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ORIGINAL ARTICLE

Modelling quality of life in children with intellectual disability using regression trees

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Aim: To identify factors associated with quality of life (QoL) in children with intellectual disability. We aimed to identify patterns of association not observable in previous hypothesis-driven regression modelling using the same data set from a cross-sectional observational study.

Method: A questionnaire was completed by 442 caregivers of children with confirmed intellectual disability and a diagnosis of autism spectrum disorder, cerebral palsy, Down syndrome, or Rett syndrome. The Quality of Life Inventory-Disability (QI-Disability) questionnaire was used to assess child QoL. Independent variables described the child's health, functional abilities, community participation, and sociodemographics. The R package *rpart* was used to build the regression trees.

Results: The mean total QI-Disability score was 69.2 out of a maximum 100. The subgroup with the lowest QoL scores comprised children with a high degree of daytime sleepiness (n=74, mean 57.5) while the subgroup with the highest QoL scores (n=91, mean 80.3) comprised children with little daytime sleepiness who participated more frequently in community activities and displayed good eye contact while listening.

Interpretation: Regression tree analysis provides insights into the relative importance of associated factors. Sleep problems and community participation were more important than functional abilities in accounting for differences in QoL.

Quality of life (QoL) refers broadly to satisfaction with life in the context of culture and goals. QoL is a multidimensional concept comprising physical, emotional, and social well-being as well as aspects of personal development and activity.[1] Beyond well-being, this set of dimensions has been expanded to also include independence (interpersonal relations and self-determination) and social participation (personal development, social inclusion, and rights) in the field of intellectual disability.[2] Many factors can threaten or enhance the QoL of children with intellectual disability and multivariate models are necessary to provide insights into these complexities. Using a QoL tool validated for children with intellectual disability,[3] we have reported that poorer functioning, notably a high level of dependence for daily needs, was associated with poorer QoL and poorer QoL can be partly explained by less frequent community participation.[4] We also found that comorbid health conditions are associated with marked negative effects on QoL,

This original article is commented on by Gómez on pages 1056–1057 of this issue.

Abbreviations: CART, Classification and regression trees; DOES, Disorders of Excessive Somnolence; ICF, International Classification of Functioning, Disability, and Health; QI-Disability, Quality of Life Inventory-Disability; QoL, Quality of life.

particularly the presence of recurrent pain and sleep dysfunction.[5] We used hypothesis-based regression modelling in each of these studies and there are likely other important factors that could influence QoL in children with intellectual disability.

Regression trees analysis is a subset of classification and regression trees (CART)[6, 7] and can be used to identify and characterize subgroups of individuals with high or low QoL scores. This alternative approach is not hypothesis-driven and could provide advantages to the more traditional linear modelling approach. First, it does not rely on any assumptions about the functional relationship, such as linearity, between independent and outcome variables, nor does it make any distributional assumptions, such as normality, about the outcome variable. Second, the process of cross-validation built into the regression trees algorithm means that the problem of overfitting from including a large number of independent variables is reduced and the prediction accuracy of the final CART model is likely better than a traditional regression model. Third, interactions between independent variables, which are difficult to interpret in a traditional model, arise naturally from the CART process with the subgroups of interest defined by a combination of high or low scores on key independent variables. Although not a novel methodology, CART has not, to the authors' knowledge, previously been used to investigate QoL in children with intellectual disability and has the potential to provide novel insights about the interplay of factors associated with QoL.

Using a broader suite of independent variables than for our previous linear modelling approach, the current study implemented a regression tree analysis and aimed to identify the salient factors associated with QoL for children with intellectual disability, potentially identifying clinical associations beyond our previous hypothesis-driven analyses.

What this paper adds

- A hypothesis-free regression tree analysis enables examination of multiple factors potentially influencing quality of life (QoL) in children with intellectual disability.
- Functional abilities were less strongly associated with QoL than sleep problems and community participation.

METHOD

Design

This was a cross-sectional observational study.

Data sources

Caregivers of a child aged 5 to 19 years with confirmed intellectual disability and a diagnosis of either autism spectrum disorder, cerebral palsy, Down syndrome, or Rett syndrome were recruited. We selected diagnoses that would represent a range of different co-occurring problems in intellectual disability, including medical, physical, communication, and behavioural difficulties, and where the investigators had access to databases on these conditions. Families were contacted through population-based registers and other data sources and were invited to complete an online questionnaire as described previously.[4] A small number of caregivers provided data using a paper format or via telephone interview. Ethics approval for this study was obtained by Human Research Ethics Committees at The University of Western Australia (RA/4/20/4276) and the Child and Adolescent Health Services (RGS2390). Primary caregivers provided informed consent to participate in the study.

Outcome variable

The Quality of Life Inventory-Disability (QI-Disability) questionnaire is a 32-item parent-report measure assessing the QoL of children with intellectual disability. The instrument has been described in detail elsewhere with evidence for satisfactory reliability and validity.[3, 8, 9] The questionnaire comprises six domains: social interaction (7 items), positive emotions (4 items), negative emotions (7 items), physical health (4 items), leisure and the outdoors (5 items), and independence (5 items). Items are linearly transformed to a scale of 0 to 100, with higher scores representing better QoL. Domain scores are calculated by averaging item scores and total scores by averaging domain scores.[3]

Independent variables

The independent variables encompassed each of the domains of the International Classification of Functioning, Disability, and Health (ICF) model,[10] including impairments, participation in activities, personal factors, and how the child lives within the environment. All of these have potential to influence a child's QoL.

