Participation of children and youth with acquired brain injury

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6

Chapter 6
Family Impact of Acquired Brain Injury in children and youth

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ABSTRACT

Objective To assess the impact of Acquired Brain Injury (ABI) in children and youth on the family and explore factors associated with the extent of family impact.

Design Follow-up study.

Setting General hospital, rehabilitation care and the community.

Participants A cohort sample of parents of children and youth, 24-30 months after the diagnosis ABI was made in a general hospital. The inclusion criteria were age 4-20 years at onset of ABI. The patients of whom the parents were included in this study had an age range of 6-22.

Interventions Not applicable.

Main outcome measures Family impact was measured with the Pediatric Quality of Life Inventory Family Impact Module (PedsQL™FIM). Additional assessments included the PedsQL™General Core and Multiple Fatigue Scales, the Paediatric Stroke Outcome Measure Short Neuro Exam (PSOM-SNE), the Child & Family Follow-up Survey (CFFS) and sociodemographic and disease characteristics.

Results The parents of 108 patients participated in the study. The median age of the patients was 13 years (range 5-22), with 60 patients (56%) being male. The cause of ABI was traumatic (TBI) in 81 patients (75%) and non-traumatic in 27 patients (25%). At the time of diagnosis 19/81 (23%) and 5/27 (19%) were classified as moderate or severe in the TBI and NTBI groups, respectively.In the total group of patients with TBI and NTBI family impact was found to be associated with the severity and type of injury and the presence of child health problems before the ABI.

Conclusion Two years after ABI, the impact on the family as measured by the PedsQL™FIM was considerable. The extent of family impact was associated with characteristics of ABI as well as the health status of the child before ABI.

INTRODUCTION

Acquired brain injury (ABI) refers to any damage to the brain that occurs after birth, due to a traumatic (TBI) or non-traumatic (NTBI) cause. In children and youth the yearly incidence of ABI is substantial, with estimated incidence rates for the age group 0-24 years in the Netherlands being 585 per 100.000 for TBI and 190 per 100.000 for NTBI,[1] similar to incidence rates reported in the international literature.[2-4] Overall it is found that TBI may have a considerable impact on the patients' functioning[5,6] and quality of life[7-9] although the results regarding the impact of severity of TBI on quality of life were conflicting.

There are various studies reporting on the occurrence of problems in patients' functioning after NTBI in comparison with their healthy peers, such as in children with stroke[10,11] and brain tumours.[12] Moreover, the literature suggests that the long-term outcome is also related to family and environmental factors (including family cohesion, resources, social support, socioeconomic status).[13-17] Vice versa, family functioning can be influenced by the consequences of pediatric ABI, with negative effects on coping, problem-solving and communication of parents,[15,17,18] reflected by increased rates of family disruption, divorce and disfunctioning of brothers or sisters[19,20] after ABI. Although many families eventually adapt favorably to the often increased demands of the situation after injury, clinically significant stress was found in approximately 40% of families more than 12 months after onset of paediatric TBI.[2,21]

Regarding the factors related to the extent of family impact, injury severity, functional impairment, health problems, behavioral changes and emotional problems after ABI, were found to have a significant association to family functioning.[14,19,22-24]

So far, studies on family impact after ABI have only been done in the United States and Australia, and were primarily focused on TBI. In addition to a relatively large variety in inclusion criteria and time since onset of injury, these studies used various instruments to measure family impact. Specific measures for family burden or impact of trauma and/or pediatric chronic health conditions, include the Impact on Family Scale (IFS),[25] Parenting Stress Index Short Form (PSI/SF),[26] Family Burden of Injury[14,27] and The Family Impact Module (PedsQL™FIM) of the Paediatric Quality of Life Inventory Interview (FBII) (PedsQL™4.0).[28] The PedsQL™FIM appears to be a useful instrument, as it includes the physical, emotional, social and cognitive functioning of parents. These domains were found to be negatively influenced after paediatric ABI in the literature.[18,21,29,30] Moreover the PedsQL™FIM is available in multiple languages including Dutch, was designed as multidimensional measure of the impact of pediatric chronic health conditions. The PedsQL™FIM showed good psychometric properties in parents of children with complex chronic health problems[28] and cancer[31] and was used in studies on children with Duchenne,[32] a diversity of disabilities[33] and chronic pain.[34]

So far, the PedsQL™FIM has not been used in studies on the family impact of ABI. The aim of the present study was therefore to determine the impact of paediatric TBI and NTBI on families in the Netherlands, 24-30 months after diagnosis, using the PedsQL™FIM.
Secondary aim was to determine associations between sociodemographic characteristics (patient and family characteristics), ABI characteristics and actual functioning on the one hand, and the family impact as measured with the PedsQL™FIM on the other hand.

