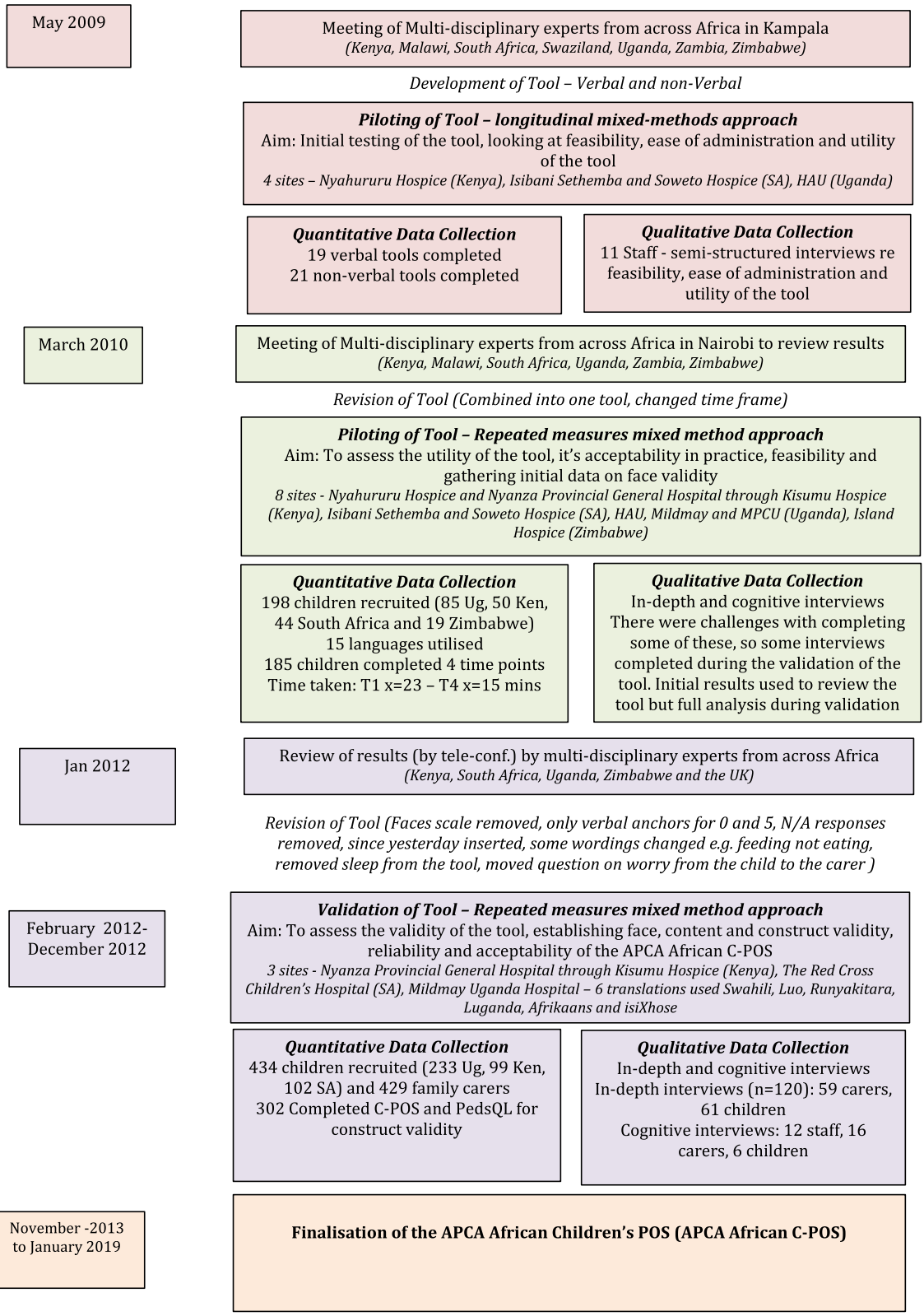

Fig. 1 Process of the development and validation of the APCA African C-POS

Paediatric Quality of Life Inventory (PedsQL)

The PedsQL [58], is a 23 item measure of quality of life designed to measure the core dimensions of health and role (school) functioning. Given that the PedsQL is a generic function-based measure of quality of life, we selected it to test for divergence from C-POS measure of palliative care-related symptoms and concerns in children with life-limiting and life-threatening conditions.

Eastern Cooperative Oncology Group Scale (ECOG)

This single item measure of functional performance [59] ranges from 0 (fully active) to 4 (completely disabled). It is used as a proxy for disease progression and its effect on daily living abilities [59]. In line with guidance, we used ECOG with children aged 5 and above [60].

Socio-demographics

A study-specific questionnaire collected data on: child's age, sex, first language, caregiver's relationship to the child, household size, primary diagnosis, phase of illness, place of care (inpatient/outpatient), and reason for referral to palliative care.

Sample size

Sample size estimation was based on factor analysis and structural equation modelling (SEM) as they required the largest sample. Ideally 10 cases/observations per indicator variable are recommended for factor analysis [61], while sample sizes of 100–150 are recommended for SEM [62]. Based on Monte and Carlo simulations with a power of 80%, p value 0.05, a sample size of > 200 was for robust weighted Least Squares or maximum likelihood for both binary or ordinal data [63, 64]. We therefore deemed a sample size > 200 for the child and proxy versions of the C-POS as adequate for modelling.

