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RESEARCH ARTICLE

Psychogenic non-epileptic (functional) seizures in adults with intellectual disability and epilepsy: A matched case–control study

Iris E. M. Kloosterman[1] | **Alexandra I. Haenen**[2] | **Esther L. G. E. Poortvliet-Koedam**[1,3] | **Richard H. C. Lazeron**[4] | **Helenius J. Schelhaas**[3] | **Jans S. van Ool**[2]

[1]Residential Care Centre, Stichting Epilepsie Instellingen Nederland (SEIN), Heemstede, The Netherlands

[2]Department of Residential Care, Epilepsy Centre Kempenhaeghe, Heeze, The Netherlands

[3]Epilepsy Centre, Stichting Epilepsie Instellingen Nederland (SEIN), Heemstede, The Netherlands

[4]Academic Center of Epileptology Kempenhaeghe, Heeze, The Netherlands

Correspondence
Iris E. M. Kloosterman, Residential Care Centre, Stichting Epilepsie Instellingen Nederland (SEIN), Spieringweg 801, 2142 ED Cruquius, Heemstede, The Netherlands.
Email: iriskloosterman@gmail.com

Abstract

Objective: To describe the characteristics of psychogenic non-epileptic (functional) seizures (PNES) in adults with epilepsy and intellectual disability (ID) and to establish differences and risk factors regarding psychosocial functioning between individuals with and without PNES.

Methods: Adults with ID and epilepsy living in epilepsy care facilities in The Netherlands were screened for PNES by a neurologist. A control group consisting of people with epilepsy and ID, without PNES, was matched according to age, sex, and level of ID. Objective data were retrieved retrospectively from clinical notes of the resident. Standardized questionnaires and tests, adjusted for people with ID, were obtained from participants and their nursing staff. Differences were analyzed using paired *t* tests, Wilcoxon signed-rank tests, or McNemar's tests, appropriate for matched case–control studies. Conditional logistic regression identified PNES risk factors.

Results: Five hundred forty individuals were screened, of which 42 had PNES (point prevalence 7.8%). In total, 35 cases and 35 controls gave consent. Proxy reports indicated that PNES impacted daily life in 79% by adjusting the individual's schedule, and caused minor injuries in one-third. Those with PNES were mainly female (69%); had a mild (46%) or moderate (37%) level of ID; showed more symptoms of depression ($p = .024$), anxiety ($p = .030$), self-injurious behavior ($p = .015$); and experienced more negative life events ($p < .001$). Clinically relevant predictors of PNES were the number of negative life events (odds ratio [OR] 1.71, 95% confidence interval [CI] 1.12–2.53) and self-injurious behaviors (OR 5.27, 95% CI .97–28.81).

Significance: Previously, PNES in individuals with ID and epilepsy were described mainly as a reinforced behavioral pattern, due to limited associations with psychiatric disorders. Our results demonstrate that this population does show individual psychosocial vulnerabilities when measured with instruments adjusted for this population, as indicated by proxy reports from daily caregivers.

Viewing PNES as an involuntary response, especially for stress-prone individuals with ID, could reduce stigma and improve treatment.

KEYWORDS
dissociative seizures, functional seizures, negative life events, psychosocial vulnerability

1 | INTRODUCTION

Psychogenic non-epileptic seizures (PNES), also known as functional seizures or dissociative seizures, are seizures characterized by altered behavioral, sensory, motor, or cognitive function, without a known neurological condition. According to the fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5), PNES are classified as Functional Neurologic Disorder (FND, or previously: conversion disorder) when the seizure is incompatible with a known medical or psychological disorder and people suffer from significant distress.[1] The term PNES is used here, acknowledging it is a manifestation of FND. A clinical assessment should exclude a neurological disease for diagnosis of PNES, such as epilepsy, although PNES and epilepsy are not mutually exclusive: almost one fourth of people with epilepsy have PNES, and 12% of people with PNES have epilepsy.[2] The highest level of diagnostic certainty for PNES ("documented PNES") is reached, therefore, by combining a history consistent with PNES and video-electroencephalography (vEEG) monitoring of a seizure that does not show epileptiform abnormalities as seen during an epileptic seizure.[3]