METHODS

Design and setting

This study on family impact was part of a larger, multicentre, hospital-based study on the incidence of ABI in the Netherlands.[1] In that study, performed in 2010, 1892 patients aged 0-24 years, with a first hospital-based diagnosis ABI made in 2008 or 2009, were identified by means of a review of the medical records of the emergency ward databases and the patient admission registries of 3 major hospitals in the Netherlands (Erasmus University Medical Centre in Rotterdam, Haga Hospital, The Hague and Medical Centre Haaglanden, The Hague). In a follow-up study we aimed to determine the health status approximately 2 years after onset of ABI. The present study on family impact is part of the follow-up study. The study (including the follow-up) was approved by the medical ethical committee (METC) of the Erasmus University Medical Centre Rotterdam (METC-2009-440). All parents and patients, as required by law from 18 years, participating in the follow-up assessment gave written informed consent.

Participants

For the larger study patients were selected from the registries of the participating hospitals using the following causes of injury for TBI: accident (e.g. in traffic, at home, in sports), (suspicion of) physical abuse and fall under influence of alcohol or drugs intoxication. For NTBI, the following causes were recorded: tumour, meningitis or encephalitis, stroke, ADEM (Acute Disseminated Encephalo Myelitis), MS (Multiple Sclerosis) or acute CNS (Central Nervous System) demyelinating disease and hypoxia-ischemia.
Patients were excluded if they were diagnosed with trauma capitis (minor head injury without brain symptoms). Inclusion criteria for the follow-up study were: age at onset ABI 4-20 years and ability to understand and complete questionnaires in Dutch. To select patients for the follow-up study in this article the total group of participants was categorized by age (4-12 or 13-20 years), year of onset (2008 or 2009), type (TBI or NTBI) and severity of injury (mild-moderate-severe), yielding 24 subgroups in total. Aiming at a total number of 400 patients to be invited for follow-up with a predicted response of 50%, 18-20 patients per subgroup

were selected. Within each subgroup, participants were at random selected using 'select cases, option select random sample of cases' in the statistical software program Statistical Package for Social Sciences (SPSS).[35] This procedure yielded a selection of 433 patients. These patients and/or their parents were subsequently approached by mail to participate in the study.

Assessments

Of all patients participating in the larger study, the age at onset, gender, year of onset, the type of injury (TBI or NTBI) and the severity had been extracted from the medical records. The severity of TBI was determined by means of the Glasgow Coma Scale (GCS) at hospital admission. According to the GCS, the severity of TBI was considered mild if the GCS was 13-15, moderate if the GCS was 9-12 or severe if the GCS was <9.[36] The severity of NTBI was determined at the time of discharge after the first admission to the hospital for this particular problem, and was scored by means of an adapted version of the modified paediatric Rankin Scale (mRS)[10,37] (school performance not taken into consideration): (1) Mild injury: no limitations (mRS 0, 1);(2) Moderate injury: mild motor impairments and/or mild problems with learning (mRS 2, 3); (3) Severe injury: severe motor impairments and/or severe problems with learning (mRS 4, 5). In addition, mRS 6 was used in cases of death during admission.

The questionnaires were in part completed at home and in part during the visit for the examination. Within 1-3 months after informed consent was given and in the week before the examination of the child in an outpatient rehabilitation clinic, parents received 4 questionnaires to be completed at home: the Child & Family Follow-up Survey (CFFS),[38] the Paediatric Quality of Life General Core Scale,[39] the PedsQL™FIM[28] and the PedsQL Multidimensional Fatigue Scale[40] (average duration 45 minutes). Subsequently, about 1 week later, the child was examined in an outpatient rehabilitation clinic. During the visit for the examination parents were interviewed by trained assessors. The structured interview included questions on the presence of physical and/or mental health problems of the parents before the ABI and/or at present (2 questions, yes/no) and/or the presence of physical and/or mental problems of the child before the ABI and/or at present (2 questions, yes/no) (4 questions in total, yes/no).