Data management

Data were entered into a pre-designed Epidata database and exported to Stata version 15 and Mplus 8.3 for analysis. The C-POS scores were explored for out-of-range scores and missing values, and items (1–4, & 8) were reversed so in all instances a score of "0" represented the least severity and "5" the most severe.

Analysis

Construct validity

We assessed for the following aspects of construct validity: a) structural properties; b) discriminant validity; c) known groups validity; d) measurement invariance; e) differential item functioning by country.

Structural properties Theoretically the C-POS has two sub-scales, i.e., child items outcomes ($n=7$) and family-items ($n=5$). We conducted multi-level confirmatory analysis using the weighted least squares to confirm this factor structure [49]. This approach is recommended where a pre-existing theory exists as confirmatory factor analysis tests a hypothesis and is hence more robust [49]. We compared competing models to identify the model with the best fit using the flowing model fit coefficients; i)the chi-square and the associated degrees of freedom and the associated P values, ii) the comparative fit index (CFI) > 0.9 is recommended, iii) the Tucker-Lewis Index (TLI)—≥ 0.9–0.95 and iv) the RMSEA -recommended cut off is ≤ 0.05 but sometimes < 0.08 acceptable [65].

Discriminant validity We hypothesised that we would find a low-moderate correlation between C-POS and PedsQL (i.e. < 0.6). The following were calculated: PedsQL psychosocial subscale scores vs C-POS psychosocial items (felt happy + felt like playing), and PedsQL physical health subscale vs the C-POS physical items (pain + other symptoms items). We used the Spearman Rank Correlation (Spearman's rho) to assess for the strength of correlation between the C-POS and PedsQL scores [66] (0 < 0.3 – low correlation; 0.3–0.5 – moderate correlation; > 0.5—strong correlation) [67].

Known groups validity We defined our known groups based on functional performance scores i.e. the principle that certain specified groups of patients are anticipated to score differently. Dividing the study population into known groups by functional performance status (for children aged 5 and above for whom it was possible to use the ECOG). We hypothesised that children with poor physical function would report more palliative care-related problems. For the self and proxy versions of the C-POS, we used analysis of variances to assess statistical significance of mean differences in the C-POS child item total scores across the levels of functional performance as measured by the ECOG.

Measurement invariance We fitted a configural model to explore the extent to which the C-POS self and proxy versions measure a similar construct, then a metric invariance model to determine the equivalence between the two versions (self and proxy report) at factor loading or structural level and then use of the scale or threshold level. At each stage, the model fit indices were examined to identify problematic items (i.e., those with large residual errors, and the level at which variance occurs). Sensitivity analyses were conducted to the explore the effect of excluding such items or allowing them to vary on the overall model fit.

Differential item functioning by country setting We used differential item functioning analysis to assess for cultural differences in the functionality of the C-POS items across Uganda, Kenya and South Africa. This was achieved using Multiple Indicator, Multiple Cause (MIMIC) modelling controlling for the effect of age. We fit three models: Model 1 self-report version of the C-POS child items; Model 2 Proxy report version of the C-POS child items; Model 3 Family items.

Reliability

Internal consistency As C-POS is a multidimensional measure, we did not assess the internal consistency of the tool as a whole, as results can be misleading [68]. Following confirmation of the hypothesised factor structure, internal consistency for the child and family subscales was assessed using the omega composite reliability coefficient. The latter statistic is robust in case of violation of the unequal factor loadings in a factorial model [69]. For items tapping a single construct, coefficients above 0.7 are acceptable and for multi-dimensional measures low coefficients of up to 0.5 are acceptable [70].

Further analysis was undertaken using item response theory to test the precision of the selected items and to identify areas for improvement along the latent construct continuum. We fitted a partial graded model and examined the internal consistency with additional information on the extent to which the various items contributed to our understanding of the variation in the latent construct. We inspected item and test information functions graphically to reflect how reliably the individual items and the test estimate the construct over the entire scale range. Values can be converted into an estimate of reliability (using this formula reliability ¼ 1 _ [1 /information]) extrapolating from Cronbach's alpha rule of thumb of 0.70 to 0.90 for interpreting reliability; with the test function curve, these values correspond to acceptable item curve information scores of 3.3 to 10 [61].

To identify potential areas for improving internal consistency, we examined the functionality of the items; good items should contribute to our understanding of the variation in the latent construct between -2 and + 2 standard deviations. Items with poor discrimination power (i.e., those with low information curves) could be targeted for removal, replacement or rephrasing.

Test–retest reliability We used the weighted kappa coefficient to assess the level of agreement between the two time point scores. More subjective items will generally show relatively low reliability, and physical outcomes may be more consistent [70, 71]. Lower test–retest coefficients were expected for the child self-report version, as test–retest reliability is affected by child developmental age [72, 73]. Interpretation of coefficients was: less than 0.2 poor agreement; 0.21–0.40 slight; 0.41–0.60 moderate 0.61–0.8 good and 0.81 -1 very high [73]. We adopted a score of 0.3 for lowest acceptable level of agreement.

