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Patient- and parent-reported outcome measures of developmental adaptive abilities in visually impaired children: The Visual Impairment Developmental Autonomy (VIDA) scale

Serena Grumi^{b,1}, Federica Morelli^{a,c,1}, Eleonora Mascherpa^c, Francesco Decortes^c, Antonella Luparia^c, Livio Provenzi^{a,b,*}, Sabrina Signorini^c, VIDA group²

^a Department of Brain and Behavioral Sciences, University of Pavia, Pavia, Italy

^b Developmental Psychobiology Lab, IRCCS Mondino Foundation, Pavia, Italy

^c Developmental Neuro-Ophthalmology Unit, IRCCS Mondino Foundation, Pavia, Italy

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ABSTRACT

In the pediatric context, parents' and patients' engagement in the care process is strongly recommended and could be pursued using patient-reported outcome measures (PROMs), which therefore become useful for planning and monitoring treatments. Nevertheless, few data are available from families of children with neurodevelopmental disorders such as visual impairment (VI). The Visual Impairment Developmental Autonomy (VIDA) project aims to develop and validate a patient- and parent-reported tool to measure the most relevant aspects concerning everyday adaptive abilities in children and adolescents with visual impairment: the VIDA scale. The present paper illustrates the Delphi process of item generation engaging parents and patients and presents a protocol for the validation of this new co-designed tool in an Italian visually impaired pediatric population. Twenty-three families and five adolescents provided a list of 192 items and assessed their relevance. Items were categorized in 5 areas of adaptive abilities (i.e., table manners, clothing, personal hygiene, orientation and mobility, and socio-affectivity) and into three age ranges based on the patient's age. The final 102-item VIDA Scale will be administered to a minimum of 300 visually impaired children together with measures of quality of life and child adjustment to investigate its psychometric properties.

1. Introduction

Over the last few years, there has been increasing interest in the use of patient-reported outcome measures (PROMs) (Black, 2013; Field et al., 2019; Greenhalgh et al., 2018; Weldring & Smith, 2013). Healthcare professionals have been encouraged to cooperate with patients and caregivers in the development and validation of tools to measure the efficacy and effectiveness of treatments, also in rehabilitation settings and pediatric populations (De Rosi et al., 2020; Minneci & Deans, 2020; Soyiri et al., 2016; Tadić et al., 2020). PROMs help clinicians and healthcare managers to gather information on intervention priorities specifically related to the health and

* Correspondence to: University of Pavia, via Mondino 2, 27100 Pavia, Italy.

E-mail address: livio.provenzi@unipv.it (L. Provenzi).

¹ Co-first authors

² Members of the VIDA Group: Giorgia Aprile^b, Daria Painsi^b, Eleonora Perotto^b, Elena Saligari^b

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quality of life of patients and caregivers. In the pediatric context, parents' participation in the care process is strongly appreciated and could be pursued using PROMs, which are useful for planning and monitoring treatments (Barello et al., 2019; Barello et al., 2015; Edbrooke-Childs et al., 2016; Fayed et al., 2020). Nevertheless, while the importance of PROMs in adult populations with chronic diseases is widely recognized and studied (Cella et al., 2010; Rothrock et al., 2010), fewer data are available from families of children with neurodevelopment disorders and especially for children with visual impairment (VI).

The annual cumulative incidence of severe VI/blindness in children under 15 years of age is estimated at around 6/10.000, with highest values of incidence in the first year of life and an estimated 19 million visually impaired children below the age of 15 years worldwide (Solebo & Rahi, 2014; World Health Organization, 2017). VI, especially when congenital or early acquired, may negatively affect multiple aspects of child's development (Bathelt et al., 2019; Bathelt et al., 2018; Campus et al., 2021; Dale and Sonksen, 2002; Levtzion-Korach et al., 2000; Martolini et al., 2020; Morelli et al., 2020; Reynell, 1978), with possible long-term implications in such domains as social inclusion, academic performances, work inclusion, and quality of life (Greenaway et al., 2017; Rainey et al., 2016; Shah et al., 2020). Therefore, considering also that many VI causes are not preventable nor curable (Tadić et al., 2020), these children and their families need continuative and personalized care to support growth process and promote autonomy and social inclusion (Elsman et al., 2019; Morelli et al., 2020). Current re-habilitation approaches for VI promote functional and effective use of residual vision and other sensorialities, such as sound and touch, to sustain development, social inclusion and quality of life (Altunbay & İdil, 2019; Morelli et al., 2020; Tadić et al., 2020). In agreement with ICF-CY guidelines (WHO, 2007), such an approach relies on a multidisciplinary work, performed by different professionals such as doctors, neuropsychomotor and occupational therapists, psychologists (Morelli et al., 2020; Rainey et al., 2014).