Health conditions

The Disorders of Initiating and Maintaining Sleep and the Disorders of Excessive Somnolence (DOES) subscales of the Sleep Disturbance Scale for Children[11] were used to describe sleep. These scales use T scores based on normative data. Parents rated their child's experiences of pain over the previous month as 'not at all', 'occasionally', or 'recurrently'. Frequency of seizures was described as 'controlled', 'fewer than once per month', 'monthly', or 'daily or weekly'. Scoliosis was classified

as 'no scoliosis', 'mild or moderate scoliosis', 'severe scoliosis treated with surgery', or 'severe scoliosis managed conservatively'. Caregiver-reported 'yes' responses for the presence or treatment of constipation or gastroesophageal reflux indicated a gastrointestinal problem. As a proxy measure of respiratory health, the number of courses of antibiotics prescribed for respiratory infections in the previous 12 months were coded as 'none', 'once', and 'twice or more'. The number of fractures in the previous 12 months was classified as 'none' or 'one or more'. Nights in hospital in the previous 12 months was classified as 'none', 'fewer than 4 nights', or '4 or more nights'. Children with mental health/behavioural problems were identified if the caregiver reported a physician-diagnosed mental health disorder or attention-deficit/hyperactivity disorder, or their child had been prescribed a psychotropic medication. A novel item was constructed to measure the primary caregiver's perception of whether their child's medical needs had been met overall during the previous 12 months with responses provided on a 7-point Likert scale.

Child functional abilities

Mobility was categorized as 'able to walk at least 500 metres with no difficulty', 'able to walk independently but for shorter distances', 'able to walk with assistance', or 'unable to walk'. Communication was categorized as 'speaks well', 'some difficulty speaking such as lack of clarity', 'difficulty speaking and only understood by those who know them well', 'non-verbal communication', and 'unable to communicate'. Parents categorized their child's function in relation to personal needs as 'independent', 'independent but needing monitoring or reminding', 'needing assistance', or 'fully dependent'. Selected questions from the Eye Contact Avoidance Scale[4, 12] were used to measure the child's eye contact during social functioning when they initiate communication. Eye contact when communicating with the parent, friends, and family, and when communicating with unfamiliar people were each rated on a 0- to 4-point Likert scale (0=never, 1=rarely, 2=sometimes, 3=often, 4=always) and then summed to give a total possible score of 12.

Child community participation

Participation was assessed using the community module of the Participation and Environment Measure for Children and Youth.[13] For each item, parents were asked how frequently their child attended activities (such as neighbourhood outings, organized physical activities) and how involved their child was participating in the activity. An overall frequency score was calculated by averaging the frequency scores from across the 10 items (scored out of 7). Involvement scores were calculated by averaging the responses to items in which the child took part as previously (scored out of 5).[14]

Sociodemographic, caregiver, and family factors

Income was classified as over or under A$84 000 per annum, and the family was classified as a single or dual (including shared parenting) parent family. The number of siblings and whether the child with intellectual disability was a firstborn were also included. Parental distress was assessed using the Kessler Psychological Distress Scale and family QoL was assessed using the Family Quality of Life Scale, a 25-item parent-report inventory.[15, 16]

Statistical analysis

The R package *rpart* (R Foundation for Statistical Computing, Vienna, Austria)[17] was used to build the regression trees. The algorithm first finds the predictor variable which, with an appropriate cut-off value, best splits the sample into two subgroups or nodes, the criterion being to optimize model fit by minimizing the within-group variability (error sum of squares) of the outcome variable. The process then continues in a similar fashion by splitting each node until the subgroups reach a minimum size (defined here as 20) or no improvement in model fit can be achieved by splitting a node. A process of 'pruning' is then employed using cross-validation to reduce the tree and avoid overfitting the data. We used 10-fold cross-validation which consists of generating 10 non-overlapping test samples from the data set and evaluating the average model fit over these test samples for a series of trees of reducing complexity. The final parsimonious tree selected has the least complexity while satisfying the prespecified criterion that model fit is within one standard error of the tree with the best cross-validated model fit. Where an observation has a missing value for a variable used in a split, a surrogate variable is used for subgroup assignation.[17]

A variable importance plot was generated for each tree. This ranks the predictor variables according to their contribution to the improvement in model fit for all the splits in which that variable is used.

A regression tree was assembled for the QI-Disability total score and separate trees for each domain.

RESULTS

Between March 2018 and October 2019, a questionnaire was completed by 442 out of 585 (75.6%) invited parents/primary caregivers, including 130 out of 162 (80.2%) with a child with autism spectrum disorder, 151 out of 229 (65.9%) with a child with cerebral palsy, 89 out of 98 (90.8%) with a child with Down syndrome, and 72 out of 96 (75.0%) with a child with Rett syndrome. Most (90.6%) respondents were biological mothers. The median (range) age of the children was 11 years 8 months (5y 1mo–19y 1mo). Distributions for the predictor and QoL variables are presented in Table 1.