Responsiveness

Responsiveness is defined as the ability of a measure to detect change [74]. A good instrument should be able to respond to changes in a patient's condition but it is important to note that responsiveness contributes more to our understanding of the variance of the population being assessed and is not a property of an instrument in the strict sense [75]. Using Wilcoxon paired sign rank sum test, we compared differences in paired scores at the following time points; 1 vs 3, 2 vs 3, 3vs 4 and 1 vs 4. The initial test–retest measures were 24 h apart, after which time points were a mean of three days apart. The test–retest data were excluded in the responsiveness analysis. We also used generalised linear regression models to assess for change in total C-POS scores for the child (self and proxy versions) and family item subscales over time controlling for age and country setting. The latter approach is more robust as it uses all observations, as opposed to the selected paired [76].

Acceptability

We measured both time and extent of completion as proxy indicators of measure acceptability [40]. For feasible use in clinical settings for palliative care populations, measures should be short [77]. In regards to completion rates, although missing values commonly range from 4 to 18%, a threshold of 8% is acceptable [78]. For this study therefore, we adopted an acceptable item completion level of 8% for acceptability.

Interpretability

Studies have consistently shown that there is little variability in the standard deviations derived from between-subject differences at baseline, change scores or difference between change in scores [79]. We therefore used the 0.5 standard deviation of baseline scores to compute the minimum important difference for the child and family total scores [80]. For the two versions of the C-POS, we computed the total C-POS scores for child and family items. To assess for cross -cultural validity, we fit three multiple indicator cause models (self-report child items; proxy child items and family subscale) controlling for age to explore differential item functioning by country setting. We set a stringent P value of 0.001 or < 0.001 as a requirement for statistical significance

considering Bonferroni corrections for multiple testing and coefficients of at least 0.64. Items with differential item functioning would be excluded from the scale to avoid inflating the type II error.

Results

Sample characteristics

Of 454 children approached, none refused and 20 were not enrolled because they were deemed too ill to participate, by their care providers. Therefore 434 children and 431 family caregivers were recruited (95.4% of those approached). The most common diagnoses were HIV/AIDS (50.0%), sickle cell (14.6%) and cancer (12.2%). Where the child completed the child-oriented items alone, the child's mean age was 9.7 years (SD = 3.4). The socio-demographic characteristics of the children are presented in Table 1.

Construct validity

Structural properties Item 7 (having questions about your illness answered) had a high number of missing values on the proxy version (item coverage 0.73) and was therefore omitted in the analysis of the C-POS proxy version (see Table 2).

For the self-report version, from model 1, item 3 (eating) had a highest residual variance and therefore was removed from the model, and model fit was examined further. From Table 3, model 2, which excludes the "feeding" item, has a much better fit compared to model 1 (CFI index is 0.941, TLI is 0.901 and RMSEA is 0.043). On average, the indices moderately satisfy the cut offs quoted by Hu and Bentler of 0.95 for the

Table 1 Participants' characteristics for the quantitative cohort ($n = 434$)

Variable		Kenya ($n = 99$)		Uganda ($n = 233$)		South Africa ($n = 102$)	
Age	Mean (SD)- self report version	7.9(2.5)		10.8(3.7)		7.4(3.9)	
	Mean (SD)-proxy report version	3(2.4)		2.6(2.3)		2.8(3.4)	
sex		n	%	n	%	n	%
	Female	44	44.4	114	48.9	63	61.76
	Male	55	55.6	119	51.1	39	38.2
place of care							
	In patient	42	42.4	70	30.0	101	99.0
	Out patient	57	57.6	163	69.96	1	1.0
Primary diagnosis	HIV/AIDS	44	44.4	177	76.3	7	6.9
	Cancer	16	16.2	52	22.4	21	20.6
	Sickle cell	39	39.4	1	0.4	4	3.9
	Epilepsy	0	0	2	0.9	23	22.5
	Renal failure	0	0	0	0	3	2.9
	Cardiac disease	0	0	0	0	19	18.6
	TB	0	0	0	0	6	5.9
	other	0	0	0	0	19	18.6
Location of home							
	Rural	47	47.5	156	66.9	13	12.7
	Urban	52	52.5	77	33.1	89	87.3
Relationship of caregiver with child	Mother	82	82.8	117	50.2	87	85.3
	Father	7	7.1	15	6.4	6	5.9
	Other relative	5	5.1	32	13.7	2	2.0
	Grand parents	4	4.0	37	15.9	3	2.9
	Auntie/uncle	1	1.0	32	13.7	4	3.9
Language used	Luo	76	76.8				
	Kiswahili	21	21.2				
	English	2	2.0	58	25	44	43.1
	Zulu	0	0	0	0	31	30.4
	Xhosa	0	0	0	0	27	26.5
	Luganda	0	0	174	75	0	0