Despite the extensive literature on developmental setbacks and needs in children with VI, normative data on specific developmental patterns in blind or severely visually impaired children are lacking (Vervloed et al., 2020). Furthermore, while several tests exist to assess *visual functions*, such as visual acuity, in children (Huurneman & Boonstra, 2016; Hyvarinen et al., 1980), providing information on how the eye functions, fewer tools are available to assess *functional vision*, i.e., how the person functions. Functional vision is related to the abilities of a person, concerning various domains, such as recognition, activities of daily living, and orientation and mobility, thus being strictly connected to social participation and quality of life (Colenbrander, 2010). Consequently, sustaining functional vision would be fundamental in the management of visually impaired children, in order to support quality of life and psychosocial aspects (Barker et al., 2015; Dahlmann-Noor et al., 2017; Tailor et al., 2017). In general, few tools are available to measure re-habilitation outcomes in visually impaired children and adolescents, especially concerning adaptive abilities (Elsman et al., 2019), and the available tools have not been co-developed in a PROM perspective by professionals and caregivers (Bevans et al., 2010; Matza et al., 2013), except for the questionnaire on the quality of life developed by Tadić and colleagues (Tadić et al., 2020). The development of tools to evaluate, plan, and monitor an intervention based on children and their families' care needs is a priority to optimize the healthcare system and provide early interventions answering these families' actual needs.

The Visual Impairment Developmental Autonomy (VIDA) project aims to develop and validate a patient- and parent-reported tool to measure the most relevant aspects concerning everyday adaptive abilities in children and adolescents with visual impairment: the

Table 1
Subjects' characteristics.

Subject	Sex	Age	Age group	Diagnosis	Low vision severity [#]
V03 *	male	17	adolescent	ONH	Severe low vision
V04 *	male	14	adolescent	ONH	Blind
V05	female	14	adolescent	IRD	Blind
V07	male	12	adolescent	IRD	Blind
V14	female	17	adolescent	IRD	Blind
V15 [§]	female	17	adolescent	RP	Mild low vision
V16 *	male	14	adolescent	IRD	Blind
V23	male	15	adolescent	IRD	Blind
V40 *	female	18	adolescent	IRD	Blind
V10	male	8	school age	IRD	Blind
V13	male	9	school age	IRD	Severe low vision
V19	female	8	school age	IRD	Blind
V24	female	9	school age	eye maldevelopment	Blind
V25	male	6	school age	ROP	Blind
V35	female	10	school age	eye maldevelopment	Blind
V39	female	8	school age	IRD	Blind
V02	female	4	pre-school	IRD	Severe low vision
V06	female	5	pre-school	IRD	Severe low vision
V12	female	5	pre-school	IRD	Blind
V18	male	5	pre-school	congenital glaucoma	Severe low vision
V20	female	4	pre-school	IRD	Blind
V27	male	4	pre-school	IRD	Blind
V36	female	5	pre-school	IRD	Blind
V38	male	5	pre-school	IRD	Blind

Note. ONH: Optic Nerve Hypoplasia; IRD: Inherited Retinal Dystrophy; RP: Retinitis Pigmentosa; ROP: Retinopathy of Prematurity. *both subject and parent participation [§] subject participation only. [#] Visual acuity tested at a 3 m distance. Severe low vision: visual acuity $\leq 1/10$. Blind: visual acuity $\leq 0.05/10$.

