

Unresponsive Wakefulness Syndrome in Children after Near-Drowning: Long-Term Outcome and Impact on the Families

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Abstract

Among 131 children admitted to our institution for early phase rehabilitation after freshwater near-drowning (ND) between the year 1986 and 2000, 87 were in unresponsive wakefulness syndrome (UWS) for at least 4 weeks after the accidents. An anonymous questionnaire was sent to the families after 0.5 to 15.0 years (median: 4.6) and 48 mothers and 51 fathers of 55 of these 87 children were interviewed after 6.6 to 23.8 years (median: 13.8) of ND. At the time of the interviews, 8/55 children were able to perform daily living activities independently, 36/55 children were not able to do so (many of them suffered from chronic medical conditions like spasticity or disorders of swallowing), and 11/55 children had died. Health-related quality of life (HRQoL) was, however, similar to the normal population for mothers, and even higher for fathers. Furthermore, the ND accident had apparently not lead to a higher rate of separations of parents but had increased their likelihood to have further children. Feelings of guilt were highly prevalent (23/47 mothers, 20/47 fathers), and correlated with lower HRQoL of the respective parent. We found correlations between duty of supervision and feelings of guilt and between outcome and HRQoL for only the fathers. In conclusion, we found that after 4 weeks in UWS, the long-term neurological outcome of pediatric ND victims is often but not always poor. Despite often severe disabilities or death of the child during long-term care, parents surprisingly report little impact on their HRQoL, on the stability of their partnership or on their wish to have further children. Our findings may help parents and physicians to choose the best treatment for a child in UWS due to different etiologies striking the balance between rehabilitation and palliative care.

Keywords

- ▶ persistent vegetative state
- ▶ children
- ▶ near-drowning
- ▶ health-related quality of life
- ▶ long-term outcome
- ▶ unresponsive wakefulness syndrome

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Introduction

Pediatric near-drowning (ND) accidents, leading to life-long disabilities due to hypoxic brain injury, are one of the worst tragedies for both the affected children and their parents. In addition to the burden that the disabilities of their children impose on them, many parents suffer from feelings of guilt related to the circumstances of the accident.¹

But although pediatric near-drowning accidents are frequent and the outcome is often unfavorable, little is known about the long-term outcome.^{2–14} Whether favorable outcome is also possible after prolonged periods (i.e., many weeks) of unconsciousness cannot be derived from these studies or is even questioned.^{3,4}

Furthermore, no study has yet prospectively addressed the impact ND accidents have on the families. Little data are available from two cross-sectional studies: Nixon and Pearn¹ interviewed 56 families in which nonfatal ND immersions had occurred; Giovannetti et al¹⁵ reported on caregivers' burden in families of 35 children in so called "vegetative and minimally conscious state" after acquired severe brain injuries (including both hypoxic and traumatic events).

The paucity of information on the long-term perspective of pediatric ND victims and their families is all the more surprising since publications have questioned the therapeutic value of prolonged resuscitation⁶ in the presence of indicators of "poor outcome" (which, in most publications, stands for death or severe neurological sequelae without distinguishing between the two).^{12,13,16}

The background for initiating our study was that we started as one of the first centers specialized for pediatric early neurorehabilitation in 1990 in Germany and often realized a conflict in the rehabilitation team between continuing or withdrawing of life-sustaining treatment when children persisted in "unresponsive wakefulness syndrome" (UWS) for months or only showed minimal improvement.¹⁴ Heindl and Laub¹⁴ had already shown in a cohort of children with UWS following hypoxic (including ND) or traumatic brain injuries in our center that after 19 months, 84% of the patients of the traumatic brain injury group and 55% of the hypoxic brain injury group had left UWS.¹⁵ For a comprehensive rehabilitation concept including counseling the parents about the prognosis, there was a need but lack of long-term outcome data of children in UWS and the impact on the families. We chose the group of fresh-water ND for a long-term follow-up because (1) this was our largest group with patients in UWS after hypoxic brain injuries, (2) of the consistent pathophysiology, and (3) because of the well-established worse prognosis compared with UWS after traumatic brain injuries.

Patient and Methods

In the year between 1986 and 2000, 131 children were admitted to our institution at a mean of 74 days (range: 13–268 days) after near-drowning accidents in freshwater for early-phase rehabilitation (i.e., directly following acute care in general pediatric hospitals). Four weeks after the

Table 1 Criteria for "persistent vegetative state" (PVS), as defined by the multi society task force on PVS^{12,13}

| |
|---|
| • No evidence of awareness of self or environment |
| • No ability to interact with others |
| • No reproducible, purposeful, or voluntary response to visual, auditory, tactile, or noxious stimuli |
| • No evidence of language comprehension or expression |
| • Presence of sleep-wake-cycles |
| • Sufficiently preserved hypothalamic and brainstem autonomic functions |
| • Variably preserved cranial-nerve reflexes and spinal reflexes |

Note: For inclusion in our study, all criteria had to be fulfilled 4 weeks or more after the accident. In our paper we use the new expression "unresponsive wakefulness syndrome" (UWS) synonymously instead of PVS.¹⁴

accident, 87 of these 131 patients were still in UWS (i.e., they fulfilled all criteria listed in ►Table 1); 41 children did not fulfill all the criteria, and three children died despite taking all medical efforts to save their lives (i.e., none of the patients in our cohort had "do-not-resuscitate" orders during the inpatient phase, and all received optimal medical and nutritional support). Of these 87 children, we excluded 3 since they came from the same family (all victims of the same car accident with submersion), leaving a study cohort in which we could investigate the outcome and also the impact of 84 near-drowning accidents on 84 families.