Intellectual disability (ID) has been mentioned as a risk factor for developing PNES.[4] Still, research into this population is lacking,[5] and the clinical relevance of PNES to the ID population might be underestimated.[6] According to the DSM-5, ID is classified based on a disability in domains of intellectual (IQ <70), social, and adaptive functioning[1] starting from childhood. People with ID make up to 3% of the general population and are more vulnerable to psychological, behavioral, and neurological disorders, such as epilepsy, than those without ID.[7] Moreover, people with ID are more often affected by both PNES and epilepsy,[8–10] frequently face diagnostic delays for PNES,[8] show a high proportion of medical encounters, more often have persistent PNES,[6] and are at additional risk of misdiagnosis, for example, due to an atypical manifestation of seizures, stereotypic movements or frequent comorbid behavioral problems, and limited abilities to communicate.[11] A meta-analysis showed that epilepsy in ID is incorrectly diagnosed in approximately one third of cases,[12] of whom 13%–50% eventually were diagnosed with PNES.[13,14] Results suggest that, although this sample consisted of children and adolescents, PNES could be underdiagnosed in people with ID and that this group might be receiving unnecessary anti-seizure medication.

Key points

- Proxy reports identified more anxiety, depression, self-injurious behavior, and negative life events in our cases with psychogenic non-epileptic (functional) seizures (PNES), epilepsy, and intellectual disability (ID) compared to controls.
- Facing multiple mildly stressful events is a specific risk factor for PNES in this population, presumably because of innate vulnerabilities in dealing with stress.
- The etiology of PNES in ID seems similar to people without ID, in which psychosocial factors and individual vulnerabilities should be assessed.
- When individuals with epilepsy and ID experience multiple life events and show self-injurious behavior, the possibility of PNES should be considered.

Multiple psychological risk factors for PNES in individuals without ID are known, such as adverse childhood experiences or trauma in adult life.[15,16] Evidence regarding risk factors in individuals with ID, such as psychopathology, is less clear, despite this population being at increased risk of exposure to adverse life events.[17,18] This ambiguity has been explained previously by the possibility of a reporting bias.[4] Due to the often-limited sample sizes in previous studies, data from earlier research[5] have been expanded upon in the current study to gain deeper insights into this specific subgroup of individuals with ID, epilepsy, and PNES. This case–controlled study, therefore, aims to (1) describe (clinical) characteristics of PNES in adults with ID and epilepsy; (2) compare epilepsy severity and psychological and behavioral characteristics between those with PNES and a matched control group without PNES by proxy reports; and (3) identify possible (psychopathological) risk factors for PNES, all in people with epilepsy and ID.

2 | METHODS

2.1 | Participants

This study is an extension of a previous one[5] in order to validate results in a larger sample size. All adults with ID living at Kempenhaeghe or Stichting Epilepsie Instellingen Nederland (SEIN), both residential epilepsy care facilities in The Netherlands, were screened and evaluated for (previous) PNES diagnosis by an experienced neurologist/epileptologist in electronic medical records between January 2014 and December 2016 (Kempenhaeghe) and between August 2018 and July 2020 (SEIN). Because the highest level of diagnostic certainty for PNES was not always achieved, the neurologist/epileptologist classified cases with a lower level of certainty (i.e., "clinically established", "probable," or "possible") based on existing criteria.[3] A control group consisting of adults with epilepsy and ID, without PNES, was matched according to age, sex, and level of ID to mitigate potential biases in the outcome measures, and randomly selected from all eligible matches. Only patients who met the following criteria were included: impaired intellectual functioning (IQ <70), age ≥18 years, and diagnosis of epilepsy and PNES following evaluation by a neurologist from medical charts and, when necessary, other medical specialists. Patients with PNES must have had more than one seizure-like event in the past 2 years.

2.2 | Procedure and instruments

This observational case-controlled study is part of the TRIANGLE study (The Relation between epilepsy, ID, And Neuropsychiatric comorbidities in a Group of patients in Long-term care for Epilepsy). TRIANGLE was approved by the local ethical committee of Kempenhaeghe (No. 15.01). All subjects or legal representatives (if appropriate) provided consent for the study. For an overview of instruments, see Table 1. A detailed written description is in a previous publication.[5] Data on seizure frequency were extracted from electronic patient records spanning the past year, where professional caregivers document daily observations of seizure type and frequency. Neurologists provide detailed descriptions of seizure types, which are recorded in the medical records.