VIDA scale. In more specific terms, the present paper [a] illustrates the process of item generation engaging parents and adolescents with visual impairment through a Delphi approach and [b] presents a validation protocol of this new co-designed tool in an Italian population of VI children and adolescents.

2. Methods, Part 1: Vida co-design

2.1. Participants

Twenty-nine families and ten adolescents referring to the Child Neuro-Ophthalmology Unit of the IRCCS Mondino Foundation, Pavia (Italy), were invited to take part to the co-design of the VIDA scale.

We included in the study families of children affected by congenital peripheral VI (i.e., a visual deficit due to the involvement of pre-chiasmatic structures, such as retina and optic nerve) without primary brain involvement (as in Cerebral Visual Impairment) or significant comorbidities. Visual acuity (VA) was measured at a 3 m distance using the Snellen optometric charts (Azzam & Ronquillo, 2020) or the LEA vision test (Hyvärinen et al., 1980), consisting respectively in lines of letters or symbols whose size decreases on each lower line of the chart. The test was chosen according to the age of the subject. According to the Italian Law, low vision was classified as mild (> 2 and $\leq 3/10$), moderate (> 1 and $\leq 2/10$), severe (> 0.05 and $\leq 1/10$), blindness ($\leq 0.05/10$) (G.U. Serie Generale, n. 93 del 21 Aprile 2001). The final sample was composed of 8 pre-school children (age 3–6 years of age), 7 children (6–11 years of age), and 9 adolescents (11–18 years of age). 23 parents and 5 adolescents participated filling the questionnaires of the three rounds. The mean value of socio-economic status of participants using the Hollingshead index (Hollingshead, 1975) was 31.94 (SD 14.43, 8–63), with 62.5 % of subjects were in the ‘low’ range (8–29), 12.5 % in the ‘middle’ range (30–39), and 25 % in the ‘high’ range (40–54). Clinical characteristics of patients are summarized in Table 1.

2.2. Procedures

The VIDA scale co-design included three rounds based on the Delphi approach. The Delphi technique is a process traditionally used to reach a group consensus about issues without definite evidence by surveying a panel of experts (Dalkey & Helmer, 1963; Thangaratinam & Redman, 2005; Vernon & Vernon, 2009). The basic principles of this method are: (a) the selection of a group of experts, called ‘panelists’, who is questioned about the issue of interest. In our case, experts were patients and their families (b) the process is anonymous in order to avoid pressure and conformity to a dominant view; (c) the procedure is iterative in its nature, comprising several rounds of enquiry; and (d) the design of subsequent rounds is informed by a summary of the group response of the previous round (Diamond et al., 2014; Hasson et al., 2000; Jünger et al., 2017; McKenna, 1994; Thangaratinam & Redman, 2005). The round questionnaires are gradually more specific and the answers from the previous round are used to structure the questionnaire of the next one (Powell, 2003; Vernon & Vernon, 2009).

The Delphi Study was performed according to published guidelines in conducting and reporting Delphi Studies (Diamond et al., 2014; Hasson et al., 2000; Jünger et al., 2017; McKenna, 1994). The aim of our study was the creation of a new patient- and