Family impact

The 36-item PedsQL™FIM yields a Parent Health Related Quality of Life (HRQoL) Summary Score (the Physical, Emotional, Social, and Cognitive Functioning Subscales; 20 items), a Family Functioning Summary Score (Daily Activities and Family Relationships Subscales; 8 items), Communication Subscale score (3 items) and a Worry Subscale score (5 items), as well as a Total Score. Higher subscale, summary and total scores indicate better functioning. If more than 50% of the items in the scale were missing, the Subcale Score was not

computed. Although there are other strategies for inputting missing values, this computation is consistent with the previous PedsQL™FIM peer-reviewed publications, as well as other well-established HRQOL measures.[41]

Overall functioning and fatigue

Two other modules of the Paediatric Quality of Life Inventory (PedsQL™4.0), pertaining to the child's health status, and both available in a Dutch language version, were used:

a. The General Core Scale,[39,42] which measures physical (8 items), emotional (5), social (5) and school functioning (5). In this study parent report versions for children 5-7, 8-12 and 13-18 years old were used.
b. The Multidimensional Fatigue Scale,[40] designed as a child self-report and parent proxy-report generic symptom-specific instrument to measure general fatigue (6 items), sleep (6) and cognitive fatigue (6) in children. The overall functioning and fatigue scores range from 0-100, with higher scores indicating better functioning). In this study the parent version was used.

Neurological functioning

The Paediatric Stroke Outcome Measure Short Neuro Exam (PSOM-SNE) was used for the neurological functioning,[43] It includes 5 areas of functioning: right sensorimotor, left sensorimotor, language production, language comprehension, and cognitive/behavioural. An overall Deficit Severity Score (DSS) of normal-mild-moderate-severe, as indicator of actual level of functioning is based on the combination of these scores, with a score range of 0-10. Lower scores indicate better functioning (less negative impact).

Participation and Environmental factors

The Child & Family Follow-up Survey (CFFS),[38,44] comprising the Child and Adolescent Scale of Participation (CASP), The Child and Adolescent Factors Inventory (CAFI) and the Child and Adolescent Scale of Environment (CASE) was used.

For both the CAFI and the CASE, higher scores indicate a greater number of problems, a greater impact of problems or a combination of the two.

Statistical analysis

Characteristics of patients and parents were analysed using descriptive statistics. All continuous variables were, according to their distribution, expressed as mean with standard deviation or median with score range.

Comparisons of sociodemographic and injury characteristics of participants in the present follow-up study as compared to those of all invited patients were done by means of the Mann-Whitney-U test.

To determine which factors were associated with family impact, the mean PedsQL™FIM Total Score, HRQoL and Family Functioning Summary Scales and the two Subscales Scores Communication and Worry were compared between subgroups of patients. Subgroups were made for the following variables: Characteristics before or at onset of ABI (sociodemographic: patient age and gender; educational level parents and single or double parent household; presence of health problems before ABI; injury characteristics: type, severity); functioning 2 years after onset of ABI (actual neurological functioning, activities and participation, fatigue, quality of life). For continuous variables, subgroups were made were made according to the mean (low/high). Comparisons of family impact scores between subgroups were done by means of independent t-tests for continuous variables or one way Analysis of Variance (ANOVA) for categorical variables.

Then, separate univariable models were used for each independent variable, again using the PedsQL™FIM Total Score, HRQoL and Family Functioning Summary Scales and the two Subscales Scores Communication and Worry as dependent variables.

Next, baseline characteristics (before or at onset of ABI) were fitted in linear multivariable regression models as independent or predictor variables, with the PedsQL™FIM Total Score as the dependent variable. The variables which were not significantly associated with this outcome were dropped from the model, after a stepwise check. Results were presented as regression coefficients and explained variance.