TABLE 1 Variables used in regression tree models stratified by diagnostic group

		Total (*n*=442)	ASD (*n*=130)	CP (*n*=151)	Down (*n*=89)	RTT (*n*=72)
	Age (years:months)	11:8 (5:1–19:1)	11:1 (5:6–18:7)	13:10 (5:11–19:1)	10:6 (5:1–18:5)	11:10 (5:2–18:11)
	Sex (female)	218 (49.4)	33 (25.4)	60 (40.0)	53 (59.6)	72 (100)
Child comorbidities						
Sleep, *n*=435	DIMS (T score)	69.6 (40.7–130.7)	72.8 (43.9–127.5)	72.8 (43.9–124.3)	63.2 (40.7–121.1)	63.2 (40.7–130.7)
n=438	DOES (T score)	57.4 (41.8–119.6)	51.5 (41.8–114.7)	57.4 (41.8–119.6)	53.5 (41.8–104.0)	65.1 (41.8–119.6)
Pain, *n*=441	None	166 (37.6)	57 (43.9)	44 (29.3)	36 (40.5)	29 (40.3)
	Occasional	199 (45.1)	62 (47.7)	69 (46.0)	45 (50.6)	23 (31.9)
	Recurrent	76 (17.2)	11 (8.5)	37 (24.7)	8 (9.0)	20 (27.8)
Seizures, *n*=436	None	264 (60.6)	108 (83.1)	62 (41.1)	84 (94.4)	10 (15.2)
	Controlled	44 (10.1)	6 (4.6)	25 (16.6)	3 (3.4)	10 (15.2)
	<Monthly	41 (9.4)	5 (3.9)	25 (16.6)	0	11 (16.7)
	Monthly	24 (5.5)	4 (3.1)	10 (6.6)	0	10 (15.2)
	Daily or weekly	63 (14.5)	7 (5.4)	29 (19.2)	2 (2.3)	25 (37.9)
Scoliosis, *n*=431	None	327 (75.9)	126 (97.7)	96 (63.3)	84 (94.4)	21 (33.9)
	Mild or moderate	50 (11.6)	2 (1.6)	23 (15.2)	4 (4.5)	21 (33.9)
	Severe, had surgery	36 (8.4)	1 (0.8)	19 (12.6)	1 (1.1)	15 (24.2)
	Severe, no surgery	18 (4.2)	0	13 (8.6)	0	5 (8.1)
GI disorders, *n*=442		242 (54.8)	55 (42.3)	101 (66.9)	42 (47.2)	44 (61.1)
Antibiotic prescribed, *n*=440	None	238 (54.1)	85 (65.4)	63 (41.7)	52 (58.4)	38 (54.3)
	Once	84 (19.1)	21 (16.2)	32 (21.2)	20 (22.4)	11 (15.7)
	Twice or more	118 (26.8)	24 (18.4)	56 (37.1)	17 (19.2)	21 (30.0)
Fractures, *n*=439		24 (5.5)	5 (3.9)	11 (7.3)	3 (3.4)	5 (7.3)
Nights in hospital, *n*=439	None	298 (67.9)	117 (90.0)	72 (48.0)	62 (69.7)	47 (67.1)
	Fewer than 4 nights	51 (11.6)	8 (6.2)	19 (12.7)	16 (18.0)	8 (11.4)
	4 or more nights	90 (20.5)	5 (3.9)	59 (39.3)	11 (12.4)	15 (21.4)
Mental health disorder, *n*=442		75 (17.0)	53 (40.8)	15 (9.9)	4 (4.5)	3 (4.2)
Med care satisfaction, *n*=420		5 (1–7)	5 (2–7)	5 (1–7)	5 (1–7)	5 (1–7)
Child functional abilities						
Mobility, *n*=442	Walks unaided	132 (29.9)	89 (68.5)	12 (8.0)	31 (34.8)	0
	Short distances only	158 (35.8)	39 (30.0)	41 (27.2)	57 (64.0)	21 (29.2)
	Assistance needed	35 (7.9)	2 (1.5)	19 (12.6)	0	14 (19.4)
	Unable to walk	117 (26.5)	0	79 (52.3)	1 (1.1)	37 (51.4)
Communication, *n*=442	Speaks well	46 (10.4)	25 (19.2)	13 (8.6)	6 (6.7)	2 (2.8)
	Some difficulty	132 (29.9)	51 (39.2)	32 (21.2)	42 (47.2)	7 (9.7)
	Some words only	86 (19.5)	28 (21.5)	18 (11.9)	34 (38.2)	6 (8.3)
	Non-verbal only	118 (26.7)	19 (14.6)	49 (32.5)	6 (6.7)	44 (61.1)
	None	60 (13.6)	7 (5.4)	39 (25.8)	1 (1.1)	13 (18.1)
Personal needs, *n*=442	Independent	14 (3.2)	7 (5.4)	6 (4.0)	1 (1.1)	0