Table 2 Completeness of Data based on the repeated measures cohort

Item	n(children)			
	T1 (*n* = 302)	**T2(*n* = 297)**	**T3 (*n* = 285**	**T4(*n* = 279)**
Child-oriented items	*Missing values: child-reported(caregiver-reported)*	*Missing values: child-reported(caregiver-reported)*	*Missing values: child-reported(caregiver-reported)*	*Missing values: child-reported(caregiver-reported)*
Item 1 pain	0 (0)	0(0)	0 (3)	0 (0)
Item 2 other body	0 (0)	0 (0)	0 (0)	2 (0)
Item 3 eating	0 (0)	0(0)	0(0)	2(0)
Item 4 cried	1 (0)	1 (0)	1 (0)	3 (0)
Item 5 happy	0 (1)	0 (0)	0 (0)	2 (1)
Item 6 playing	1 (2)	0 (1)	6(1)	7 (1)
Item 7 questions answered	3(57)	2(*)	8(*)	8(*)
	n (family caregivers)			
Caregiver/family-oriented items	*N = 299*	*N = 282*	*N = 272*	*N = 270*
Item 8 worry	2	0	3	5
Item 9 Shared own feelings	2	0	4	5
Item 10 Received adequate info	1	0	5	5
Item 11 Received enough help and advice	5	2	0	0
Item 13 Family confidence	1	1	3	6

* no further analysis was conducted because of the high number of missing values resulting into a low item coverage

Table 3 Confirmatory factor analyses model fit indices for the models evaluated

Model	n	X (df)	Comparative fit index	Tucker-Lewis index (TLI)	Root mean square error of approximation (RMSEA)	RMSEA -90%CI	Standardized Root Mean Square Residual (SRMR)
Self-report version							
Model 1 with all 7 items	221	34.553*(14)	0.933	0.899	0.062	0.048–0.116	0.046
Model 2 minus feeding item (n for items = 6)	221	25.888*(9)	0.941	0.901	0.043	0.052- 0.135	0.044
Proxy-report							
Model 1 with 6 items Excludes item on questions answered (n for items = 6)	213	52.733(9)	0.925	0.876	< 0.001	0.113- 0.192	0.056
Model 2 Excludes feeding and questions answered items (n of items = 5)	213	39.950(5)	0.936	0.871	< 0.001	0.132–0.235	0.050
Assessing the self and proxy report versions for invariance by type of respondent							
Configural	434	63.218*(10)	0.933	0.866	0.157	0.121- 0.195	0.048
Metric	Model does not converge due to item 4 invariance item						
Scalar	434	146.091(33)	0.858	0.914	0.126	0.105- 0.147	0.065
Metric invariance model – allowing item 4 to vary							
Metric invariance model – allowing item 4 to vary	434	63.218(10)	0.933	0.866	0.157	0.121- 0.195	0.048
Family version – 5 items	215	15.892(5)	0.862	0.724	0.057	0.048 -0.158	0.044
Family version – 4 items Less item 12	215	12.312(6)	0.918	0.864	0.234	0.000 -0.126	0.045

Df degrees of freedom, *CFI* comparative fit index, *TLI* Tucker–Lewis index, *RMSEA* root mean square error of approximation, *95% CI* 95% confidence intervals

comparative fit index, 0.06 for the standardized root mean square residual [81].

For the *proxy version, model* 1 of the proxy version has 6 items (pain, other body problems, feeding, cried, happy and play). Inspecting the model modification indices, the feeding item showed a residual variance. Removing the feeding item from the model did not meaningfully impact on fitness indices (see Table 3). Given that feeding was an emergent theme from the qualitative interviews, and the fact that it is negatively worded and hence susceptible to construct distortion bias, we decided to retain it and the wording problem can be addressed.

For the family sub-scale, the model with 5 items showed moderate fit, any further changes in content did not improve the model fit.

Discriminant validity A total of 302 children/family caregivers completed the C-POS and the PedsQL. The Spearman's rho correlation coefficients for the selected best fit C-POS and PedsQL subscales scores for child items (self and proxy) and the PedsQL scores were low as hypothesized (all < 0.6) (see Table 4).

Known groups' comparison For both the child and proxy versions, patients with worse functional performance (ECOG) reported more severe palliative care-related symptoms and concerns (CPOS) compared to those with better functional performance (see Fig. 2).

Measurement invariance We found partial metric variance due to item 4 (cried). We inspected the source of variance and noted it was at the threshold level and caregivers were more likely to rate cried highly or severely as compared to the children. Allowing item 4 to vary depending on type of respondent improved the model fit, comparing the scalar model fit indices with the model that allows item 4 to vary by type of respondent, shows that the latter had a better fit (see Table 3).

Differential item functioning For our MIMIC model assessing item functioning variation by country controlling for age, at a *P* value cut off of < 0.001, we did not find any item functioning variation by country setting for the self-report and proxy report versions (see supplementary results e-table S1 and S2).