2.3 | Analyses

Analyses were performed using the SPSS package, version 29.0. A power analysis for a two-tailed paired-samples *t* test or Wilcoxon signed-rank test indicated that the minimum sample size to yield a statistical power of at least .8 with an alpha of .05 and a medium effect size ($d = .5$) is 35. Clinical characteristics of PNES are reported. To avoid possible bias, analyses were not performed when more than 10% of the data were missing. The correlation between the frequency of PNES and epileptic seizures in the past year was examined using Spearman's rank correlation analysis. As neither variable met the criteria for a normal distribution, a log-transformation was performed before the analysis.

Differences between subjects with PNES and the control group were analyzed with statistical analyses appropriate for matched case–control studies,[28] that is, paired *t* test for continuous variables or Wilcoxon signed-rank as non-parametric alternative test, and McNemar's test for dichotomous variables.

Because PNES is not always diagnosed with the highest level of certainty, post hoc analyses were performed to examine possible bias regarding the output per subgroup, by splitting and comparing output according to the highest levels of certainty ("clinically established" and "documented") vs the lowest levels of certainty ("probable" and "possible").[3] To disclose whether specific characteristics are significantly associated with the level of certainty (i.e., possibly explanatory for whether or not video and/or v-EEG was performed), subgroups were compared by independent sample *t* test, or Mann–Whitney when assumptions were not met, and by chi-square test for categorical variables. The ordinal IQ is recoded into a dichotomous variable (mild ID vs moderate, severe, and profound ID) because of the expected small cell frequencies.

A conditional logistic regression was performed to identify risk factors that specifically contribute to having PNES. To avoid type-1 errors, the model was based only on variables significantly associated with PNES in the previous analysis. Because of the small sample size per category, multicollinearity was checked by Pearson correlation for linear variables and by Kruskal–Wallis for categorical data. Likelihood ratio statistics were used to determine the effect sizes of the predictor variables. All analyses were conducted two-tailed, with *p*-values <.05 considered statistically significant.

3 | RESULTS

Our screening of 540 eligible patients yielded 42 residents with PNES (8%). Seven individuals did not consent to the study, resulting in 35 participating cases. After matching, 70 participants were included. The age of cases ($n = 35$) ranged from 19 to 74 years (mean = 46.0 years, $SD = 17.1$) and did not differ from the controls (mean difference = .6,

TABLE 1 Overview of instruments, measurements, and informants.

Instrument	Measurement	Reference
Self-report		
WAIS-IV, abbreviated version	Level of ID (conceptual) mild–moderate, according to DSM-5 (American Psychiatric Association, 2013)	van Ool, Hurks[19]
PPVT-III	Level of ID (conceptual), profound (American Psychiatric Association, 2013)	Dunn and Dunn[20]
Informant: individuals' professional caregiver		
Vineland-II, socialization and daily living skills	Level of ID (social and practical) according to DSM-5 (American Psychiatric Association, 2013)	Sparrow, Cicchetti[21]
Questionnaire about PNES	PNES characteristics: objective (e.g., frequency, time and location, and injuries as a result of PNES) and subjective (e.g., suspected triggers, impact on daily life and how generally is responded to PNES)	van Ool, Haenen[5]
ADAMS	Anxiety and depressive symptoms, higher scores reflecting more severe symptoms	Hermans, Jelluma,[22] Hermans and Evenhuis[23]
BPI	Behavioral problems, higher scores reflecting more severe behavior	Rojahn, Matson[24] Dumont, Kroes[25]
CLE	Negative life events, higher scores reflecting more negative life events	Hermans and Evenhuis[26]
Source: Electronic patient records		
Chart reviews	Age, sex, epilepsy characteristics, number and type of anti-seizure medication and psychotropic drugs, psychiatric history	
EPIEK	Severity of epilepsy, based on seizure frequency, number of anti-epileptic drugs, use of emergency anti-epileptic drugs, use of protective measures for epilepsy, and adjustments in the subject's daily schedule after a seizure, leading to a score from 0 to 10 (higher score indicating a more severe form of epilepsy)	van Blarikom, Tan[27]

Abbreviations: ADAMS, Anxiety, Depression and Mood Scale; BPI, Behavior Problem Inventory; CLE, checklist life events; EPIEK, Epilepsy Impact Scale Kempenhaeghe; ID, intellectual disability; PNES, psychogenic non-epileptic seizures; PPVT-III, Picture Peabody Vocabulary Test—Third Edition; WAIS-IV, Wechsler Adult Intelligence scale— Fourth Edition.

$SD = 3.46$, $p = .312$). Most were female (69%) and had a mild (46%) or moderate (37%) ID. All subjects were taking anti-epileptic drugs.