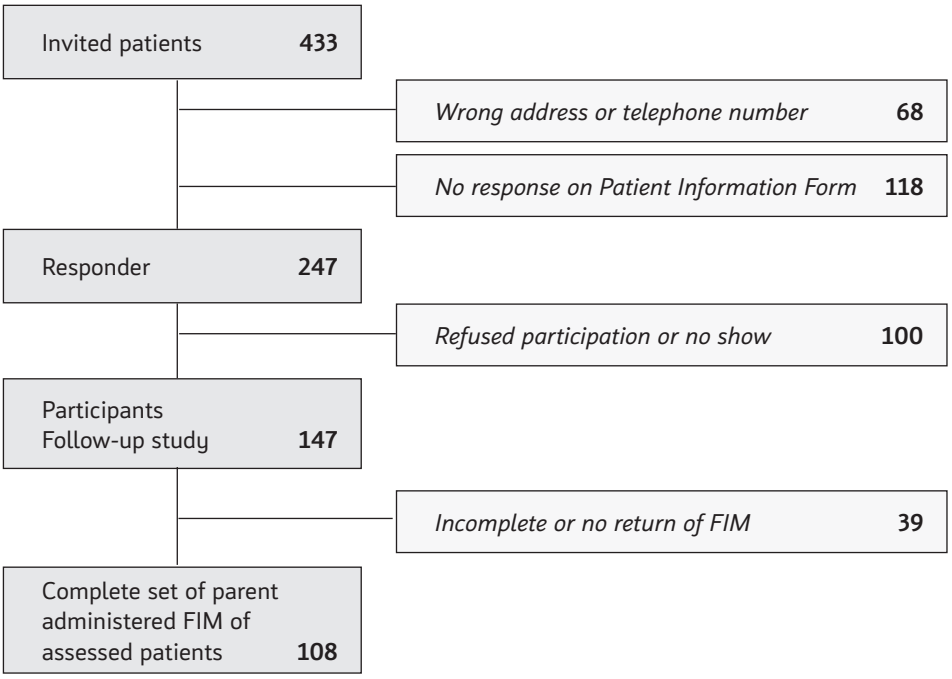
The sample size of n=108 supports the number of analyses conducted. The use of different classification systems for severity in TBI and NTBI warranted the need to conduct the analyses separately in those subgroups. As this categorization yielded a relatively small number of patients in the various categories of severity, we also combined TBI and NTBI to examine the impact of severity on family impact. In addition, for all analyses, a p-value less than 0.05 was adopted as the criterion for statistical significance. All data were analysed using SPSS version 21.0 software.[35] Missing values were processed according to instructions of each questionnaire.

RESULTS

Participants

The flow of patients is presented in Figure 1. In total, 147 participants were included in the present follow-up study. Of those, a total of 108 (60%) parents filled in the PedsQL™FIM. Eighty-one (75%) of the patients had TBI, with 62 (77%) being classified as mild and 19 (23 %) as moderate/severe. There were 27 patients with NTBI (25%), of whom 22 (81%) were classified as mild and 5 (19 %) as moderate/severe.

Figure 1 Flow chart recruitment



Regarding the presence of health problems among parents, the numbers (%) of parents reporting the presence of mental or physical health problems were 26 (22%) before ABI and 27 (23%) at present. For the presence of health problems among children, these numbers (%) were 23 (26%) before ABI and 36 (38%) at present.
Comparisons between participants in the follow-up study (n=147) and all invited patients (n=433) showed no significant differences regarding the distribution in age groups and types of injury. However, among the participants the proportion of patients with mild TBI was somewhat lower than in the total group (84 (78%) versus 359 (83%)).

Table 1 shows the characteristics of the 108 included participants with ABI and their parents. In the TBI group (75% of participants) the severity ratio mild: moderate/severe was 77:23. In the NTBI group (25%) the severity ratio mild: moderate/severe was 81:19. In the total ABI group 27 cases (26 %) reported pre-injury health problems versus 39 cases (38%) with health problems 2 years after onset of ABI. Parents reported a low educational level in 13 cases (13%) versus intermediate in 40 (40%) and high in 47 (47%) cases. Being a single parent household was reported by 31 (30%) parents.

Table 1 Characteristics of patients with Acquired Brain Injury and their parents in a study on family impact approximately 2 years after onset

		Cohort (n=108)
Age in years; median (range)		13 (5-22)
	Age Group ≤ 14 years old; number (%)	65 (60)
	Sexe, male; number (%)	60 (56)
Cause and severity		
Traumatic[1]	Total; number (% of total ABI)	81 (75)
	Mild; number (% of total TBI)	62 (77)
	Moderate/ Severe; number (% of total TBI)	19 (23)
Non-traumatic[2]	Total; number (% of total ABI)	27 (25)
	Mild; number (% of total NTBI)	22 (81)
	Moderate/ Severe; number (% of total NTBI)	5 (19)
Pre-injury physical or mental health problems; number (%) (n=104)		27 (26)
Actual physical or mental health problems; number (%) (n=103)		39 (38)
Educational level of parents; number (%) (n = 100)		
Low[3]		13 (13)
Intermediate		40 (40)
High		47 (47)
Single parent household; number (%) (n=102)		31 (30)