Table 4 Correlations of PedsQL and C-POS subscale summary scores for discriminant validity

PedsQL summary scores	C-POS subscale scores	Spearman's correlation (proxy version)	Spearman's correlation (self-report version)
Total PedsQL score	C-POS patient items	0.308	0.117
PedsQL Physical health summary score	Sum- C-POS (Pain) + (other problems)	0.291	0.029
PedsQL Psychosocial health summary score	Sum C-POS (felt happy) + Q6(felt like playing) + Q (questions about illness)	0.256	0.133

C-POS items –includes 6 patient items, we excluded one item (questions answered because of the high number of missing values)

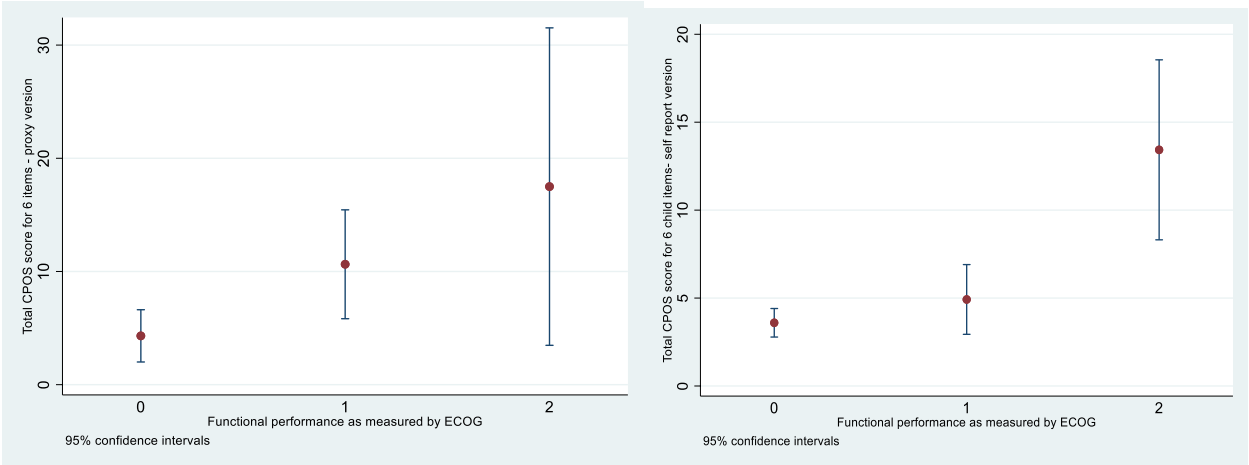

Fig. 2 Comparison of CPOS mean scores by functional performance as measured by the ECOG for known groups validity. Note: this analysis is based on a sub-sample of children aged 5 and above to whom it was feasible to use the ECOG; proxy -report version (*n* = 31); self-report version (*n* = 101). Higher scores indicate more palliative care related concerns

Reliability

Internal consistency For both versions of the C-POS, the omega reliability coefficients for the child item subscale showed acceptable internal consistency (omega coefficients proxy version- 0.71 and child version 0.67. We also found moderate internal consistency for the family sub-scale omega coefficient (0.49). Inspecting the item information functions, C-POS proxy report (achieved satisfactory internal consistency, item information range 3–9), the self-report version, item information range was 3–5, be strengthened by including more items that addressed concerns of children with less severe palliative care problems. The family subscale could be strengthened by including items that reflect the concerns of carers for children with more severe palliative care-related concerns, as that is where the standard error is biggest. For item information (i.e., reliability ¼ 1 _ [1 /information]) the values of 3.3 to 10 [42], correspond to the recommended Cronbach's alpha scores of 0.70 to 0.90 for interpreting reliability (see Fig. 3 in Appendix for details).

Test-re-test reliability The C-POS was completed by 297 children at two visits; the test–retest analysis is based on a sub-sample of 152 in-patients whom we hypothesised remained stable during the test–retest period. The weighted Kappa scores were good for all items, with more subjective items generally showing lower reliability than the physical outcomes (see Table 5).

Responsiveness

The GEE analysis showed a positive change (improvement/reduction in palliative care related concerns) for most items (5/7 questions for child self-report version and 6/7 questions for the proxy-report version) (see Table 6). For both versions, the tool did not demonstrate responsiveness for the feeding item. For the self-report version, the tool did not demonstrate responsiveness on the items of cried, felt happy and questions about illness answered.

Controlling for country and age, generalised linear regression analysis showed statistically significant reductions in palliative care concerns over time for both the self and proxy report versions of the C-POS and the family sub-scale. We found no differences due to age and country setting variation in palliative care-related concerns over time for self-report version of the C-POS (see supplementary Table 2).

Acceptability of the C-POS

For the C-POS child self-report version, the median completion time reduced from 25 min (IQR 20–30) at visit 1 to 10.5 (IQR (10–15) at visit 4. For the C-POS proxy version, the median time for completion reduced from 20 (IQR 15–25) at visit 1 to 10 min (IQR 7–13 min) at visit 4. The C-POS items were acceptable to participants, as shown by the infrequent missing values for any time point (0% for "pain" to 2.8% for "questions answered"). Data coverage for item 7 – question about illness

Table 5 Test–retest reliability (children *n* = 152, carers *n* = 127)

