

Parenting stress and perceived stigma in mothers of young children with epilepsy: A case-control study

Colin Reilly^{1,2}

Patricia Atkinson³

Ayesha Memon³

Chloe Jones¹

Lyvia Dabydeen¹

Krishna B Das^{2,4}

Christopher Gillberg^{1,2,5}

Brian GR Neville^{1,2}

J Helen Cross^{1,2,4}

Rod C Scott^{2,4,6}

Affiliations

¹Research Department, Young Epilepsy, Lingfield, Surrey, RH7 6PW, UK.

²UCL Great Ormond Street Institute of Child Health (ICH), 30 Guilford Street London WC1N 1EH UK.

³Child Development Centre, Crawley Hospital, West Green Drive, Crawley, RH11 7DH, West Sussex, UK.

⁴Great Ormond Street Hospital for Children NHS Trust, Great Ormond Street, London WC1N 3JH, UK.

⁵Gillberg Neuropsychiatry Centre, University of Gothenburg, Kungsgatan 12, Gothenburg, Sweden.

⁶Department of Neurological Sciences, University of Vermont College of Medicine, Burlington, Vermont, U.S.A

Correspondence to: Colin Reilly, Research Department, Young Epilepsy, Lingfield, Surrey, RH7 6PW, UK. creilly@youngepilepsy.org.uk 01342 832243

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Abstract

The aim was to provide data on parenting stress and perceived stigma in mothers (n=47) of young children with epilepsy, and to compare findings with those of mothers (n=48) of developmental, age and gender matched children with non-epilepsy related neurodisability (neurological and/or neurodevelopmental concerns). The mothers of young children (1-7 years) with epilepsy and mothers of children with neurodisability in a defined geographical area of the UK, completed the Parenting Stress Index-4th Edition (PSI-4) and a measure of perceived stigma. Factors associated with parenting stress and stigma were analyzed using linear regression. 38% of mothers of children with epilepsy scored in the at-risk range (>85th percentile) on the Total Stress score of the PSI-4 (Neurodisability 21%) (p=0.06). Significantly more mothers of children with epilepsy scored in the at-risk range on the Parent-Child Dysfunctional Interaction subscale than mothers of children with neurodisability (Epilepsy 45% vs. Neurodisability 21%; p=0.01), but not on the Parental Distress subscale (Epilepsy 32% vs. Neurodisability 23%; p=0.33) or Difficult Child (Epilepsy 57% vs. Neurodisability 46%; p=0.26) subscales. There was not a statistically significant difference on perceived stigma between mothers in both groups (p=0.51). Factors significantly associated with increased parenting stress in the epilepsy group were child behavior difficulties (p<0.001) and maternal sleep difficulties (p=0.02). Lower child developmental level was the only factor independently associated with increased stigma in the epilepsy group (p=0.08). Mothers of young children with epilepsy report high levels of parenting stress and higher levels of difficulties with parent-child interaction compared to mothers of children with non-epilepsy related neurodisability. Parenting stress and stigma in epilepsy were not associated with epilepsy factors. Efforts at reducing parenting stress and stigma should focus on interventions targeting child development and maternal sleep.

Highlights

- 38% of mothers of children with epilepsy had significant parenting stress
- Parenting stress was associated with child behavior and parental sleep
- Perceived stigma was related to developmental delays and not epilepsy factors

1. Introduction

Childhood onset epilepsy can have a very significant impact on the family system with mothers in particular being at risk for suboptimal functioning. Mothers of children with epilepsy experience elevated rates of anxiety and depression^{1,2} and frequently have problems with sleep and fatigue³. In addition to increased difficulties in maternal functioning, epilepsy has long been associated with stigma that is often greater than other chronic illnesses and on a par with mental health problems⁴.

Epilepsy in early childhood is often characterised by difficult to treat epileptic seizures and has a very high association with behavioral and developmental difficulties⁵ with a high frequency of developmental and epileptic encephalopathies^{6,7}. These difficulties often are not identified but have a very significant impact on health related quality of life^{8,9} and add significantly to the economic cost of the condition¹⁰.

Parenting stress is stress directly related to the role of being a parent and is an important factor when considering family functioning¹¹. In the context of childhood illness, increased parenting stress is associated with poorer psychological adjustment in caregivers and also their children¹². Additionally, parenting stress may also affect child health related outcomes as it could potentially interfere with management of the child's condition¹². Difficulties with parenting stress in childhood epilepsy have been shown to be associated with increased child behavioral-emotional difficulties^{13,14} and symptoms of parental depression¹⁴ underlining the need to identify and provide support for these problems.