E1: In the years 2001 and 2002, all 84 families were sent anonymous questionnaires with (semi-)structured and open responses covering demographic information, the child's physical condition, caregiving, any positive changes that developed in the family after the accident, and the psychosocial situation of the parents (English translation of questionnaire see additional material: **S1**; supplementary file is available in online version only).

E2: In 2010, the same 84 families were sent letters asking for the willingness of mother and father to be contacted again for structured telephone interviews led by an experienced pediatric neurologist (G.J.K.). These interviews took place in 2010 and 2011 and consisted of 23 (semi)structured and open questions covering all items of E1, and also the short form-8 (SF-8) health survey questionnaire for health-related quality of life (HRQoL; see questionnaire in the additional material **S2**; supplementary file is available in online version only). A second telephone interview took place with an occupational therapist (M.H.), who performed a semistructured interview, based on the remission profile for children and adolescents after severe acquired brain lesion (RemiPro), in which the level of responsiveness, activities, and participation of each patient was determined. Six levels were distinguished (►Fig. 1).¹⁷

Data were subsequently anonymized and handed over to another pediatric neurologist (M.S.) who, assisted by a statistician (H.A.) and analyzed the data and deducted interpretations. These were discussed among the authors and modified until consensus was reached. Data acquisition and

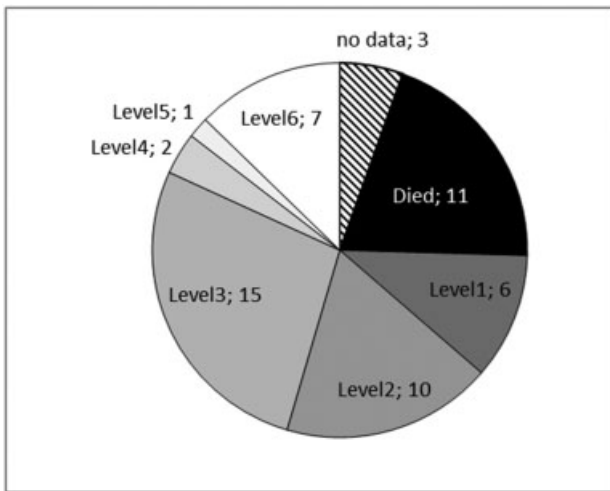


Fig. 1 Levels of responsiveness, activities, and participation for the 55 children at E2. Level 1: The patient does not or only occasionally show reactions in participative situations. Level 2: The patient repeatedly shows reactions but does not initiate actions. Level 3: the patient initiates simple actions. Level 4: The patient performs activities (as sequences of actions) but only in supervised and prestructured situations. Level 5: The patient performs activities independently in familiar everyday situations; however, with reduced speed and greater effort. Level 6: The patient performs all age-appropriate activities in normal speed and also in unfamiliar environment.

analysis were separated to prevent contamination of results and conclusions from any subjective and emotional factors that might have evolved during the interviews.

The study was approved by the ethical committee of the Bavarian Chamber of Physicians. All participants gave written informed consent for E2.

Statistics: For all analyses of dichotomic variables, we used Fisher’s exact test; for all other correlations, Kendalls Tau was calculated, both with $p < 0.05$ as the level of significance. To achieve maximum sensitivity for differences between groups and correlations, we did not correct for multiple comparisons.

Results

Demographic information for the cohorts studied at E1 and E2 is given in ►Table 2. The Kaplan–Meier survival curve is illustrated in ►Fig. 2.¹⁸ ►Fig. 1 displays the results of the interview on responsiveness, activities, and participation which was conducted for 41/44 patients who still lived at E2. Medical problems were reported for 35 of 43 patients who still lived at E2 and many suffered from neuro-orthopedic problems like spasticity or dystonia (28/43), scoliosis (26/43), and hip luxation (14/22). Tube feeding via gastrostomies was needed in 19/43 patients, 17/43 had intrathecal baclofen pumps implanted, and 2/43 patients were respirator-dependent. Epileptic seizures were noted in 22/43 patients and/or

Table 2 Demographic information for the cohorts studied at E1 and E2

| | E1 | E2 |
|--------------------------------|---|--|
| Response rate | 42/84 families (one questionnaire per family) | 55/84 families (48 mothers, 51 fathers) |
| Patients’ age at near-drowning | 0.8–8.3 y, median: 2.4 y | 0.7–8.3 y, median: 2.3 y |
| Time after near-drowning | 0.5–15.0 y, median: 4.6 y | 6.6–23.8 y, median: 13.8 y |
| Patients’ age at interview | 2.4–21 y, median: 8.5 y | 8.4–28.7 y, median: 16.7 y |
| Death since discharge | 3/42 | 11/55 |
| Divorce/separated | 6/39 (15%) | 12/55 (22%) |
| Feeling of guilt | 26/35 families (74%) | 23/47 mothers (49%) 20/47 fathers (43%) |