TABLE 1 (Continued)

		Total (*n*=442)	ASD (*n*=130)	CP (*n*=151)	Down (*n*=89)	RTT (*n*=72)
	Generally able	87 (19.7)	41 (31.5)	11 (7.3)	33 (37.1)	2 (2.8)
	Requires assistance	112 (25.3)	50 (38.5)	23 (15.2)	36 (40.5)	3 (4.2)
	Fully dependent	229 (51.8)	32 (24.6)	111 (73.5)	19 (21.4)	67 (93.1)
Eye contact,[a] *n*=430	Speaking	8 (0–12)	6 (0–12)	8 (0–12)	8 (0–12)	9 (0–12)
n=429	Listening	7 (0–12)	6 (0–12)	7 (0–12)	8 (2–12)	8 (0–12)
Participation[d]						
n=442	Frequency	1.9 (0.0–5.0)	2.1 (0.2–4.8)	1.5 (0.0–4.1)	2.4 (0.2–5.0)	1.7 (0.0–4.2)
n=437	Involvement	3.0 (1.0–5.0)	3.0 (1.0–5.0)	2.7 (1.0–5.0)	3.8 (`1.0–5.0)	2.3 (1.0–5.0)
Sociodemographic, caregiver, and family factors						
n=442	Any siblings	358 (85.5)	111 (85.4)	128 (84.8)	73 (82.0)	66 (91.7)
n=442	Firstborn	225 (50.9)	72 (55.4)	78 (51.7)	43 (48.3)	32 (44.4)
n=441	Single parent	65 (14.7)	21 (16.1)	24 (15.9)	10 (11.2)	10 (14.1)
n=365	Income (>$84 K pa)	171 (46.9)	59 (51.3)	39 (33.3)	45 (48.4)	28 (50.01)
n=439	Maternal distress[b]	20 (10–50)	20 (10–50)	20 (11–50)	18 (10–35)	21.5 (10–47)
n=433	Family quality of life[c]	3.7 (1.1–5.0)	3.6 (1.5–4.7)	3.7 (1.1–5.0)	3.8 (2.7–5.0)	3.9 (2.4–4.9)
Quality of life						
QI-Disability, *n*=442	Total	69.2 (12.7)	68.3 (10.9)	66.6 (13.5)	77.5 (11.7)	66.1 (11.2))
	Physical health	70.1 (16.7)	74.6 (15.6)	67.2 (16.6)	71.6 (16.6)	66.3 (16.9)
	Positive emotions	77.1 (17.8)	74.1 (16.3)	75.2 (19.2)	86.6 (14.7)	74.6 (16.9)
	Negative emotions	65.3 (18.9)	57.6 (18.6)	67.6 (19.8)	70.9 (16.4)	67.6 (16.0)
	Social interactions	70.4 (19.0)	61.5 (17.7)	70.9 (20.1)	80.0 (15.9)	74.3 (15.0)
	Leisure	70.4 (20.1)	72.5 (18.7)	64.5 (21.2)	78.3 (17.5)	69.5 (19.5)
	Independence	61.9 (24.0)	69.8 (16.3)	54.2 (26.0)	77.7 (17.7)	44.6 (20.6)

Median (range) for continuous variables. *n* (%) in category for ordinal variables. Mean (SD) for QI-Disability outcome variables. There was a small amount of missing data where questionnaire items had not been completed. The number of available responses for each variable is shown in the left-hand column.

Abbreviations: ASD, autism spectrum disorder; CP, cerebral palsy; Down, Down syndrome; RTT, Rett syndrome; DIMS, Disorders of Initiating and Maintaining Sleep; DOES, Disorders of Excessive Somnolence; GI, gastrointestinal; Med care, medical care; QI-Disability, Quality of Life Inventory-Disability.

[a]Eye Contact Avoidance Scale.

[b]Participation and Environment Measure for Children.

[c]Kessler Psychological Distress Scale.

[d]Beach Center Family Quality of Life Scale.

Total QoL score

The mean total QI-Disability score was 69.2 and the final tree is shown in Figure 1. The first split, corresponding to the variable which explains most of the variance in QoL, was based on the degree of daytime sleepiness (DOES), with high scores (≥74) associated with poorer QoL. Further splits were based on the frequency of participation in community activities and the levels of eye contact while speaking and listening. The terminal subgroup or leaf with the highest QoL scores (*n*=91, mean 80.3) comprised children with little daytime sleepiness who participated more frequently in community activities (Participation and Environment Measure for Children and Youth scale >2.1) and also displayed good eye contact while listening (Eye Contact Avoidance scale ≥8). Subgroups with the lowest QoL scores were those with a high degree of daytime sleepiness (*n*=74, mean 57.5) and those with less sleepiness but a lower level of participation and eye contact (Eye Contact Avoidance Scale <11) while speaking (*n*=163, mean 65.8). The most important variables used in assembling the tree are shown in Figure 2. The composition of the terminal subgroups (leaves) according to diagnostic category, needs dependence category, and communication category are shown in Table 2.