Stigma refers to the loss of status that arises from being in possession of an attribute, for example a health condition, that has been culturally defined as “undesirably different” and also

as “deeply discrediting”¹⁵. A person’s perceptions of stigma or “perceived stigma” can result from previous negative experience or from anticipation of future negative experience. Perceived stigma in childhood epilepsy has been associated with child mental health/behavioral problems^{16,17} and impairments in child quality of life^{18,19}. The reduction of stigma in epilepsy has been identified as a priority by the ILAE and IBE²⁰.

It is essential to understand the psychosocial impact of epilepsy to guide possible intervention efforts that are likely to improve quality of life for mothers and their children. Comparing mothers of children with epilepsy to mothers of other children with a similar level of developmental difficulties allows us to better understand the specific role of seizures in parenting stress and stigma. The Sussex Early Epilepsy and Neurobehaviour (SEEN) study is a population-based study focussing on behavior and development in young children with epilepsy as well as functioning in parents. The aim of this paper was to provide population-based data on the parenting stress and perceived stigma in mothers of young children with epilepsy. A secondary aim was to use a case-control design to compare findings with those of mothers of developmental, age and gender matched children with non-epilepsy related neurodisability (neurodevelopmental and/or neurological difficulties). A final aim was to provide data on associations between variables related to child epilepsy, child behavior/development and parental socioeconomic status with maternal parenting stress and perceived stigma in young children with epilepsy.

2. Methods

2.1 Recruitment in the SEEN study

Recruitment in the SEEN study has been previously described²¹. Children with epilepsy (a history of two or more unprovoked seizures more than 24 hours apart), born between 2008 and

2014, who were resident in defined geographical districts of the south of the United Kingdom between September 30th 2014, and February 29th 2016 were included in the study. Children needed to be at least one year of age during the study period in order to be included. The prevalence of lifetime epilepsy (a history of 2 or more unprovoked epileptic seizures) in the study area was calculated by using the 2011 UK census population data of 1 to 7-year-olds (19,393) provided by the Office of National Statistics (2011 UK census total population 217,006). With respect to ethnic make-up, the area is similar to the UK average (study area: white 87%, non-white 13%, United Kingdom: white 88% non-white 12%: Office of National Statistics, UK).

Once a child with epilepsy was enrolled in the study, the parents of children with similar attributes (age, gender and estimated developmental level) without epilepsy attending the same clinics in the study area were approached by collaborating paediatricians. The children were referred for a neurological/neurodevelopmental concern. Developmental level was estimated based on school/preschool placement (special or mainstream), previous psychological/developmental assessment or clinician judgement. Estimated developmental level was used as we wanted to match the children with neurodisability with the children with epilepsy as close as possible with regard to level of development. Primary referral concerns for the children with neurodisability were classified as 'global development', 'social communication only', or 'motor only'.

2.2 Child assessment

Children in both groups underwent comprehensive psychological assessment in their homes, including measures of global development, sleep and emotional-behavioral functioning between November 1st 2014, and April 30th 2016. Global development was assessed using the

Griffiths Mental Development Scales (GMDS)²² or the Griffiths Mental Development Scales-Extended Revised²³ which yield an overall Developmental Quotient (DQ). Child sleep was assessed using the Child Sleep Habits Questionnaire (CSHQ)²⁴ and child behavior by the Strengths and Difficulties Questionnaire (SDQ)²⁵.

Clinical information on all children was extracted (using a standardised proforma) including data on current AEDs and seizures. Clinical data of the children with epilepsy were reviewed by 2 paediatric neurologists who independently classified seizures (as primarily generalised or focal), and epilepsy syndromes/epilepsies and aetiology/cause proposed by the Task force of the International League Against Epilepsy (ILAE) in 2010²⁶. When the assessors disagreed on the classification, conflicts were resolved by a third rater.

2.3 Assessment of Maternal Functioning

The Parenting Stress Index –Fourth Edition (PSI-4)²⁷ Short Form and a five-item scale assessing parental perception of stigma toward the child²⁸ with epilepsy/neurodisability was part of the research pack given to the mothers at the time of child assessment.