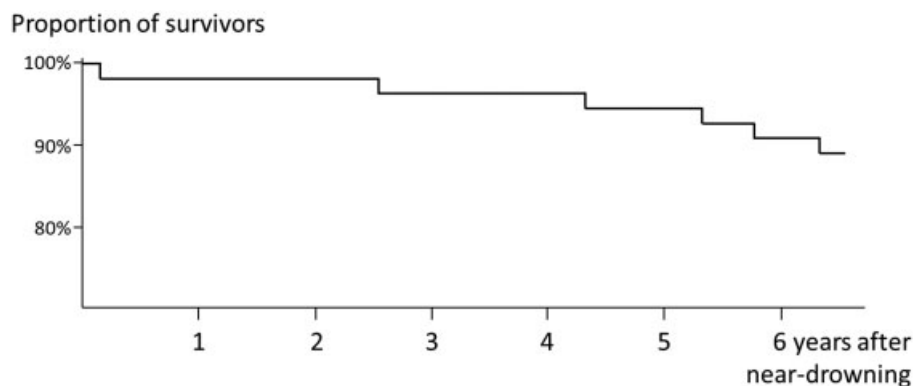


Fig. 2 Kaplan–Meier survival curve illustrating the times of death of the 12 children of our cohort who had died during the minimum follow-up period of E2. At least five more children had died in the years thereafter (9, 10, 13, 14, and 15 years after the accident) but these could not be included in the diagram, since their deaths had occurred beyond our minimum follow-up period of 6.6 years, and we cannot exclude that more children of our cohort died after the interviews had taken place.

were still on antiepileptic medication. No chronic medical problems were noted in 8/43 patients—the same eight patients who were able to independently perform activities (Levels 5 and 6, ►Fig. 1). Not all answers were obtained from all participating families; hence, the numbers often do not sum up to the number of participants in the respective cohort.

Intolerable suffering: At E2, among 22/55 patients (40%), at least one parent stated that it would have been better if their child had died in the acute phase (both parents: 7/44; 16%). On the other hand, for 39/55 patients (71%), at least one parent stated that they were glad that their child had survived the acute phase (both parents: 26/44; 59%). Diverging answers were given by 11/44 couples: six of these were definitively diverging, while in five couples, one partner (four fathers, one mother) was not sure how to answer this question. Comparing mothers and fathers who provided definitive answers, we found no significant differences (“better died”: 12/48 mothers, 17/51 fathers, $p > 0.3$; “not better died”: 35/48 mothers, 33/51 fathers, $p > 0.3$; Fisher’s Exact). At E1, with only one questionnaire available per patient, parents responded for 19/41 patients (46%) that they sometimes had wished that their child had died in the acute phase, they were not sure in 2/41 patients, and 20/41 patients (49%) responded that they had not wished for the death of their child. Note that, due to the anonymity of E1 and the different methodology (one questionnaire per patient at E1, one or two parents’ interviews at E2 per patient), we do not know how many parents had really changed their attitude toward this question between E1 and E2. With all due caution, the data might indicate that the parents’ attitude had shifted from “I sometimes wish that the child would have died” (E1: 46% → E2: 16–40%) toward “I am glad the child survived” (E1: 49% → E2: 59–71%).

Caregiving: Most patients lived with their families, with only a subtle decrease of the proportion over time from E1 (35/38 patients [92%]) to E2 (36/44 patients [82%]). For 16 of the 36 patients who lived at home at E2, the parents were the only caregivers. For the other 20 patients, additional care was provided by professional outpatient nursing services (7/20), relatives (12/20), friends (4/20), and other persons (5/20). At E2, 42/44 patients received professional therapies like physiotherapy, occupational therapy, or speech therapy, most often two (10 patients) or three sessions per week (13 patients).

Demographic situation of the families: Of the 55 families at E2, 43 couples reported that they still lived together, while 12 had separated (22%). Since no normative data exist for this proportion, we can only give for comparison that in the general German population, 31% of all couples who married in the year 1994 (the median birth year of our patients) were divorced by the year 2011 (the median year of E2; data provided by the Statistisches Bundesamt). Furthermore, 17 families reported that at least one further child had been born after the near-drowning accidents (31%). In the general German population (raw data provided by the Statistisches Bundesamt), an estimated average of only 13% of the families (range: 0.6–34%) in the same situation (with respect to year of the accident, number, and age of children) would have been expected to have at least one further child (see ►Appendix 1 for calculation). Thus, the affected families had a clearly

increased probability to have further children after the near-drowning accidents compared with families without near-drowning accident in the same constellation of children.