Negative emotions

The mean score for this subdomain was 65.3 and the final tree is shown in Figure 3. The first split was based on the degree of insomnia (Disorders of Initiating and Maintaining Sleep Scale) with high scores (>93) associated with poorer QoL. Further splits were based on the level of maternal

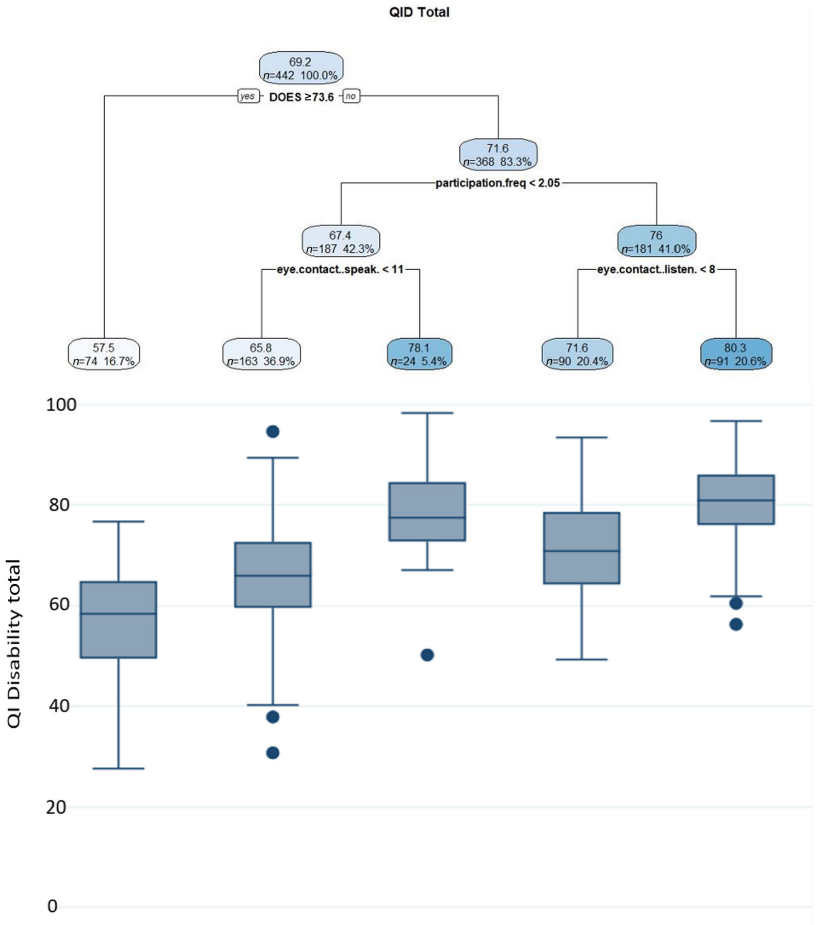

FIGURE 1 Regression tree for Quality of Life Inventory-Disability (QI-Disability) total score showing mean value and *n* (%) for each node. Boxplots show distribution of scores within each terminal node. The boxes show, from bottom to top, the 25th centile, median, and 75th centile values, and the error bars indicate the lower and upper adjacent values. The upper adjacent value is defined as the largest data point ≤75th centile +1.5 interquartile range (IQR), and the lower adjacent value is defined as the smallest data point ≥25th centile – 1.5 IQR. The outside values, which are data points more extreme than the upper and lower adjacent values, are individually plotted. DOES, Disorders of Excessive Somnolence

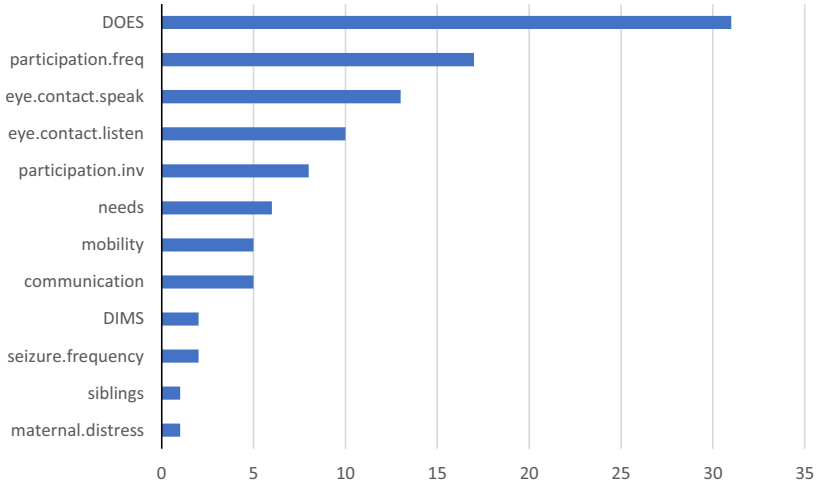

FIGURE 2 Variable importance. Indicates the variance reduction contribution of variables in generating the regression tree (before pruning) for total Quality of Life Inventory-Disability score. x-axis values are scaled to sum to 100. DOES, Disorders of Excessive Somnolence; DIMS, Disorders of Initiating and Maintaining Sleep