In patients (152)							
Child version (*n* = 33)					**Proxy version (*n* = 119)**		
Child items				**Child items**			
	Weighted Kappa statistic	Agreement within one score	Expected agreement		Weighted Kappa statistic	Agreement within one score	Expected agreement
Item 1 pain	0.5364	80.63%	58.20%	Q1	0.5239	81.87%	61.91%
Item 2 other body symptoms	0.4995	80.63%	61.29%	Q2	0.4791	77.33%	56.49%
Item 3 eating	0.5741	82.50%	58.91%	Q3	0.6421	83.73%	54.55%
Item 4 cried	0.4237	86.72%	76.95%	Q4	0.3477	76.53%	64.02%
Item 5 happy	0.4863	78.75%	58.63%	Q5	0.5950	81.74%	54.92%
Item 6 playing	0.4348	73.75%	53.55%	Q6	0.5861	81.08%	54.29%
Item 7 questions answered	0.5224	80.00%	58.12%	Q7	0.3613	71.91%	56.03%
Family items (*n* = 127)							
Item 8 worry	0.5047	76.52%	52.60%				
Item 9 Shared own feelings	0.6814	88.63%	64.33%				
Item 10 Received adequate info	0.6487	88.55%	67.41%				
Item 11 Received enough help and advice	0.6942	88.89%	63.67%				
Item 12 Family confidence	0.7086	95.68%	85.19%				

Table 6 Responsiveness analysis results using Wilcoxon rank sum test for matched pairs

	T1 Mean (SD) Median (IQR)	T2 Mean (SD) Median (IQR)	T3 Mean (SD) Median (IQR)	T4 Mean (SD) Median (IQR)	*P* values for differences between paired observations			
					T1 -T3	T2-T3	T3-T4	T1-T4
Self-report version ($n=71$)								
Item1 Pain	2.4(1.9) 2(0–4)	1.9(1.7) 1(0–3)	1.4(1.7) 1(0–3)	1.3(1.6) 0(0–2)	< 0.001*	0.055	0.4452	0.0001*
Item2 other body problems	2.1(1.8) 2(0–4)	1.7(1.8) 1(0–3)	1.5(1.7) 1(0–3)	1.5(1.8) 1(0–3)	0.008*	0.065	0.963	0.011*
Item 3 Feeding	2.5(2.0) 3(0–5)	2.3(1.9) 2(0–4)	2.3(2.2) 2(0–5)	2.3(2.2) 2(0–5)	0.402	0.887	0.3652	0.417
Item 4 Cried	0.7(1.4) 0(0–1)	2.1(1.6) 2(1–3)	0.6(1.2) 0(0–1)	0.6(1.5) 0(0–0)	0.855	0.433	0.786	0.224
Item 5 felt happy	1.6(1.7) 1(0–3)	1.6(1.8) 1(0–3)	1.3(1.6) 1(0–3)	1.3(1.6) 0.5(0–2)	0.150	0.142	0.884	0.217
Item 6 playing	2.2(2.2) 2(0–5)	2.0(2.1) 1(0–5)	1.6(2.2) 0(0–3)	1.9(2.5) 0(0–3)	0.007*	0.073	0.861	0.130
Item 7 questions answered	1.7(1.9) 1(0–3)	1.6(1.9) 1(0–3)	1.5(2.2) 0(0–3)	1.6(2.4) 0(0–3)	0.112	0.191	0.897	0.631
Proxy version ($n=159$)								
Item1 Pain	2.1(1.8) 2(0–3)	1.3(1.6) 1(0–3)	1.4(1.7) 1(0–3)	1.3(1.7) 0(0–3)	< 0.001*	0.2089	0.3550	< 0.001*
Item2 other body problems	2.6(1.9) 3(1–4)	2.0(1.7) 2(0–3)	1.6(1.7) 1(0–3)	1.5(1.6) 1(0–3)	< 0.001*	0.005*	0.1987	< 0.001*
Item 3 Feeding	2.7(2.0) 3(0–5)	2.7(2.0) 3(0–5)	2.8(2.0) 3(0–5)	2.8(2.1) 3(0–5)	0.402	0.678	0.5237	0.459
Item 4 Cried	2.1(1.6) 2(1–3)	1.0(1.7) 0(0–2)	1.6(1.6) 1(0–3)	1.3(1.5) 1(0–2)	0.001*	0.4186	0.001*	0.001*
Item 5 felt happy	2.4(1.9) 3(0–4)	2.2(1.9) 3(0–4)	1.9(1.8) 2(0–3)	1.7(1.8) 1(0–3)	0.001*	0.032	0.0602	0.001*
Item 6 playing	2.3(2.0) 2(0–5)	2.3(1.9) 2(0–4)	2.1(2.0) 2(0–4)	1.7(2.0) 1(0–3)	0.062	0.148	0.001*	0.001*
Item7 questions answered	2.7(1.9) 3(1–5)	2.3(1.9) 3(0–4)	2.1(2.0) 2(0–4)	2.4(2.4) 2(0–4)	0.002*	0.1458	0.901	< 0.001*
Family ($n=293$)								
Item 8 worry($n=293$)	3(2.1) 3(1–5)	2.8(2.0) 3(1–5)	2.9(1.8) 3(2–5)	2.7(2.0) 3(1–5)				
Item 9 Shared own feelings ($n=272$)	1.5(1.9) 0(0–3)	1.5(1.9) 1(0–3)	1.5(1.9) 0(0–2)	1.1(1.8) 0(0–2)	0.030*	0.014*	< 0.001*	< 0.001*
Item 10 Received adequate information ($n=294$)	1.6(1.8) 1(0–3)	1.4(1.7) 1(0–2)	1.4(1.7) 1(0–2)	1.2(1.7) 0(0–2)	< 0.001*	0.066	0.006*	< 0.001*
Item 11 Received enough help and advice ($n=290$)	2.2(2) 2(0–5)	1.9(1.8) 2(0–3)	1.9(1.9) 1(0–3.5)	1.8(1.8) 1(0–3)	0.001*	0.272	0.0006	< 0.001*
Item 12 Family confidence ($n=268$)	0.9(1.6) 0(0–1)	0.8(1.4) 0(0–1)	0.6(1.2) 0(0–1)	0.5(1.1) 0(0–1)	0.001*	0.023*	0.004*	0.001*