Duty of supervision and feelings of guilt: Of the families responding at E2, the duty of supervision at the time of the accident was with (multiple naming possible): the mothers (35/47), the fathers (14/47), other persons (16/47). As expected, many parents who had the duty of supervision at the time of the accident continued to have feelings of guilt (18/35 mothers, 10/14 fathers; not significant). Feelings of guilt were, however, also present in parents without duty of supervision (5/12 mothers, 6/33 fathers; n. s.). We hypothesized that parental feelings of guilt would be aggravated by, and thus correlated with, the duty of supervision, poor outcome, the perception of intolerable suffering of the child, and by death of the child during long-term care (►Fig. 3). Surprisingly, for the mothers, none of these expected correlations could be confirmed. Similarly, for the fathers, we found only a correlation between the duty of supervision and feelings of guilt ($p < 0.001$; Fisher’s Exact) but no other significant correlations.

Health-related quality of life (►Fig. 4): For the mothers of near-drowning victims, neither of the HRQoL sum scores “mental component summary” (MCS) nor “physical component summary” (PCS) were, on a group level, different from female controls (raw data of female and male controls provided by the Robert Koch Institute, Germany). Looking at the subscores, we found that only the item “global health” (GH) was significantly lower than in controls. For the fathers of near-drowning victims, we also found, on a group level, no difference to male controls for the MCS. For the PCS, fathers of near-drowning victims scored significantly higher than controls. Looking at the subscores, we found this superior HRQoL especially for the item “bodily pain” but also for “physical functioning,” “vitality,” and “social functioning.” In terms of correlations (►Fig. 3), we hypothesized that the parental HRQoL (analyzed for MCS and PCS) would be reduced by and thus correlate with feelings of guilt, poor outcome, the perception of intolerable suffering of the child, and with death of the child during long-term care. We found for mothers ($p = 0.039$) and fathers ($p = 0.027$) that feelings of guilt indeed correlated negatively with MCS (Kendalls Tau). For the mothers, this was the only significant correlation, whereas for the fathers, outcome of the child also correlated with the MCS ($p = 0.002$; Kendalls Tau).

In the free text sections, positive changes after the accident were reported by 28/41 families (68%) at E1 and 73/102 parents (72%) at E2. The most frequently mentioned positive aspect was that parents had the feeling that the accident had helped them “to find out what really mattered in life.”

Discussion

Our study provides unique long-term data about outcome and impact on the families in a large cohort of children in UWS after ND who had been sent for pediatric early rehabilitation to a specialized center. As already shown in a study

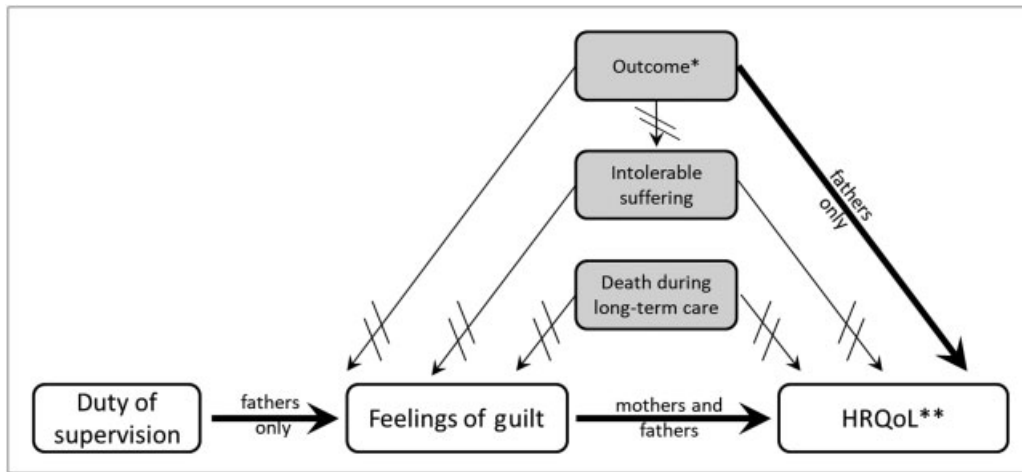


Fig. 3 Schematic illustration of hypothetical correlations (= arrows) between patient-related factors (gray boxes) and parent-related factors (white boxes). Significant correlations are marked as thick black arrows, insignificant correlations are crossed out. Note that only one-tailed statistics was used, except for correlations with “death during long-term care,” where both directions seemed plausible. *level of responsiveness and participation on ordinal scale (levels 1–6). **significant correlations were only found with the mental component summary (MCS) but not with the physical component summary (PCS). HRQoL, health-related quality of life.