TABLE 2 Composition of the terminal subgroups (leaves) for the total quality of life score regression tree, *n* (%) by diagnostic, needs dependence, and communication categories

		Diagnosis				
Leaf number	**Mean quality of life score**	**Autism spectrum disorder**	**Cerebral palsy**	**Down syndrome**		**Rett syndrome**
1	57.5	18 (13.9)	28 (18.5)	6 (6.7)		22 (30.6)
2	65.8	52 (40.0)	64 (42.4)	21 (23.6)		26 (36.1)
3	78.1	1 (0.8)	16 (10.6)	4 (4.5)		3 (4.2)
4	71.6	41 (31.5)	25 (16.6)	17 (19.1)		7 (9.7)
5	80.3	18 (13.9)	18 (11.9)	41 (46.1)		14 (19.4)
		Needs dependence				
		Fully independent	Largely independent	Assistance required		Fully dependent
1	57.5	1 (7.1)	7 (8.1)	11 (9.8)		55 (24.0)
2	65.8	4 (28.6)	18 (20.7)	36 (32.1)		105 (45.9)
3	78.1	1 (7.1)	2 (2.3)	3 (2.7)		18 (7.9)
4	71.6	3 (21.4)	25 (28.7)	34 (30.4)		28 (12.2)
5	80.3	5 (35.7)	35 (40.2)	28 (25.0)		23 (10.0)
		Communication				
		Speaks well	Speaks with some difficulty	Some words only	Non-verbal only	Unable to communicate
1	57.5	3 (6.5)	11 (8.3)	12 (14.0)	32 (27.1)	16 (26.7)
2	65.8	13 (28.3)	32 (24.2)	35 (40.7)	48 (40.7)	35 (58.3)
3	78.1	4 (8.7)	5 (3.8)	2 (2.3)	11 (9.3)	2 (3.3)
4	71.6	13 (28.3)	41 (31.1)	15 (17.4)	15 (12.7)	6 (10.0)
5	80.3	13 (28.3)	43 (32.6)	22 (25.6)	12 (10.2)	1 (1.7)

distress and presence of a child mental health diagnosis. The leaf with the highest score (*n*=197, mean 74.0) included those without serious insomnia and with lower levels of maternal distress (Kessler Psychological Distress Scale <20). Leaves with the lowest scores included children with serious insomnia (*n*=73, mean 50.1) and those without insomnia but high levels of maternal distress and a mental health diagnosis (*n*=35, men 49.0).

Social interactions

The mean score for this subdomain was 70.5. The final tree, consisting of only one split, is shown in Figure 3. A higher level of eye contact while listening (Eye Contact Avoidance Scale ≥6) divided the sample into a subgroup with higher-than-average social interactions scores (*n*=328, mean 75.0) compared to a subgroup with substantially lower scores (*n*=114, mean 57.5). Further splitting did not improve the predictive ability of the tree.

Independence

The mean score for the independence subdomain was 61.9 with the final tree shown in Figure 3. The primary split was based on dependence for personal needs and a further split was based on communication ability. The leaf with the highest scores on this subdomain consisted of children displaying some level of independence (*n*=213, mean 78.3). The lowest independence scores were displayed by children totally dependent for their needs and with no communication, either verbal or non-verbal (*n*=57, mean 30.4).

Physical health

The mean physical health subdomain score was 70.1. The final tree comprised only one split (Figure 3). A low level of daytime sleepiness (DOES <59.3) divided the sample into a subgroup with higher-than-average physical health scores (*n*=254, mean 76.0) compared to a subgroup with lower scores (*n*=188, mean 62.2). Further splitting did not improve the predictive ability of the tree.

Leisure and the outdoors

The mean overall score for the leisure subdomain was 70.4 with the final tree shown in Figure 3. The primary split was again based on daytime sleepiness with a further split