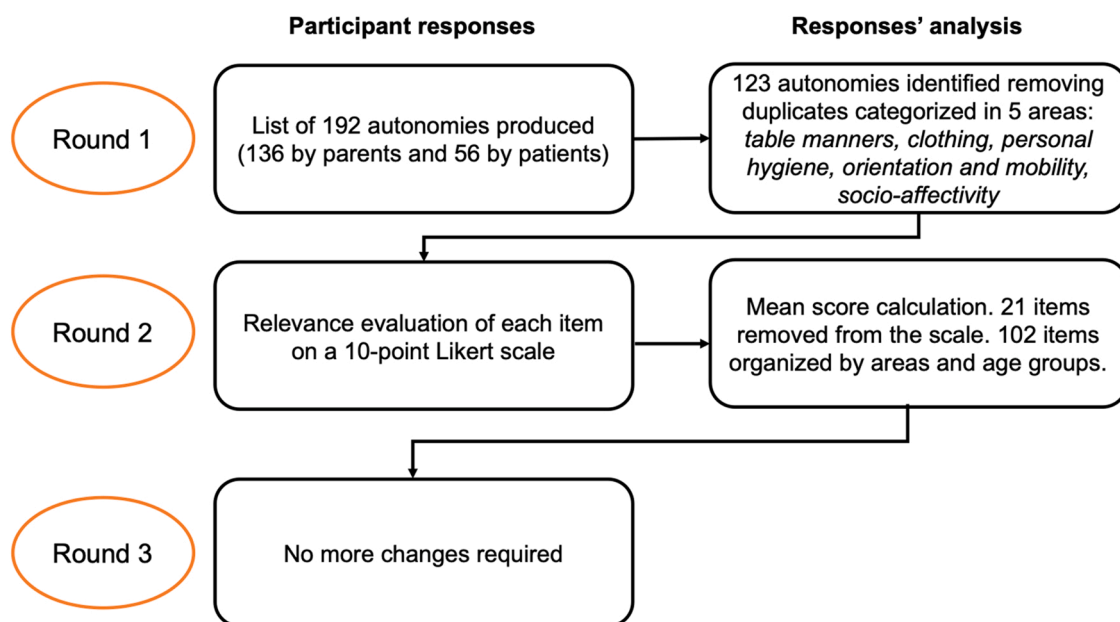


Fig. 1. Development of the patient- parent-reported VIDA scale using the Delphi process.

Table 2

Patient- and parent-reported outcome measure of developmental adaptive abilities in visually impaired children (VIDA scale).

A AREA - Table manners

Score: 2 = performs without help 1 = performs with help 0 = does not perform

3 – 6 years	He/she drinks from the glass He/she finds objects on the table He/she uses the fork He/she pours liquids He/she help himself/herself with both hands in collecting food (e.g., one hand holds the fork and the other helps in the search when he finds nothing) He/she takes part in the meal without getting distracted or moving excessively He/she takes part in the meal without talking excessively
6 – 11 years	He/she uses the knife He/she uses the spoon He/she is able to take any type of food with cutlery without touching it with hands He/she seats at table adequately He/she recognizes what he/she is eating He/she manages to eat all the dish's content He/she wipes his/her hands and mouth in the napkin during and after meals He/she actively participates in the conversation during meals He/she helps to set and clean up the table
11 – 18 years	He/she is able to eat without dropping food off the plate He/she is able to stir He/she is able to serve himself/herself from the refrigerator He/she is able to make a sandwich He/she collaborates in the preparation of dishes (i.e., transformation of foods)

B AREA - Clothing

3 – 6 years	He/she is able to fasten a jacket with press studs or with the ends already fixed He/she is able to take off the pants He/she is able to take off the shirt He/she puts on the shoes without tying them or puts on velcro shoes He/she is able to take his/her pajamas from under the pillow himself/herself He/she hangs his/her jacket/scarf/hat on the hanger
6 – 11 years	He/she puts on his/her socks He/she puts on his/her pants He/she puts on his/her shirts He/she manages to close zippers and buttons He/she finds what he/she is looking for in the closet He/she ties the shoelaces He/she can wear clothes the right way
11 – 18 years	He/she manages to wear all the clothing items in complete autonomy He/she chooses and matches the clothes to wear, also according to the season He/she folds clothes properly He/she tidies up his/her closet

C AREA – Personal hygiene

3 – 6 years	He/she opens and closes the tap He/she washes his/her hands with soap He/she dries his/her hands after washing them He/she can use the toilet by himself/herself He/she washes face and eyes He/she brushes his/her teeth
6 – 11 years	He/she takes a shower or bath He/she brushes his/her teeth (putting the toothpaste on the toothbrush and closing the tube) He/she cleans up after he/she's done his/her needs He/she washes his/her private parts He/she washes and dries his/her feet He/she washes his/her glasses He/she combs his/her hair
11 – 18 years	He/she washes taking a reasonable amount of time He/she washes his/her hair himself He/she distinguishes the various bathroom products He/she takes care of his/her personal hygiene He/she uses the hair dryer and manages to style his/her hair He/she takes a shower or bath alone, dosing the shampoo and shower gel correctly He/she manages how to shave/wear makeup