3.1 | PNES characteristics

PNES were classified according to the highest level of certainty in 57% of the cases (37% by vEEG and 20% by video). In the remaining cases, PNES diagnosis was classified as "probable" (29%) and "possible" (14%). In 69% of the subjects, the semiology of PNES showed similarities with the typical epileptic seizure of the subject. Usually, tonic-like, tonic–clonic-like, and absence-like seizures were presented. PNES occurred at various times of the day (83%) and locations (80%). Data regarding PNES onset was missing in nine cases (36%) and was not further analyzed. The frequency of PNES was mostly yearly (37%), hereafter monthly (26%), and weekly (20%). Six subjects (17%) did not show PNES the previous year, but presented them in the year before. Medical records showed that epileptic seizures were recorded more frequently than PNES in 63% of cases. There was no correlation between the frequency of PNES and epileptic seizures (Spearman's $r = -.257$, $p = .136$). A psychiatrist was involved in the clinical care of more than half of the subjects (54%), and more than a third (37%) had a comorbid psychiatric diagnosis. The daily use of psychotropic medication for the treatment of psychiatric, psychological, or behavioral problems was common (40%). According to the nursing staff, triggers for PNES were identified in the majority (85%). These triggers involved stress, negative mood, unexpected events, (over)demanding situations, and overstimulation. Minor injuries were reported in 27% and severe injuries in 9%. The PNES impacted daily life in 79% of cases, for example, by adjusting their daily schedule. The nursing staff reported responding to the PNES mostly by attending the individual and trying to start a conversation (41%), ignoring the PNES (23%), distracting the individual (18%), or acting as they would to an epileptic seizure (18%).

3.2 | PNES vs controls

Differences in epilepsy-related and psychological characteristics between the cases and control group are presented in Table 2. Of the continuous variables, the severity of aggressive and self-injurious behavior did not meet the normality assumption. Both groups were using anti-seizure medication daily. Epilepsy in both groups was severe, with a median severity score of nearly 7 of 10 in the PNES group and 6 in the control group (not statistically significant). Most of the participants had weekly seizures (49% of the cases vs 40% of the controls), and only two cases (6%) and seven controls (20%) were seizure-free last year. The cases differed from the control group concerning psychological characteristics. Paired *t* tests indicated that the group with PNES had significantly more anxiety symptoms (mean difference = 2.1, t (34) = 2.37, $p = .024$, medium effect size), depressive symptoms (mean difference = 4.2, t (34) = 2.27, $p = .030$, medium effect size), showed significantly more self-injurious behavior ($Z = -2.43$, $p = .015$, large effect size), and encountered substantially more negative life events in the past year (mean difference = 2.26, t (34) = 3.97, $p < .001$, large effect size), such as major injuries, frequent moving or change in nursing staff, severe illness, or death of a friend or family member. A higher percentage of cases used psychotropic drugs (40.0% vs 31.4%), had a comorbid mental disorder according to the DSM-5[1] (37.1% vs 28.6%; e.g., autism spectrum disorder, depression), and more often, a psychiatrist was involved (54.3% vs 37.1), although these differences did not reach statistical significance. Although not statistically significant, a discrepancy between the two domains of adaptive deficits within the ID classification (according to DSM-5) was found between cases and control groups ($p = .077$); Cohen's *g* suggests that the observed difference is large.

Post hoc analyses were performed to compare the results between participants with the highest level of certainty, $n = 20$, vs the lowest level of certainty, $n = 15$, by splitting the data in subgroups. Regarding the variables significantly associated with PNES, the variables of depression, negative life events, and self-injurious behavior remained significantly associated with PNES in the highest level of certainty subgroup. The subgroup in which no video/vEEG was performed, had a significant lower level of intellectual functioning compared to the other group (13 vs 3 with a mild ID, respectively, χ^2 (1) = 6.994, $p = .008$, large effect size) and were significantly older (53 vs 40 years, $U = 75$, $n_1 = 20$, $n_2 = 15$, $p = .012$, large effect size). The frequency of PNES in the lowest level of certainty group was lower (mean frequency = 54.9 vs 27.9 seizures a year), although this difference between subgroups was not significant ($U = 108$, $n_1 = 20$, $n_2 = 15$, $p = .169$, medium effect size), possibly due to a lack of power (20%).

TABLE 2 Differences between cases with PNES and controls.