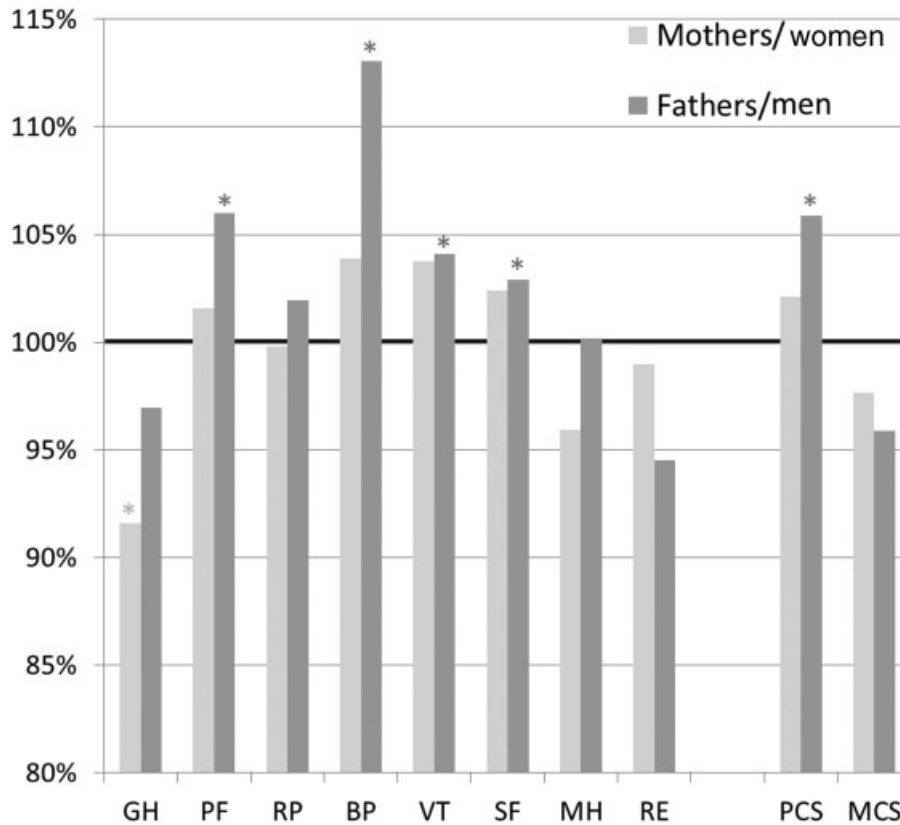


Fig. 4 Ratio of HRQoL scores (parent/normal population) of the SF-8 questionnaire, with numbers above 100% indicating higher scores for the parents of near-drowning victims (light gray = mothers/women; dark gray = fathers/men). *significant differences between parents and the normal population; BP = bodily pain; GH = global health; HRQoL, health-related quality of life; MCS = mental component summary (comprising VT, SF, MH, RE); MH = mental health; PCS = physical component summary (comprising GH, PF, RP, BP); PF = physical functioning; RE = role emotional; RP = role physical; SF = social functioning; VT = vitality.

of our center and other studies the outcome of children in UWS is worse after hypoxic brain injury (including ND) compared with UWS after traumatic brain injury.^{14,15,19–22} Therefore we think that the data of our study are helpful for a rehabilitation team to counsel parents of children in pro-

longed disorders of consciousness of different etiologies when there is a conflict between continuing or withdrawing of life-sustaining treatment. This was the primary goal of our long-term study in 1998. In addition, we think that our data might also be of interest discussing medical, legal, and

ethical aspects in emergency situations or intensive care units initiating or withholding life-sustaining treatments in children with severe brain injuries.²²

At the end of our study (i.e., interviews at E2) all life-sustaining treatments including tube feeding and ventilation were strictly not withdrawn during the rehabilitation in our center, following legal and ethical aspects in Germany at those days. Although not included as a standardized question at E2, to the best of our knowledge, in all deceased patients at E2, death did not occur because these life-sustaining treatments had been withdrawn (note: two patients at E2 were still in UWS and respirator-dependent). Therefore our long-term data are probably different from other studies of children in UWS after ND and other etiologies in countries with different access to early rehabilitation and/or attitude to palliative care.

It is likely that the results of our study would be different in the nonresponders. But the response rates at both E1 and E2 were high (50 and 66%, respectively), although as recommended by the ethical committee, the questionnaire at E1 was an anonymized written postal questionnaire and the telephone interview at E2 took place after parents had given written informed consent sent by mail.

With respect to patient outcome, the major finding of our study was the wide range of outcomes. With our inclusion criteria of UWS 4 weeks after ND, we were not surprised to learn that several patients had died during long-term care (11 / 55; 20%); however, we had not expected to find such a high proportion of recovered patients (7/55; 13%) who participated in all age-typical aspects of life at the time of the interviews at E2, and for whom no chronic medical problems were reported. This finding contradicts a study⁴ reporting that all satisfactory survivors after ND (17/44 patients in the study cohort) were sufficiently awake to have spontaneous, purposeful movements within 24 hours after the drowning event, whereas all children (27/44) who did not show this fast recovery survived with severe deficits or died. In this context, it is important to clarify that this cohort with “poor recovery” included three children who “were removed from mechanical ventilation because of persistent severe neurologic injury on the 4th or 5th days after the submersion,” without specifying what the “persistent severe neurologic injury” was.⁴ Our findings also contradict a study³ reporting that all 7 out of 49 nearly drowned children who made good recoveries regained consciousness within a 2-week period.

We cannot conclude after how many weeks of UWS after ND a full recovery was possible and if magnetic resonance imaging (MRI) can help to predict the prognosis as this was not included in our study design. But according to a retrospective analysis of entries in the medical records, the seven patients with full recovery showed first clear responses to environment (i.e., eye contact) at the latest of 8 weeks after ND.

Nevertheless, our outcome data with shows full recovery in 15% of children in UWS 4 weeks after ND advocate for optimal emergency medicine and intensive care support not only in the acute phase^{19,20} but also at least during the first weeks after ND and against an overhasty withdrawal of

life-supporting measures during early rehabilitation in these children.