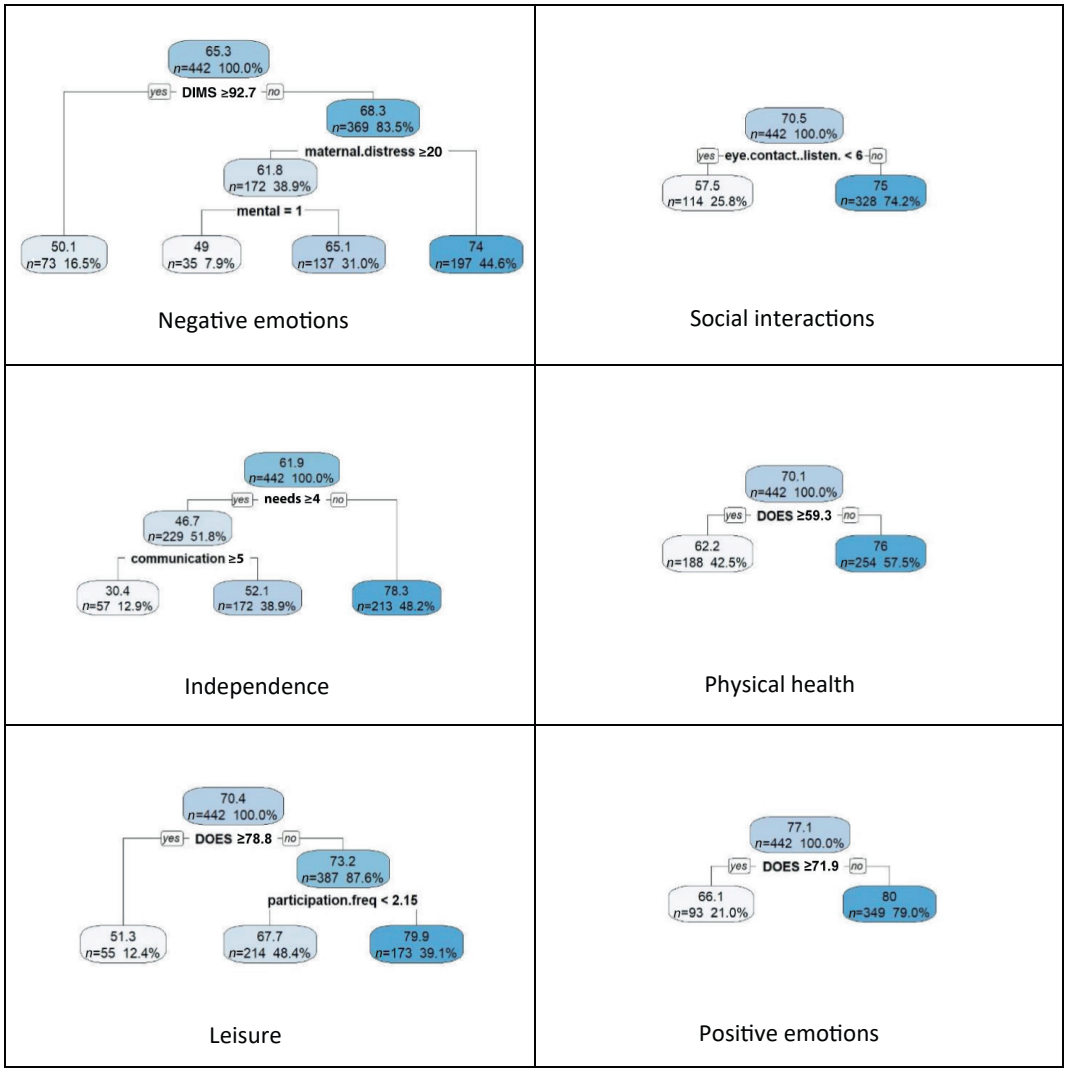

FIGURE 3 Regression trees for the Quality of Life Inventory-Disability (QI-Disability) subdomains showing mean score and *n* (%) for each node. DIMS, Disorders of Initiating and Maintaining Sleep; DOES, Disorders of Excessive Somnolence

based on frequency of community participation. The leaf with the highest scores on this subdomain (*n*=173, mean 79.9) consisted of children experiencing a lower level of sleepiness (DOES <78.8) and a higher level of community participation (Participation and Environment Measure for Children and Youth ≥2.15). The lowest leisure scores were displayed by children with a high level of sleepiness (*n*=55, mean 51.3).

Positive emotions

The mean positive emotions subdomain score was 77.1. The final tree consisted of only one split (Figure 3). A lower level of daytime sleepiness (DOES <71.9) divided the sample into a subgroup with higher-than-average positive emotions scores (*n*=349, mean 76.0) compared to a subgroup with lower scores (*n*=93, mean 66.1). Further splitting did not improve the predictive ability of the tree.

DISCUSSION

We previously identified that community participation[4] and sleep dysfunction[5] were potentially modifiable determinants of QoL in children with intellectual disability, using multivariate models and accounting for relevant confounders. Using data-driven CART analysis methods and a very large number of predictor variables beyond those included in our previous hypothesis-driven models, we again found that sleep dysfunction, particularly daytime sleepiness, and participation in the community were important factors associated with the child's QoL. Our results again suggest that worse impairments (notably sleep problems) and poorer community participation were associated with poorer QoL. These factors indicate pathways that can be exploited to improve QoL.

The range of diagnoses in our sample captures the common sleep problems experienced by children with intellectual disability in different neurodevelopmental conditions.[18]

The majority (60%–86%) of children with autism spectrum disorder are vulnerable to insomnia,[19] and 23% to 44% with cerebral palsy are vulnerable to insomnia, daytime sleepiness, or sleep-related breathing disorders.[20] The majority with Rett syndrome live with sleep problems, notably insomnia,[21] and more than half of children with Down syndrome have sleep-related breathing or other sleep disorders.[22] In our sample, daytime sleepiness was the most important factor to account for variance in QoL scores and also featured as a prominent factor associated with the physical health, positive emotions, and leisure and the outdoors domain scores. This finding resonates with known adverse consequences of poor sleep for daytime functioning.[23] While we cannot prove a causal link between insomnia and daytime sleepiness, 73% of those with abnormally high levels of daytime sleepiness in our sample also had abnormal insomnia levels. Treatments to reduce insomnia could also have the effect of reducing daytime sleepiness. We note consistency with a recent CART analysis that evaluated the roles of comorbid health conditions, functioning, and demographics on the QoL of adults with autism where sleep dysfunction was a vital factor in reducing QoL.[24]

The leaf node with the lowest total QoL scores (n=74) comprised those with abnormal levels of sleepiness. No further splits were found which would correspond to variables associated with improved QoL among this subgroup. We could hypothesize that excess daytime sleepiness was a proxy for clinical severity. However, only 24% of children who were totally dependent for their personal needs and 27% of children who were unable to communicate were members of this subgroup. In our combined analysis, the terminal nodes with the lowest and highest mean scores contained children from each diagnostic group, providing evidence that there are common factors which influence QoL in children with intellectual disability regardless of their specific diagnosis and clinical severity.