Characteristics	Cases with PNES		Controls		*p* Value	Effect size
Epilepsy severity	*M* = 6.29	*SD* = 2.02	*M* = 6.09	*SD* = 2.82	.724[a]	.1[d]
Number of ASMs	*M* = 2.91	*SD* = .92	*M* = 2.80	*SD* = .99	.594[a]	.1[d]
Depressive symptoms	*M* = 14.06	*SD* = 7.73	*M* = 9.89	*SD* = 7.51	.030[a]	.6[d]
Anxiety symptoms	*M* = 7.83	*SD* = 4.85	*M* = 5.69	*SD* = 4.26	.024[a]	.5[d]
Social avoidance symptoms	*M* = 6.29	*SD* = 5.15	*M* = 4.54	*SD* = 4.56	.109[a]	.4[d]
Negative life events	*M* = 5.89	*SD* = 2.68	*M* = 3.63	*SD* = 3.08	<.001[a]	.8[d]
Aggressive behavior	*Mdn* = 2	IQR = 0–6	*Mdn* = 0	IQR = 0–4	.210[b]	.4[d]
Self-injurious behavior	*Mdn* = 0	IQR = 0–2	*Mdn* = 0	IQR = 0–0	.015[b]	.9[d]
Psychiatric diagnosis	37.1%		28.6%		.607[c]	.2[e]
Daily use PTDs	40.0%		31.4%		.629[c]	.2[e]
Psychiatrist involved	54.3%		17.0%		.263[c]	.3[e]
ID domain discrepancy*	68.6%		45.7%		.077[c]	.5[e]

Abbreviations: ASM, anti-seizure medication; ID, intellectual disability; IQR, interquartile range; M, mean; Mdn, median; PNES, psychogenic non-epileptic seizures; PTD, psychotropic drugs; SD, standard deviation.

[a]Paired *t* test.

[b]Wilcoxon signed-rank test.

[c]McNemar's test.

[d]Cohen's *d* (≤.4 small effect; .5–.7 medium effect; ≥.8 large effect).

[e]Cohen's *g* (≤.15 small effect; .16–.24 medium effect; ≥.25 large effect).[29]

*A discrepancy indicated a significant intra-individual difference between two of the three domains of adaptive functioning (conceptual, social, and practical domain).

3.3 | Risk factors PNES

Because the variable of self-injurious behavior included an outlier, the variable was recoded into a binary variable (self-injurious behavior yes/no) and added to the model. Conditional logistic regression analysis revealed a significant prediction model (χ^2 (4, 35) = 19.655, p = .001). The amount of negative life events is a predictor of PNES (odds ratio [OR] 1.71, 95% confidence interval [CI] 1.12–2.53, p = .007). Self-injurious behavior was not statistically significant, but it is considered clinically relevant with an OR of 5.27 (95% CI .97–28.81, p = .055).

4 | DISCUSSION

We investigated the characteristics of people with PNES, epilepsy, and ID in Dutch residential care facilities using informant reports; compared them to controls; and identified risk factors associated with PNES. The established impact on daily life (in 79%) and number of injuries (27% small and 9% severe) in the cases emphasize the relevance of investigating PNES in this under-investigated subgroup. Our sample findings aligned with previous research on people without ID, both in terms of gender[4,30] and the presence of psychosocial vulnerabilities.[4,16]

Our sample of cases had elevated levels of anxiety and depression, showed significantly more self-injurious behavior, and experienced significantly more negative life events compared to controls. Research in the ID and PNES population is less conclusive about the existence of intrapsychic factors, which often are presumed not to be involved in this subgroup.[8,31] In these studies it is unclear how data on psychiatric comorbidity were collected and whether instruments were suitable to detect psychological symptoms in this specific group with ID. Psychiatric symptoms in people with ID are challenging to identify and are reported less frequently than in those without ID,[4] due to diagnostic overshadowing or an atypical presentation of symptoms, for instance, in behavioral problems and self-harm.[32,33] One study reported that although only 2% of their sample with ID had a posttraumatic stress disorder (PTSD) diagnosis, up to 40% of participants met DSM-5 PTSD criteria after screening with an appropriate instrument.[34] Our method consisted of instruments designed specifically for people with ID, which are likely to be more sensitive to detect psychological symptoms in this group.