[1] determined by means of the Glasgow Coma Scale (GCS) at hospital admission
[2] determined by means of a disability scale based on the Modified Rankin Scale (mRS) at hospital discharge
[3] low (pre-vocational practical education or less), intermediate (pre-vocational theoretical education and upper secondary vocational education) or high (secondary education, higher education and/or university level education)

Family Impact after pediatric ABI

Regarding the PedsQL™FIM Total Score and the Summary Scores Parent Health Related Quality of Life Score and Family Functioning, the median scores varied from 81.7-85.4, with comparable score ranges (Table 2). The median scores for the Subscales Communication and Worry were 100 and 90, respectively.

In addition to the data presented in Table 2, the highest possible score, meaning no problems, was reported by 12 parents (11%) for the PedsQL™FIM Total Score, 26 parents (24%) for the Parent Health Related Quality of Life Score and 27 parents (25%) for the Family Functioning Score. The lowest possible score, meaning maximal family impact, was only reported once (1%), for the Subscale Worry.

Table 2 Scores on parent reported questionnaires and neurological outcome (PSOM) approximately 2 years after onset of ABI

Family Impact	Peds QL FIM-DLV ; median (range)	
	Total (range 0-100) n=108	82.9 (33.6 -100)
	Parent HR QoL (range 0-100) n=107	85.4 (33.5 -100)
	Family functioning (range 0-100) n=107	81.7 (30.8 -100)
	Communication (range 0-100) n=107	100.0 (33.3 -100)
	Worry (range 0-100) n=106	90.0 (0 -100)
Quality of Life	PedsQL General Core Scale-DLV; median (range) n=105	
	Total (0-100)	79.3 (40.8 -100)
Fatigue	PedsQL Fatigue-DLV; median (range) n=83	
	Total (0-100)	77.8 (36.1-100)
	Cognitive fatigue (0-100)	70.8 (37.5-100)
Activities and Participation	CFFS-DLV; median (range)	
	CASP Total (0-100) participation problems n=104	97.5 (42.5 -100)
	CAFI Total (33.3-100) limitations in functions n=107	40.0 (33.3-67.5)
	CASE Total (0-100) environmental limitations n=93	33.3 (33.3-59.3)
Actual functioning	PSOM professional reported; median (range) (0-10)	0.5 (0 -7)

The correlations of the PedsQL™FIM Total Score with the Parent Health Related Quality of Life Summary Score (r=0.971) and the Family Functioning Summary Score idem (r=0.879) were high ($p < 0.01$). The mutual correlation between the Summary Scores Parent Health Related Quality of Life parents and Family Functioning was 0.871, and significant at the 0.01 level (2-tailed).

Table 3 shows the results of the univariable analysis with the PedsQL™FIM Total Score, Summary and Subscale Scores as dependent variables and sociodemographic, pre-injury and injury characteristics and actual functioning ad independent variables. For the independent variables, patients were divided in subgroups according to fixed categories for nominal variables or by the mean score for numeric variables.

The FIM Communication and Worry Subscales were significantly different between younger and older patients, with lower scores in older patients. There was a significant difference between the FIM Total and Parent Health Related Quality of Life Summary Score and Family Functioning Summary Scores and the two Subscale Scores Communication and Worry in subgroups of patients with and without health problems before ABI, and between the TBI and NTBI groups.

Table 3 Results of univariable analysis of Total, Summary and Subscale Scores on PedsQL™FIM as dependent variables, related to baseline characteristics at onset (sociodemographic, pre-injury health, injury characteristics) and results at follow-up (actual functioning) approximately 2 years after onset of ABI, as independent or predictor variables*