Frequency of participation was identified as the second most important variable for overall QoL and the most important variable amongst those children with normal levels of daytime sleepiness. Children with intellectual disability frequently live with social isolation and disconnectedness from peers[25] and our findings underline the importance of considering the social environment for opportunities in which children with intellectual disability can engage, collaborate, and prosper.[26] Also consistent with our multivariate models,[4] CART analysis indicated that eye contact while speaking and listening were the third and fourth in variable importance for the total QoL score, and eye contact while listening was also important for the social interactions subdomain analysis. Potentially, these skills provide the child with greater opportunity for social interactions and strategies to modify caregiver communication styles around the child could facilitate this component of QoL. In disability, support and environmental factors can improve QoL and counter the negative effects of difficulties. This is known as the 'disability paradox'.[27] Poor functioning and participation should not equate to poor QoL; rather interventions are needed to mitigate their effects where they are modifiable.

Other factors were relevant to the negative emotions and independence domains. For example, child insomnia was the most important variable and maternal distress the second most important variable associated with the negative emotions domain. This subdomain comprises items describing behaviours relating to aggression, anxiety, and agitation which could be exacerbated by lack of sleep. It is difficult to interpret the importance of maternal distress within this domain because a causal association could feasibly operate in either direction. Our data nevertheless suggest that prioritizing behavioural and mental health interventions for children and parents to promote positive behaviours and reduce distress is indicated. Independence in daily activities and communication abilities accounted for most of the variability in the independence domain. Rapidly expanding access to augmentative and alternative technologies has potential to improve the child's capacity for social communication, and choice and control.[28]

Children with intellectual disability have poorer QoL compared with typically developing children.[29] In response, we have searched a comprehensive set of predictor variables to identify modifiable determinants of QoL and redress this imbalance. This is a novel approach in the paediatric QoL field. Other strengths of this study include using a validated measure of QoL and recruiting a large sample representing a wide spectrum of clinical and disability issues.

We acknowledge some limitations. As is the case with any cross-sectional observational study, inferences cannot be made about causal processes. QoL was determined based on caregiver report because self-report on complex topics including the many things that are important to them would not be feasible for many of our participating children. While self-report is the criterion standard, we suggest that using proxy-report for QI-Disability is supported by its development processes where we only coded qualitative data that was observable to create items to reduce the subjectivity of the measure as far as is possible and avoid a parent interpreting how the child feels.[3] QI-Disability does not measure functioning or impairments but what is observed when life is going well or when life is challenging, and so a child with very impaired functioning could still score well on QoL. As expected, the domains of QI-Disability are consistent with the ICF as we have published when examining the content validity of QI-Disability.[30] The items do not equate to the ICF elements but a further qualitative study mapping QI-Disability items to groups of ICF elements and contextual factors would expand capacity to interpret QoL scores. There is also recent evidence that parent psychological distress does not mediate or moderate the relationship between functioning and QoL in children with intellectual disability.[31] Ideally, a regression tree should be validated by testing on an independent sample. Our sample size was not large enough to justify splitting into two independent samples, but it was sufficiently large to enable the growth of trees which had significant predictive ability after cross-validation.

We conclude that a data-driven method such as CART is a useful adjunct to the more traditional hypothesis-based methods to provide insights into the relative importance of associated factors. Specifically, we highlight the importance of sleep problems, in particular daytime sleepiness, community participation, and how children use eye contact during social interactions as important factors associated with QoL. The primacy of sleep problems and community participation in accounting for differences in QoL is encouraging. Little improvement in functional abilities such as mobility and communication can, at present, be achieved for these children but our findings indicate that these factors are secondary and do not prevent children with intellectual disability from enjoying good QoL. Insomnia is potentially modifiable. Clinicians need to be aware of the problem, remember to enquire about it, and make a careful diagnosis of the cause, to identify whether sleep quality is poor because of insomnia, sleepiness, or sleep breathing disorder. Management can include education on healthy sleep practice strategies or medication.[18] Increasing community participation is feasible and essential when building inclusive communities, offering opportunities for the child to connect with others, develop independence, and engage in meaningful activities.[32] We note that there is no high-level evidence for how to improve sleep and community participation in children with intellectual disability despite their influences on the child's QoL. We propose that these pathways indicate needs for trials of new treatments and supports for these targets, and that QoL would be an appropriate outcome measure.

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DATA AVAILABILITY STATEMENT

Data available on request from the authors.

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