In our study, the number of negative life events and self-injurious behaviors were considered risk factors for PNES. The importance of negative life events is stated previously,[35] by recognizing that next to PTSD or chronic stress, also mildly or moderately stressful experiences contribute to PNES in individuals with a high biological susceptibility to stress. The questionnaire we used included items like "change of staff" and "moving to different room/building," which are considered mildly or moderately stressful experiences, although frequently faced by institutionalized people with IDs. People with an ID are at increased risk of exposure to adverse life events[17,18] and are more prone to stress due to biological and psychosocial vulnerabilities, for example, aberrations in brain development, having limited coping strategies to deal with the many negative life events they encounter, and difficulties interpreting their emotional states and expressing themselves. People with ID also often have a limited social network, which is associated with psychopathology.[36–38] Another study found that for every additional stressor in adults with ID, the likelihood of having a psychological diagnosis increased by 20% and behavioral problems by 19%.[39] Although no causal conclusions can be drawn from this study, these results suggest that self-injurious behavior and PNES could be a manifestation of heightened emotional distress in individuals with ID and epilepsy,[4,40] who already have biological and psychosocial vulnerabilities in dealing with stress.

Possible limitations are the study's retrospective nature; the (small) sample size from which results may not be generalized to populations not living in residential care facilities or to people without epilepsy; a potential attention bias from the individuals' professional caregiver specifically for the cases; and the difference in time of data collection between the two sites. However, comparable results were obtained.[5] The relatively few diagnostics by the gold standard of PNES highlight the diagnostic challenges in this population, even for subjects living in residential care facilities for epilepsy. A previous study reached the gold standard in less than one third of people with ID.[6] In our sample, the group in which video/vEEG was not performed had a more severe ID and a higher age, and although not statistically significant, this group had a lower frequency of PNES. Undergoing vEEG might be challenging for these individuals, and could have influenced the chance and feasibility of a recording being carried out. Most importantly, subgroup comparisons between the high and low levels of certainty in diagnosing PNES within our cases revealed comparable results in relation to our main findings.

Our study stresses the importance of considering the role of negative life events and stress, as reported by caregivers, in the etiology of PNES, and also for people with ID. Previously, a different mechanism was proposed in a sub-group of people with PNES and ID, in which no intrapsychic factor was suspected.[8,31] Suggesting that this subgroup unconsciously "simulates" seizures at times,

for example, to avoid (high) demands or unpleasant situations, implies a voluntary element and could be stigmatizing for the subgroup of people with ID,[41] leading to inappropriate treatment, including unwanted caregiver or health care professionals responses that could perpetuate PNES.[4] Adequate information should be given about PNES and environmental factors; biological and psychological vulnerabilities should be assessed with appropriate instruments for ID. An example of a useful explanatory model is the theoretical model of five factors involved in PNES.[15] This model includes five different levels, from psychological, biological, shaping, triggering, and prolongation factors, which could all contribute to PNES. Even when presumed to be mild, the number of negative life events could be an important (perpetuating) factor that needs considerable attention and treatment in this subgroup. Conversely, when individuals with ID and epilepsy encounter many negative life events or show self-injurious behavior, the possibility of PNES should be considered. Future work should be prospective and focus on suitable diagnostics, for example, instruments adjusted for people with ID and combining self-report (when applicable) and proxy reporting, and investigate treatment options for people with ID, PNES, and epilepsy.

AUTHOR CONTRIBUTIONS

Iris E. M. Kloosterman: investigation, data curation (Stichting Epilepsie Instellingen Nederland (SEIN) sample + combined sample), formal analysis, draft writing, review and editing, and conceptualization. Alexandra I. Haenen: draft writing and review and editing. Esther L. G. E. Poortvliet-Koedam: investigation (SEIN sample) and review and editing. Richard H. C. Lazeron: investigation (Kempenhaeghe sample) and review and editing. Helenius J. Schelhaas: supervision and review and editing. Jans S. van Ool: conceptualization (lead), investigation and data curation (Kempenhaeghe sample), draft writing, and review and editing.

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CONFLICT OF INTEREST STATEMENT

None of the authors has any conflict of interest to disclose.

DATA AVAILABILITY STATEMENT

Data are available on request from the authors due to privacy/ethical restrictions.

ETHICS STATEMENT

We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this report is consistent with those guidelines.

ORCID

Iris E. M. Kloosterman https://orcid.org/0009-0006-6690-0599
Alexandra I. Haenen https://orcid.org/0009-0005-2724-3242
Richard H. C. Lazeron https://orcid.org/0000-0001-5570-8872
Helenius J. Schelhaas https://orcid.org/0000-0002-8594-4478
Jans S. van Ool https://orcid.org/0000-0003-0645-4452

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