	Predictor Variables		Family Impact Total Score	Quality of Life Summary Score	Family Functioning Summary Score	Communi-cation Subscale Score	Worry Subscale Score
Socio-demographic Characteristics	Age (at Onset)	≤14 y (N=65) †	82.8 (16.9)	84.4 (17.7)	79.8 (18.5)	89.3 (17.5)	81.9 (22.5)
		>14 y (N=63)	76.7 (18.8)	78.2(19.5)	77.0 (20.8)	77.5 (24.2)	70.8 (27.9)
	Sex	Male †	81.3 (16.9)	82.8 (17.3)	80.2 (18.2)	85,4 (20.2)	76.1 (24.6)
		Female	79.3 (19.1)	80.8 (20.1)	76.7 (20.8)	83.5 (22.5)	79.2 (26.3)
	Educational Level Parents	Low (N=11) †	75.7 (16.3)	77.8 (17.0	77.6 (20.5)	82.1 (22.0)	63.8 (28.6)
		Intermediate (N=40)	78.7 (16.9)	79.3 (18.3)	77.9 (19.3)	86.1 (19.9)	78.0 (23.1)
		High (N=47)	82.7 (19.9)	85.3 (19.6)	79.3 (20.6)	84.6 (22.4)	79.3 (26.8)
	Single Parent Household	Yes (N=31) †	77.0 (17.7)	78.3 (19.2)	76.5 (19.0)	85.2 (20.0)	72.7 (28.0)
		No (N=71)	82.4 (17.8)	84.2 (18.0)	79.9 (19.9)	85.7 (21.2)	79.6 (24.6)
Pre-injury Functioning	Health Problems	Yes (N=27) †	71.1 (21.7)	72.7 (23.5)	71.2 (22.0)	83.3 (26.0)	65.4 (25.9)
		No (N =77)	83.4 (15.5)	84.9 (15.8)	81.3 (18.0)	88.6 (17.7)	82.0 (24.1)
Injury Characteristics	Type of Injury	TBI (N=81) †	83.6 (16.1)	85.1 (17.2)	80.8 (18.3)	89.7 (17.1)	83.2 (21.6)
		NTBI (N 27)	70.8 (19.6)	72.6 (19.7)	72.3 (21.4)	69.4 (24.7)	60.7 (28.0)
	Severity of Injury	Mild (N=84) †	81.8 (16.2)	83.4 (16.8)	79.8 (18.2)	84.9 (20.3)	79.5 (22.8)
		Moderate/ severe (N=24)	75.6 (22.5)	76.8 (23.4)	74.8 (23.0)	83.3 (20.3)	70.6 (31.9)
	Severity TBI	Mild TBI (N=62)	84.1 (15.4)	85.8 (16.2)	80.4 (18.3)	88.5 (17.9)	84.6 (18.6)
		Moderate/ severe TBI (N=19)	82.0 (18.7)	82.7 (20.3)	82.0 (18.6)	93.4 (14.3)	78.9 (29.5)
	Severity NTBI	Mild NTBI (N=22)	75.5 (17.0)	76.7 (17.1)	78.0 (18.1)	75.0 (23.7)	65.9 (27.7)
		Moderate/ severe NTBI (N=5)	50.1 (18.3)	54.4 (22.0)	47.2 (16.9)	45.0 (9.5)	38.8 (18.1)

	Predictor Variables		Family Impact Total Score	Quality of Life Summary Score	Family Functioning Summary Score	Communi-cation Subscale Score	Worry Subscale Score
Actual Functioning	PSOM-SNE §	Low (=0) (N=43) ‡	85.1 (15.3)	86.6 (16.7)	81.7 (17.4)	89.8 (17.3)	85.9 (20.8)
		High (>0) (N=63)	76.5 (19.0)	78.1 (19.3)	76.1 (20.7)	80.2 (23.1)	70.5 (26.6)
	CASP	Low (≤97) (N=51) ‡	74.8 (17.8)	76.9 (18.5)	72.9 (19.7)	78.5 (22.8)	70.3 (26.4)
		High (>97) N=53)	85.2 (16.8)	86.3 (18.0)	83.6 (17.8)	90.3 (17.4)	83.9 (23.2)
	CAFI §	Low (≤40) (N=59) ‡	88.7 (12.2)	89.8 (12.8)	85.6 (15.8)	95.2 (11.7)	90.0 (18.1)
		High (>40) (N=48)	69.9 (18.4)	72.1 (20.1)	69.5 (19.9)	70.9 (22.7)	61.3 (24.3)
	CASE	Low (≤33) (N=53) ‡	88.6 (13.1)	89.4 (13.4)	86.6 (16.0)	92.6 (16.2)	89.2 (19.6)
		High (>33) (N=50)	71.1 (18.6)	73.4 (20.4)	69.8 (19.6)	75.5 (22.7)	64.0 (24.9)
	PedsQL QoL	Low (≤80) (N=53) ‡	69.9 (17.8)	71.8 (19.2)	70.2 (19.6)	74.9 (23.4)	61.8 (26.0)
		High (>80) (N=52)	91.3 (100)	92.9 (9.9)	87.1 (15.5)	95.4 (10.6)	92.9 (12.6)
	PedsQL Fatigue	Low (≤78) (N=42) ‡	68.8 (17.5)	70.3 (18.7)	69.0 (20.1)	76.6 (23.4)	60.9 (26.9)
		High (>78) (N=41)	93.1 (7.8)	95.0 (6.8)	88.7 (13.5)	97.2 (6.9)	94.0 (12.4)