Statistically significant *p* values are bolded—Significant at *5% **1% ***0.01% levels

Time difference between T1 and T2 is 1 day, T2 and T3 is 3 days T3 and T4 is 3 days

answered was 0.72, suggesting that the item is problematic. Details of these findings were presented in Table 2 at point of assessing item level data completeness.

Interpretability

The mean scores for the child and proxy versions were 14.07 (SD- 6.8)-proxy and 10.69 (6.03) respectively. Therefore, the minimum important difference for total score on the six child items alone is 3 on a scale of 0–30. For the family 11 item sub-scale, the mean scores were 20.1 (SD 8.6) (proxy version) and 23.9 (SD8.2) (self-report version) yielding a minimum important difference of 4, on a scale of 0–55.

Discussion

This study provides evidence for the validity, reliability, responsiveness to change and acceptability, and interpretability of both the child and proxy versions of the novel C-POS. This comprehensive multi-dimensional person-centred outcome measure for use in children's palliative provides an important contribution to the advancement of the science and practice of outcome measurement for children and young people.

Due to the absence of a gold standard multi-dimensional outcome measure, we selected a comparative tool: the PedsQL. We found evidence for the divergent validity of the C-POS using the PedsQL. The C-POS total scores and subscale scores showed low/moderate correlation to the PedsQL, suggesting the instrument discriminates dissimilar constructs. The PedsQL is a function-oriented quality of life measure while the C-POS is a multidimensional outcome measure and therefore measures different constructs from this measure [82, 83].

Although the psychometric properties of the C-POS were found to be acceptable, a low test–retest reliability score was obtained for the 'cried' and 'questions answered' items on the proxy version of the C-POS. This could be because of the subjectivity of the items or an indication of measuring difficulties of very subjective constructs in young populations [70, 84]. For example, children with advanced cognitive development age may show other emotions beyond crying. For the self-report version C-POS, responsiveness was not demonstrated for the two items of "cried" and "felt happy." This could be explained by the fact that the two items are inversely related, the happiness item within this subsample will tend to have a more restricted range of scores than for the full sample. Also, these items had floor and ceiling effects which limits variability and hence ability to show change over time. Lastly, the proxy version of the C-POS did not demonstrate change for the item of feeding, possibly due to the challenge of scoring feeding.

The acceptability of the C-POS is demonstrated by its minimum patient burden in terms of its completion time and very limited missing data. Moreover, the C-POS items were acceptable, the scale was judged as relevant to the population, and instructions were clear. Item 7 ("how much have questions about your sickness have answered since yesterday") had the highest number of missing values on the proxy version. It might be explained by common difficulties proxies face in reporting unobservable constructs without input from patients themselves [85]. We suggest that this question is asked directly to children or and observable indicators can be used for the proxy version, the question should be avoided for very young children who cannot ask questions given their cognitive development age.

Themes developed from the qualitative interviews highlighted spiritual and existential concerns particularly amongst older children [6]. This finding demonstrates the robustness of the spiritual /existential domain of health and well-being which is equally important in children as for adults [86]. We therefore recommend inclusion of an item to cover this domain of well-being. The themes mirrored how patients and families adjusted to self, community and environment as it related to the perceived purpose of life, life satisfaction and life experiences [87]. The theme of financial problems was raised by adolescents but should be interpreted with caution as respondents commonly anticipate financial support if they present such difficulties. As countries move towards integrating palliative care into healthcare systems using a public health approach, additional items or modules might be developed and validated to add as necessary to this core outcome measure. This approach has been used for newer versions of the POS, such as an expanded symptom module and for specific diagnostic groups.

Measure refinement

Based on the findings, we recommend the following minor revisions:

First, remove the feeding item as it provides the least information explaining variation in the latent construct; moreover, it has considerable overlap with the "other body problems" item for the self-report version. This item was, however, negatively worded, and this could affect its functionality; re-wording it may therefore be an option also worth exploring.

Second, all negatively worded items should be re-worded as evidence has consistently shown that such items tap different constructs. Item 8 (family worry) and item 3 (feeding), fall in this category.

Third, item 7 (questions about illness answered) should be revisited; it can be posed to the children, but an observable alternative may be more useful for the proxy version. Moreover, the question should be omitted for very young children who cannot ask questions because of developmental/cognitive age problems.