However, most patients survived with often severe and chronic medical conditions consisting of neuro-orthopedic problems, the need for gastrostomy tube feeding, for intrathecal baclofen or, in two cases, even for chronic mechanical ventilation. But still, the majority of parents stated that they were glad that their child had survived the acute phase of the accident which we interpret as absence of intolerable suffering of their child. Again, this justifies full emergency medicine and intensive care support in the acute and subacute phase of these children and questions whether this outcome should be regarded as “poor” and to be avoided by early withdrawal of life-supporting medical measures.

On the other hand, for 7/44 patients (16%), both parents agreed that it would have been better if their child had not survived the acute phase after the accident. We interpret this as indicating the parental perception of periods of intolerable suffering of their child. Surprisingly, we found no correlation of this parameter with the outcome of the patient (regarding responsiveness and participation), indicating that periods of intolerable suffering are perceived not only by patients with “poor recovery.” The high proportion of patients who apparently experienced periods of intolerable suffering is a serious finding of our study, since it indicates failure of all medical efforts to overcome the problems leading to this perception. This finding has enormous implications for professionals involved in the long-term care of these patients, calling for intensified monitoring for and medical efforts against negative symptoms in these patients. It is in accordance with the Statement of the Ethics Working Group of the Confederation of the European Specialists of Pediatrics,²² to go into discussions both within the team of professional supporters and with the families about withholding or withdrawing life-sustaining measures when these efforts fail.

With respect to the affected families, the major and most surprising finding was that the impact of near-drowning accidents was little, despite the fact that long-term outcome was often severe. Subjectively, the parents HRQoL was comparable to the normal population (details see below), although, at least for fathers, we found the expected correlation between patient outcome and the MCS score of the SF-8. In contrast, HRQoL of the mothers did not seem to be influenced by the outcome of the child. This is highly important information for communication with the parents in all phases of rehabilitation; normal quality of life can be possible for the parents even when the long-term outcome of their child is severe. This is surprising since the accident had dramatically changed the lives of the families: Even at E2, many years after the accident, 82% of the children still lived with their families, and 93% of them received professional therapies (like physiotherapy) at least on a weekly basis. The high proportion of children living at home is similar to data from Giovannetti et al,¹⁵ reporting that 77% of Italian children with vegetative state and minimally conscious state lived at home.

Also in objective measures, the accident did not have major impact on the psychosocial situation of the families: the expected rate of divorce/separations had probably not

increased and the wish to have further children had probably even increased after the near-drowning accidents from an expected probability of 13% (families in the same constellation of children in the general German population, i.e., without the accident) to 31% (after the accident). This figure demonstrates that the burden of having a near-drowning victim in the family was obviously not so severe as to reduce the parents' wish to raise more children. These observations are compatible with Nixon and Pearn¹ who report that none of 54 families of pediatric near-drowning victims separated (contrasting with 24% of the families of fatal drowning accidents).

Our results differ, at first glance, from those reported by Giovannetti et al,¹⁵ who using the SF-12 in caregivers of 35 Italian children in vegetative and minimally conscious state, found PCS within the normal range but MCS below the normal population. The overall higher HRQoL scores in our study might be attributed to several factors: first, the study¹⁵ comprised only patients in "vegetative state and minimally conscious state" at the time of the survey, whereas our sample also included many patients who had recovered from these conditions. Therefore, the difference could be due to a less severely affected cohort of patients in our study. Second, we interviewed both parents, whereas Giovannetti et al¹⁵ included only data from the "main caregiver" who in 30/35 cases was the mother and mothers indeed showed similar results in our study with PCS in the normal range and a reduction in one subscore of the MCS. Similar to Giovannetti et al,¹⁵ the HRQoL of mothers' did not correlate with their child's outcome. Further studies about the long-term effect of children in UWS should include measurements of the brothers and sisters and focus on an even longer follow-up as in our study, that is, adult age when the parents are no longer able to take care of their grown-up children.

On the other hand, feelings of guilt were highly prevalent in the parents of our study, even many years after the near-drowning accidents (49% of mothers, 40% of fathers), and apparently did not become better (nor worse) when the child died during long-term care. Surprisingly, the expected correlation between feelings of guilt with the duty of supervision was only found for the fathers but not for the mothers. From the interviews, we had the impression that the reason for this might be that many mothers reported to feel guilty because they had left the child in the care of other persons (e.g., the father or a grandparent). We were also surprised by the lack of correlation between the outcome of the child and the feelings of guilt. Both aspects are important findings, since they help a professional team to know that they have no predictors for development of feelings of guilt, so that they must expect this phenomenon irrespective of the circumstances of the accident and outcome parameters. Furthermore, the high prevalence of feelings of guilt should be taken into account when discussing palliative aspects of care. It is difficult to predict how far the feelings of guilt change when the child dies as a consequence of withholding or withdrawing life-sustaining measures. In our analysis, we found no influence of death during long-term care, neither on feelings of guilt nor on parental quality of life, while others reported less severe grief disorders in parents whose children were still alive.¹ At E2, 40/49 (82%) of the mothers and

34/50 (68%) of the fathers answered "yes" to the last question "Did the illness of your child lead to any positive change in your life?". On the other hand only a few parents could give an advice on what was helpful against their persistent feelings of guilt, for example, talking within the family ($n = 8$), professional psychotherapy ($n = 8$), religion ($n = 3$), time ($n = 3$).