D AREA – Orientation and mobility

3 – 6 years	He/she spontaneously participates in simple games with peers He/she finds someone who calls him/her He/she senses the presence of an obstacle He/she finds objects in his/her house He/she calls a friend/schoolmate to get help in moving and playing He/she plays simple motor games
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(continued on next page)

Table 2 (continued)

6 – 11 years	He/she manages to carry out a short-known path (e.g., at the kindergarten) He/she is able to move in sudden and new situations He/she orientates himself/herself in a noisy environment He/she manages to go up and down the stairs autonomously He/she manages to carry out a known path (e.g., moves between different classrooms at school, home-school trip) He/she recognizes faces and voices He/she recognizes landmarks in a small center
11 – 18 years	He/she practices sports He/she makes short path in unknown environments He/she practices group sports He/she uses the stick to move independently He/she crosses the street He/she takes public transports He/she goes out with his/her peers/friends independently
E AREA – Socio-affectivity	
3–6 years	He/she exhibits behaviors to build friendships towards children of his/her own age He/she is aware of what he/she is able or unable to do and knows how to ask for help He/she asks to play with his/her schoolmates He/she doesn't react excessively in case of defeats or difficulties in facing a task He/she performs simple group games for a common goal He/she respects the rules of simple games He/she respects his/her turn in a game He/she recognizes the others' emotional state and modulates his own behavior He/she shares his/her toys with his/her friends/schoolmates
6 – 11 years	He/she makes the first move to play with peers He/she is integrated into the peer group He/she involves others in his/her own adapted games (when present) He/she is autonomous in carrying out his homework He/she he meets friends even outside school hours, with adult supervision He/she nurtures a passion
11 – 18 years	He/she uses the PC in a functional and autonomous way He/she spontaneously starts the conversation He/she does easy household chores He/she can have a conversation with an adult He/she relates to another person in whom he/she is romantically interested He/she cooperates with others in planning and implementing activities He/she knows how to verbalize his/her own experiences with respect to the situations he/she lives He/she meets friends regularly without adult supervision He/she takes part in trips and outings even lasting several days (more than one day)

parent-reported tool about adaptive behaviors in visually impaired children, therefore the consensus has been considered achieved when all participants approved the final version of the scale and requested no further changes.

Round 1. Between December 2020 and February 2021, a pool of parents and visually impaired adolescents was invited to participate in the design of an adaptive behavior scale for visually impaired children. Characteristics of parents and patients are summarized in the next paragraph. Parents and patients received a web-link by email with the request to fill out the first questionnaire consisting of open-ended questions. As per other adaptive behavior evaluation tools (Harrison & Oakland, 2015; Sparrow & Cicchetti, 1989), our scale was thought to be structured in different domains of independent living. Participants were asked to provide as many suggestions as they could about four domains of adaptive abilities in everyday living: *table manners, clothing, personal hygiene, orientation and mobility*. Moreover, participants were asked to provide suggestions about other relevant domains of adaptive abilities and to generate new items accordingly. Parents provided suggestions according to the age range of their children.

Analysis. The responses of the first round were analyzed qualitatively by unifying similar items and, if necessary, categorizing them in areas of adaptive abilities. Moreover, the listed abilities were organized also in three different groups of developmental stages based on the age of the patients (3–6 years; 6–11 years; 11–18 years).

Round 2. Participating parents and adolescents received a second survey containing the list of areas and adaptive abilities and were asked to evaluate the relevance of each item on a 10-point Likert scale according to their point of view. This part took place between March and April 2021.

Analysis. Responses were analyzed quantitatively and items with a mean score lower than 8 were excluded.

Round 3. Between May and June 2021, participants received the last version of the scale with the request to validate it or suggest any changes (e.g., moving items from a developmental stage to another one, removing items, or adding new items).

Analysis. Researchers defined the final version of the scale according to participants' comments.