* all variables are expressed as mean SD; high score indicates better functioning, except for §: high score indicates bigger problem
† group split in categories or ‡ group split in categories by median score
Dark gray cells indicate significant difference between groups, tested by t-test and One war ANOVA (for Educational level parents)

Moreover, for the total group of ABI the severity of injury was associated with family functioning. This association was seen in the NTBI subgroup as well, but not in the TBI subgroup.
Moreover, almost all measures of functioning and participation at follow-up participation (CAPE, CASP), environmental factors (CAFI), fatigue (PedsQL™Fatigue) and quality of life (PedsQL HR QoL). Similar results were found for neurological functioning (PSOM), except for the association with the Summary Score on Family Functioning, which did not

reach statistical significance. There was no indication of possible collinearity among the independent variables to be entered in the multivariable model (sociodemographics: patient age and gender; educational level parents and single or double parent household; presence of health problems before and after ABI; injury characteristics: type, severity) (tolerance values of all variables > 0.2). Table 4 shows that in a multivariable model the type of ABI (NTBI > TBI), severity (moderate/severe > mild) , and the presence of health problems before ABI were associated with more family impact, according to the PedsQL™FIM Total Score, with the final model accounting for 21.4% of the variance. As the type of injury (TBI or NTBI) was included in the model, no analyses for TBI and NTBI separately were done to examine the impact of severity on the PedsQL™FIM Total Score within subgroups of ABI. Sex (p=0.929), age (p=0.655), single parent household (p=0.356) and parents' educational level (p=0.426) were not significantly associated with family impact.

Table 4 Results of multivariable regression analysis, with Total Score on the PedsQL™FIM as dependent variable, approximately 2 years after onset of ABI, related to significant predictive baseline characteristics at onset of ABI

		Regression coefficient B	Significance level*	95% Confidence Interval	
				Lower Bound	Upper Bound
Intercept		54.929	.000	44.394	65.464
Pre-injury health problems	No	12.628	.001	5.250	20.007
	Yes	0[a]	.	.	.
Type of injury	TBI	11.740	.002	4.445	19.035
	NTBI	0[a]	.	.	.
Severity of injury	Mild	9.140	.020	1.449	16.830
	Moderate/ Severe	0[a]	.	.	.
		R^2 = .214			

* $p < 0.05$

[a] this parameter is set to zero because it is redundant

DISCUSSION

In a selected group of children and youth with ABI, with relatively many children with mild TBI or NTBI and only few being treated for consequences of ABI, the impact on the family as measured by the PedsQL™FIM was considerable.

The results of our study suggest that the PedsQLTM FIM is a promising, multidimensional instrument to measure family functioning, parental health related quality of life, communication and worries after pediatric ABI, with high mutual correlations between Total, Summary and Subscale Scores. The Subscales Communication and Worries are additional to other specific family impact measures.[25,26] The availability of an appropriate

instrument to measure and monitor family impact and functioning after ABI is important, as has been previously suggested in the literature.[13-17] Subsequently, the development and implementation of specific family centered interventions in rehabilitation and chronic care for youth with ABI has been advocated.[45,46,47]

The results of the univariable analysis showed that functioning 2 years after ABI was associated with the PedsQL™FIM Total Score (Family Impact), Summary Scores (parents' Quality of Life and Family Functioning) and Subscale Sores (Communication and Worries).

This is in line with other studies concerning patients' actual functioning (in our study measured with PSOM, CAFI),[14,22] participation (CASP), quality of life (PedsQL HR QL)[18,30] and environmental factors (CASE).[16,17] Our results support the importance of (measuring) fatigue as associated with family impact, just as others published on fatigue after pediatric ABI related to general health-related problems[23] and sleep problems.[24] Similar to other studies[14,48] it was found that current health problems of children were found to impact family functioning after ABI. The variables concerning actual functioning were not entered into the multivariable prediction model, as they concerned the outcome of ABI rather than its starting point.