Truly, our study has several shortcomings. First, the return rate was only 65% (at E2), so we do not know how far our findings are representative for our cohort. And it seems likely that our findings regarding parental coping might be biased in a positive direction, since especially those parents with less efficient coping mechanisms might not have agreed to be interviewed. Second, our time points E1 and E2 were chosen as short periods during which the data could be collected and therefore not at a fixed time after the accident. Therefore, the time that had elapsed between the accident and the collection of the data varied considerably and even overlapped between E1 (0.5–15 years) and E2 (6.6–22 years). Third, due to the anonymity of E1, we do not know how many families participated in both, so we cannot tell in how many parents the answers had really changed between E1 and E2. Still, with the similar age ranges between E1 and E2, it is justified to compare, even if not statistically, the response patterns of the parents between "earlier" and "later" during long-term care. Fourth, the outcome scale we used was rather crude, and the highest level of our scale can definitely include children with marked neuropsychological difficulties (in fact, for many of these patients, the parents reported difficulties in concentration, visuomotor abilities, long-term memory, or working speed; several children also in this level still had ongoing physiotherapy, speech therapy, or occupational therapy sessions). Future studies need to include neuropsychological testing in children with apparently good outcome. The reader must also acknowledge that all numbers provided by the Statistisches Bundesamt are only rough estimates, since all proportions are calculated with indicators from 1 year (e.g., number of divorces) and denominators from another year (e.g., number of marriages), thus ignoring many factors like migration of families, etc.

Conclusion

In conclusion, our study on long-term outcome after severe pediatric ND accidents demonstrated that favorable outcome is possible even after prolonged periods (here: 4 weeks in UWS) of severe disorders of consciousness, calling for optimal emergency medicine and intensive care support for all victims in UWS after ND and other severe brain injuries. On the other hand, several parents apparently perceive periods of intolerable suffering of their children which calls for intensified efforts in the field of palliative care. Parents can be reassured that for themselves, a normal quality of life can be achieved despite the accidents-with hardly any identifiable correlations with outcome factors. Furthermore, parents can be informed that, on average, the accident will most likely not destabilize their partnership or marriage and that many families decided to have further children after the accident, probably more than in the normal population.

Professionals dealing with these parents must be aware that parental feelings of guilt are highly prevalent even years after the accident and that such feelings must be expected irrespective of the patient's outcome (including death) during long-term care and even when the duty of supervision had not been with the respective parent when the accident occurred.

Overall, the primary goal of our study is fulfilled. We have long-term data about children in UWS and can therefore better counsel the parents. We considered the very complex medical, legal, ethical, and psychological aspects and changes in perception and the legal situation while having treated more than 2,500 children in UWS in our center over the past 28 years. Through this, we have made an important contribution to our debate about withholding or withdrawal of life-sustaining treatment in children in UWS. Now, we put an even stronger emphasis on a collaborative decision with the families to define which goals are best for each child during all stages of rehabilitation. Moreover, we now aim to choose the optimal treatment that enables participation in society and personal satisfaction in life.

Conflict of Interest

None.