3. Results

As summarized in Fig. 1, during the first round of the scale co-design parents and adolescents provided 136 and 56 new items respectively. The items provided by parents and adolescents were qualitatively analyzed together to identify redundant adaptive behaviors. During the analysis, 69 items were excluded for redundancy. A sub-group of the new items was clustered in the area socio-

affectivity. As such, the new version included 5 domains of independent living: *table manners, clothing, personal hygiene, orientation and mobility, and socio-affectivity*. Moreover, for each area items were organized into three ranges of age (3–6 years, 6–11 years, and 11–18 years) based on the patients' age. During the second round, parents and adolescents rated on a 10-point Likert scale the relevance of each item and the 21 items with a mean score lower than 8 were excluded. During the third round no more changes were required by participants. The final version of the scale is reported in [Table 2](#).

4. Methods, Part 2: validation protocol

4.1. Enrollment

Visually impaired children and their parents will be enrolled at the Child Neuro-Ophthalmology Unit of the IRCCS Mondino Foundation, Pavia (Italy). They will be enrolled consecutively according to the following inclusion criteria: child's age between 3 and 18 years; parental mastery of Italian language; peripheral (i.e., involving pre-geniculate structures and pathways) visual deficit resulting in low vision (defined as a visual acuity $\leq 3/10$ at a 3 m distance measured with a standardized test, below the threshold defined by the Italian legislative system). Exclusion criteria include a diagnosis of developmental delay and/or intellectual disability (i.e., intellectual or development quotient <70 as per standardized evaluations such as Wechsler, for which verbal subtests will be considered in case of severe visual impairment/blindness, Griffiths, Bayley, or Reynell-Zinkin Scales) and the presence of neuromotor disorder, central nervous system involvement or chronic comorbidities.

4.2. Procedures

The co-designed VIDA scale will be administered to a group of visually impaired children stratified by age together with a scale about the quality of life (Preschool Children's Quality of Life questionnaire or Children's Quality of Life questionnaire on the basis of child's age; Vision-Related Quality of Life Instrument for Children and Young People with Visual Impairment for children over 8 years old) in order to test the convergent validity. Parents will be invited to complete the Child Behavior Checklist (CBCL), while adolescents will complete the Youth Self Report ([Achenbach & Rescorla, 2001](#)) to investigate the association between developmental adaptive abilities and child adjustment.

A factorial analysis and the internal consistency of the VIDA scale will be computed to obtain psychometric characteristics of the instrument. The concurrent validity will be assessed through the level of correlation between the VIDA scale's completion by the patient/parent and by the therapist.

4.3. Measures

Quality of life. On the basis of the child's age, participants will complete the *Children's Quality of Life questionnaire* (TACQOL; [Vogels et al., 1998](#)) or the *Preschool Children's Quality of Life questionnaire* (TAPQOL; [Fekkes et al., 2000](#)). The TAPQOL is a 43-item questionnaire that measures the parent's perceptions of health-related quality of life in preschool children. It consists of 12 scales that cover four domains: physical functioning, social functioning, cognitive functioning and emotional functioning. The 56-item TACQOL cover seven domains of health-related quality of life: physical complaints, motor functioning, autonomous functioning, cognitive functioning, social functioning, positive moods, and negative moods. Both the parent- and the child-form of the TACQOL will be administered. Children over 8 years old will fill-in also the Vision-Related Quality of Life Instrument for Children and Young People with Visual Impairment (VQoL_CYP; [Tadić et al., 2020](#)). The PROM VQoL-CYP is a developmentally sensitive measure of vision-related quality of life. Two age-specific extensions were adapted for children (8–12 years; 20 items) and young people (13–17 years; 22 items).

Child adjustment. The *Child Behavior Checklist* (CBCL; [Achenbach & Edelbrock, 1991](#); [Achenbach & Rescorla, 2001](#)) is an extensively used questionnaire to assess children's adjustment. The respondent (usually a parent or another caregiver) is asked to estimate the degree to which his or her child exhibits a set of problem behaviors using a 3-point Likert scale ranging from 0 (not true) to 2 (very or often true). It provides scores for internalizing (e.g. withdrawn-depressed) and externalizing problems (e.g., aggressive behavior) and a total score on the child's overall psychological adjustment. Two equivalent versions of the scale depending on the age of the child are available: CBCL 1^{1/2}-5 (100 items) and CBCL 4-18 (113 items). Adolescents will be asked to complete the *Youth self-report* (YSR; [Achenbach & Rescorla, 2001](#)). The 119-item YSR is a self-report questionnaire developed to assess problems in youth ages 11–18, which is consistent and comparable to the parent-reported CBCL.