Fourth, the crying item showed metric variance should be revisited as older children, who complete the self-report version, are more likely to express emotions such as sadness or feel like crying as opposed to crying.

Fifth, we suggest additional items to address the following conceptual gaps: spiritual/existential and normalcy. Items with small discriminating power could be replaced

or additional items added to maximise impact. The developmental and chronological age differences in themes should be addressed, age group-specific prototypes should be considered, and the current questions can be modified slightly to address this gap.

Strengths and limitations

The C-POS has been developed according to highest scientific principles and guidelines for tool development and validation. Importantly this measure has been developed across a range of countries, settings (inpatient, outpatient and daycare) and diagnostic and cultural groups. Second, the self-report version of the C-POS was not used in children who were too ill to complete the tool, a common occurrence in paediatric palliative care. These children may be unable to self-report due to young age, advanced illness, type of illness or fatigue as they near death [88] Proxy reporting from family caregivers is acceptable in such situations, given the time spent together during their illness and this can be adopted as best practice [56].

There are several limitations of this study that need to be taken into consideration. First, the development of person-centred outcome measures is an evolutionary process, and we identified some areas for improvement which should be considered. Second, we also did not explore minimum important clinical difference, which is useful for interpreting changes in scores in clinical settings. Third, grouping children by stage of disease trajectory would be useful for informing construct validity assessment by known groups but this is complicated by the unpredictability of disease trajectories being mindful of the 400 conditions that require palliative care in young people [89]. More so, due to language diversity in the countries, the interviews were conducted in different languages which were later translated into English and combined in the analysis. This could introduce some language level biases.

Conclusion

With the minimal changes proposed above, the C-POS has potential to measure person-centred outcomes in paediatric palliative care. The vast majority of evidence for the concept of practice of person-centredness in serious physical illness has focused on adults [90, 91]. The data in this study expands this to conceptualising and measuring child- and family-centred outcomes. The palliative care of children and young people has become a global health priority [92]. It is therefore important that resources are allocated for adaptation of the C-POS where required, and sustained implementation [93] to improve outcomes and inform a culture shift to outcome-focused services.

Appendix

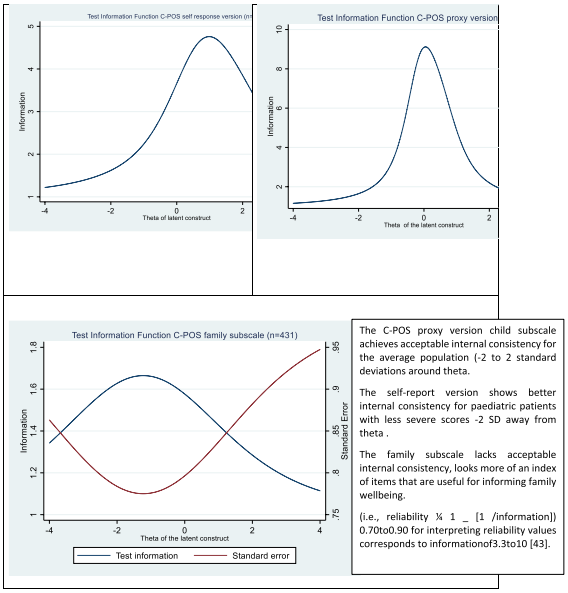

Fig. 3 Item test function curves illustrating internal consistency properties of the C-POS subscales

Abbreviations

APCA	African Palliative Care Association
CFI	Comparative Fit Index
C-POS	Children's Palliative Care Outcome Scale
ECOG	Eastern Cooperative Oncology Group Scale
LLCs	Life-limiting and life-threatening conditions
MIMIC	Multiple Indicator, Multiple Cause Modelling Controlling
PCOM/PCOMs	Person-Centred Outcome Measure(S)
PedsQL	Paediatric Quality of Life Inventory
RMSEA	Root Mean Square Error of Approximation
SD	Standard Deviation
SEM	Structural Equation Modelling
SRMR	Standardized Root Mean Square Residual
TLI	Tucker-Lewis Index

Supplementary Information

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Supplementary Materials 1 and 2

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Authors' contributions
Study concept and design, analysis and interpretation of the data drafting of the manuscript critical revision of the manuscript for important intellectual content: EN, KB, FEM, JD , MA , MS, RH . Analysis and interpretation of the data, drafting of the manuscript, critical revision of the manuscript for important intellectual content: IJH , LF, MA, FM-P, RAP Study supervision, drafting of manuscript, administrative, technical, and material support: ZA , MM. All authors read and approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

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Data availability
The data can be accessed by contacting the authors.

Declarations
Ethical approval and consent to participate
Ethical approval was obtained from King's College London, Research Ethics Office (LRS-15/16–3524) and in the three countries: Uganda National Council for Science and Technology (UNCST SS2366), Kenya Medical Research Institute (KEMRI/RES/7/3/1), and the South Africa Hospice and Palliative Care Association (HPCA 03/10).
All participating children aged 8 years and above gave assent, while adult caregivers gave informed written consent for children to participate.

Competing interests
The authors declare no competing interests.

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