References

- Nixon J, Pearn J. Emotional sequelae of parents and sibs following the drowning or near-drowning of a child. *Aust N Z J Psychiatry* 1977;11(04):265–268
- Suominen PK, Vähätalo R. Neurologic long term outcome after drowning in children. *Scand J Trauma Resusc Emerg Med* 2012;20:55
- Bell TS, Ellenberg L, McComb JG. Neuropsychological outcome after severe pediatric near-drowning. *Neurosurgery* 1985;17(04):604–608
- Bratton SL, Jardine DS, Morray JP. Serial neurologic examinations after near drowning and outcome. *Arch Pediatr Adolesc Med* 1994;148(02):167–170
- Hughes SK, Nilsson DE, Boyer RS, et al. Neurodevelopmental outcome for extended cold water drowning: a longitudinal case study. *J Int Neuropsychol Soc* 2002;8(04):588–595
- Kieboom JK, Verkade HJ, Burgerhof JG, et al. Outcome after resuscitation beyond 30 minutes in drowned children with cardiac arrest and hypothermia: Dutch nationwide retrospective cohort study. *BMJ* 2015;350:h418 10.1136/bmj.h418
- Kruus S, Bergström L, Suutarinen T, Hyvönen R. The prognosis of near-drowned children. *Acta Paediatr Scand* 1979;68(03):315–322
- Pearn J. Neurological and psychometric studies in children surviving freshwater immersion accidents. *Lancet* 1977;1(8001):7–9
- Suominen PK, Korpela RE, Silfvast TG, Olkkola KT. Does water temperature affect outcome of nearly drowned children. *Resuscitation* 1997;35(02):111–115
- Suominen PK, Vähätalo R, Sintonen H, Haverinen A, Roine RP. Health-related quality of life after a drowning incident as a child. *Resuscitation* 2011;82(10):1318–1322
- Suominen PK, Vallila NH, Hartikainen LM, Sairanen HI, Korpela RE. Outcome of drowned hypothermic children with cardiac arrest treated with cardiopulmonary bypass. *Acta Anaesthesiol Scand* 2010;54(10):1276–1281
- Multi-Society Task Force on PVS. Medical aspects of the persistent vegetative state (1). *N Engl J Med* 1994a;330(21):1499–1508
- Multi-Society Task Force on PVS. Medical aspects of the persistent vegetative state (2). *N Engl J Med* 1994b;330(22):1572–1579
- Heindl UT, Laub MC. Outcome of persistent vegetative state following hypoxic or traumatic brain injury in children and adolescents. *Neuropediatrics* 1996;27(02):94–100
- Giovannetti AM, Pagani M, Sattin D, et al. Children in vegetative state and minimally conscious state: patients' condition and caregivers' burden. *ScientificWorldJournal* 2012;2012:232149 Doi: 10.1100/2012/232149
- Giacino JT, Katz DI, Schiff ND, et al. Comprehensive systematic review update summary: disorders of consciousness: report of the guideline development, dissemination, and implementation subcommittee of the American Academy of Neurology; the American Congress of Rehabilitation Medicine; and the National Institute on Disability, independent living, and rehabilitation research. *Arch Phys Med Rehabil* 2018;99(09):1710–1719
- Kluger G, Romein E, Hessenauer M, Pringsheim M, Berweck S. Pediatric Klüver-Bucy syndrome: report of two cases and review of the literature. *Neuropediatrics* 2018;49(05):353–354
- Kaplan EL, Meier P. Nonparametric estimation from incomplete observations. *J Am Stat Assoc* 1958;53(282):457–481
- Sachdeva RC. Near drowning. *Crit Care Clin* 1999;15(02):281–296
- Waugh JH, O'Callaghan MJ, Pitt WR. Prognostic factors and long-term outcomes for children who have nearly drowned. *Med J Aust* 1994;161(10):594–595, 598–599
- Howell K, Grill E, Klein AM, Straube A, Bender A. Rehabilitation outcome of anoxic-ischaemic encephalopathy survivors with prolonged disorders of consciousness. *Resuscitation* 2013;84(10):1409–1415
- Kurz R; Ethics working group of Confederation of the European Specialists of Paediatrics. Decision making in extreme situations involving children: withholding or withdrawal of life supporting treatment in paediatric care. Statement of the ethics working group of the Confederation of the European Specialists of Paediatrics (CESP). *Eur J Pediatr* 2001;160(04):214–216

Appendix 1

Calculation of the probability to have further children in the general German population

Y_{bv} = Year of birth of the victim; Y_{acc} = year of the accident; n = position of the victim in the order of siblings

For families with no children born between birth of the victim and the accident, the probability was calculated as:

$P = [(\text{number of } (n + 1^{\text{st}}) \text{ children born in } (Y_{acc} + 1 \text{ year}) \text{ with a distance between } n + 1^{\text{st}} \text{ and } n^{\text{th}} \text{ child of } (Y_{acc} - Y_{bv} + 1 \text{ year})) + (\text{number of } (n + 1^{\text{st}}) \text{ children born in } (Y_{acc} + 2 \text{ years}) \text{ with a distance between } n + 1^{\text{st}} \text{ and } n^{\text{th}} \text{ child of } (Y_{acc} - Y_{bv} + 2 \text{ years}) + \dots)] / (\text{number of } n^{\text{th}} \text{ children born in } Y_{bv})$

For families with one child born between birth of the victim and the accident, the birth year of this younger sibling (Y_{bys}) was arbitrarily set to ($Y_{bv} + 2$ years), since these data had not been obtained, and 2 years was, in the median year of the accident 1997, the most frequently observed distance between two subsequent children in the same family. Here, the probability was calculated as:

$P = [\text{number of } (n + 2^{\text{nd}}) \text{ children born in } (Y_{acc} + 1 \text{ year}) \text{ with a distance between } n + 2^{\text{nd}} \text{ and } n + 1^{\text{st}} \text{ child of } (Y_{acc} - Y_{bys} + 1)] + [\text{number of } (n + 2^{\text{nd}}) \text{ children born in } (Y_{acc} + 2 \text{ years}) \text{ with a distance between } n + 2^{\text{nd}} \text{ and } n + 1^{\text{st}} \text{ child of } (Y_{acc} - Y_{bys} + 2 \text{ years}) + \dots] / (\text{number of } n^{\text{th}} \text{ children born in } Y_{bv})$

In none of the participating families, more than one child was born between birth of the victim and the accident.

*continued until either ($Y_{acc} - Y_{bv} + i$ years) or ($Y_a - Y_{bys} + i$ years) exceeds 9 years** – or until ($Y_{acc} - Y_{bv} + i$ years) or ($Y_{acc} - Y_{bys} + i$ years) reaches the year of the interview.

**This was done since the Statistisches Bundesamt does not provide data in sufficient detail for (the very few) children born 10 years or more after the previously born child in the family. For children born in 1997 (the median year of the accidents), this was true for only 19,472 of 364,469 second and further children (5.3%); hence, our algorithm leads to a marginal underestimation of the calculated probabilities.