4.4. Sample size estimation

The minimum sample size was estimated based on [Boateng and colleagues \(2018\)](#) review of recommendations across the literature about developing and validating scales. Despite the ratio for the minimum sample size estimation having often been contentious, Boateng and colleagues identify the minimum sample size of 300 as the most shared criteria. Considering the specific characteristics of this clinical population, a multi-center study will be considered to reach the minimum sample size established. Moreover, the sample will be stratified by age according to the developmental stages included in the scale (i.e., 3–6 years; 6–11 years; 11–18 years).

4.5. Plan of analyses

Descriptive statistics (i.e., frequencies, means and standard deviations), Cronbach's alpha reliability (for each area and for the overall scale), factorial analysis will be performed as preliminary analyses to explore the psychometric characteristics of the scale. The Pearson's correlation between the VIDA and the TAPQOL or TACQOL scores will be tested to evaluate the convergent validity. The bivariate Pearson's correlation between the VIDA scores by parents and professionals will be performed to test the concurrent validity. To explore the association between developmental adaptive abilities and adjustment in visually impaired children bivariate Pearson's correlation indexes will be performed between the VIDA score and the CBCL or YSR subscales and total scores.

4.6. Ethics

The study has received the approval of the Ethics Committee of Pavia Area, Fondazione IRCCS Policlinico San Matteo, Pavia (Italy) on 5th June 2021, Protocol Number p-20200048762. All the procedures are consistent with the Declaration of Helsinki ethical principles for research involving human subjects. The procedures do not imply any harm to the participating subjects nor any change to rehabilitation programs at the Child Neuro-Ophthalmology Unit of the IRCCS Mondino Foundation, Pavia (Italy).

5. Discussion

5.1. Impact and implications

The VIDA project is supposed to develop and validate a questionnaire for the evaluation of adaptive skills in visually impaired children and adolescents. The development of such a tool in a PROM perspective, with the active engagement of patients and their families, could possibly fill a gap in the literature and availability of specific instruments to measure rehabilitation outcomes in VI. The VIDA questionnaire hold premises to be useful both in the clinical and in the research settings. Indeed, on one hand it will serve as an outcome measure for re-habilitation in visually impaired children; in this way, a periodic assessment using the questionnaires can be helpful for the therapist to constantly monitor and tailor his/her intervention. On the other hand, proposing the VIDA questionnaire to a vast population could shed a light on the adaptive skills acquisition trajectories in the visually impaired population. Furthermore, the questionnaire has the undoubtable advantage that it can be filled in both by the patient/parent and by the clinician. This peculiarity could provide useful insight on the differences (if present) between the perceived and the observed adaptive abilities and difficulties.

5.2. Limitations

First, only adolescents actively participated in the Delphi procedure as the type of data collection used did not allow the engagement of younger patients. Second, in the same way, the scale includes a patient version only for adolescents. Third, given the characteristics of this specific clinical population, it will be challenging reaching a huge sample size for the questionnaire validation. Fourth, the co-designed measure may be affected by cultural variables, therefore it may not be appropriate to use this scale in different cultural settings before an appropriate adaptation.

CRedit authorship contribution statement

Serena Grumi: Conceptualization, Methodology, Data curation, Writing – original draft. **Federica Morelli:** Conceptualization, Investigation, Data curation, Writing – original draft. **Eleonora Mascherpa:** Investigation, Data curation. **Francesco Decortes:** Writing – review & editing, Visualization. **Antonella Luparia:** Writing – review & editing, Visualization. **Livio Provenzi:** Conceptualization, Resources, Writing – review & editing, Visualization. **Sabrina Signorini:** Conceptualization, Resources, Writing – review & editing, Visualization.

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