The prediction model of family functioning after ABI using only sociodemographic, pre-injury and injury characteristics, showed that the presence of NTBI, a greater severity of either TBI or NTBI and the presence of pre-injury health problems were associated with more family impact. These findings are largely in line with the literature.[5,6,14,49] However, the impact of the type of ABI on family impact has been scarcely studied, as most studies were so far done among specific diagnosis groups. The finding that NTBI had a greater impact on family functioning than TBI. This difference may be due to the different nature of the two types of ABI, with TBI having a transient and/or steady course in many patients, whereas the underlying conditions in NTBI may have other consequences, such as side effects of medical treatment and risk of recurrence or relapse.[10,12]

In our study 'younger age at onset' was not significantly associated (r=0.655) with family impact. This is surprising, as younger age at onset, has been previously associated with poorer outcomes in the literature.[13,27] A potential explanation may be the so called 'growing into deficit' theory.[49] This theory implies that children and youth may experience a decline in functioning later on in their development, when brain development is supposed to support age-specific cognitive and behavioral competences that are required for more complex demands,[50] e.g. in transitions to higher levels of education, work, social intimacy or living independently. In contrast with the literature, we found 3 baseline characteristics being not significantly associated with family impact: parents' educational level,[14,15] single parent household,[15] and sex.[50]

Several limitations of our study should be noted. First, the generalizability of the results is probably limited by the selection of the cohort. Patient recruitment was done in hospitals and not in the rehabilitation setting. Therefore, the population consisted of patients with

predominantly mild ABI, not requiring treatment. The results are therefore not generalizable to groups of patients with ABI who are currently treated for the consequences.[5,6,38] According to literature[51,52] approximately 20% of children with mild TBI is hindered by consequences after 3 and 10% after 12 months, respectively. Differences with other studies may be explained by these limitations. The relatively high number of non-responders may be a confounder. This is likely due to the relatively high percentage of children and youth without consequences after a mild ABI. Another reason for non-response could be that the invitation for participating in the study was sent by mail two years after the hospital based diagnosis. We did not systematically record the reasons for non-participation. Some of the non-response was due to wrong addresses, and is probably random. Although response bias cannot be excluded, the characteristics of the patients at hospital admission or discharge in the present study are fairly similar to those of the larger population, which was described in a previous publication.[44] The relatively low response resulted in an overall small sample size, which may have limited the statistical power of the study.

Another limitation is time since onset: 2 years after the hospital based diagnoses is a relatively long period in which many other factors may influence outcomes such as family functioning as well, and for parents it is a long period to reflect on.

Another limitation is the difference in the classification systems and time points used for severity grading between TBI and NTBI employed in the present study. For this reason we did analyses within the two subgroups separately. The association between severity and FIM appeared to be stronger in NTBI than in TBI. Despite the observation that the impact of severity on family functioning remained in the multivariable model including the type of ABI as a separate independent variable, it could be hypothesized that severity as determined at hospital admission is a better predictor for future functioning in NTBI than in TBI. This finding underscores the need to take the differences between the two types of ABI, as well as the classification systems for their severity, always into account when conducting research in this area. Another limitation concerning the assessment of neurological functioning was that we used the PSOM, which has only been found to be a reliable and valid measure in paediatric stroke, but not in other forms of NTBI or in TBI. However, at the time the study was designed, it was considered the best available quantitative instrument providing a standardized neurological assessment in all diagnosis groups.

Finally, a limitation of the study relates to the interpretation of the magnitude of the observed PedsQL™FIM scores in the group of patients with ABI. To our knowledge, there is no literature on this subject in this patient group available yet. Future studies are needed to define the Minimal Clinically Important Difference (MCID) of the PedsQL™ FIM, the difference in scores that can be interpreted as clinically meaningful, in children with ABI.

To overcome these shortcomings, a larger scale and longitudinal study including sufficient numbers and proportions of children with mild, moderate and severe ABI would be needed.

CONCLUSION

Family impact following a diagnosis of paediatric ABI involves risks of long-term psychosocial problems for parents and families, partly due to the specific consequences of pediatric ABI. The results of this study support the importance of the systematic monitoring of family impact to enable tailor-made psycho-education, follow-up and support for parents, brothers and sisters. The PedsQL™FIM appears to be an appropriate, multidimensional instrument for measuring and monitoring family impact after pediatric TBI and NTBI. Further research on family impact after ABI is required to further elucidate associated factors and examine the utility of cumulative risk index.[53]

Declaration of Interest statement

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

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