The PSI-4 is a 36-item measure in which parents respond to items within the domains of (1) Parental Stress, (2) Parent–Child Dysfunction Interaction, and (3) Child Difficulty. It is used to consider a parent’s relationship with one of his or her children between the ages of 1 month and 12 years. Participants rate items on a 5-point Likert scale ranging from *Strongly Disagree* (1) to *Strongly Agree* (5). Items across the domains are summed yielding a total parenting stress score in which higher scores reflect greater parenting stress. Scores (i.e. total raw score) between the 85th and 89th percentile are considered ‘high’ and 90th percentile or higher are

considered ‘clinically significant’. The PSI-4 also contains a ‘defensive responding’ scale indicating if respondents have responded to items in a biased manner. A ‘defensive responding’ raw score of 10 or less indicates potential biased responding.

The perceived stigma measure²⁸ contains five items each rated on a scale ranging from *Strongly Disagree* (1) to *Strongly Agree* (5) (All questions are in Supplement 2). A higher score reflects greater perceptions of stigma associated with their child having epilepsy. In the version given to parents of children with neurodisability the word ‘epilepsy’ was replaced by the word ‘disability’.

Mothers also completed the short-form version of the Depression Anxiety Stress Scales (DASS-21)²⁹ a measure of mental health, the Pittsburgh Sleep Quality Index (PSQI)³⁰ a measure of sleep and the Iowa Fatigue Scale (IFS)³¹. Mothers provided information on their highest level of education (categorised as attending full-time education up to 16 years (formal education) or fulltime education beyond 16 years (beyond formal education), total amount of hours worked per week, age in years and employment (in paid employment/not in paid employment). Socioeconomic deprivation status was determined by the Index of Multiple Deprivation (IMD) 2015 rankings (Department of Communities and Local Government, *English indices of deprivation*. Retrieved from <http://imd-by-postcode.opendatacommunities.org/> (Accessed December 15th 2016). Lower scores are associated with lower deprivation.

2.4 Statistical Analysis

Descriptive statistics were used to indicate mean scores on the PSI-4 and stigma scale and also the number of parents scoring in the at-risk range (defined as scoring in the ‘high’ or clinically ‘significant range’ i.e. 85th percentile or higher) on the PSI-4.

Independent t-tests or chi-square analyses/fishers exact tests were used to compare the epilepsy group to the neurodisability group with respect to maternal age, education level, deprivation, employment status, hours worked previous diagnosis of mental health problems and children with respect to age, developmental quotient, child sleep difficulties (Total score on CSHQ) and child behavior difficulties (Total SDQ score).

Chi-square analyses were used to compare the epilepsy group with the neurodisability group with respect to the proportion of mothers in the clinically significant range on the PSI-4 subscales and total score. Independent-sample t-tests were used to compare mothers' PSI-4 symptom scores (i.e. total raw score and subscale raw scores) and total scores on the perceived stigma scores between the epilepsy and neurodisability groups.

Cohen's effect size (d) is shown for the comparisons between group mean scores on PSI-4 total scores and subscales. Effect size magnitudes were interpreted against the criteria suggested by Cohen: trivial (0 to <0.2), small (≥ 0.2 to <0.5), moderate (≥ 0.5 to <0.8), and large (≥ 0.8)³². Cohen's d was determined by calculating the mean difference between the relevant two groups, and then dividing the result by the pooled standard deviation. Holm-Bonferroni corrections were used to adjust for multiple comparisons.

Linear regression modelling was used to identify factors associated with the total score on the PSI-4 and total score on the perceived stigma scale in the epilepsy and the total sample

Principal Component Analysis (PCA) (Varimax rotation with Kaiser normalization) was used to reduce the total number of epilepsy factors. The epilepsy factors included in the PCA analysis

were Aetiology (Genetic/presumed genetic, Structural/metabolic, Unknown/undetermined), Predominant seizure type (focal/generalized), seizure frequency (monthly/more often), status epilepticus (seizures longer than 30 minutes), Polytherapy (monotherapy/polytherapy) and age of seizure onset (in years). The analysis resulted in a 3 factor solution accounting for 65% of the variance (see supplement 1). These three factors were subsequently used in the regression analysis.

Additional child factors included were age at time of assessment (in years), gender, global developmental level (based on Developmental Quotient score from GMDS/GMDS-ER), sleep (total CSHQ score), behavior (total SDQ score) and deprivation. Parent factors included were mothers age (in years), maternal education level (formal education/beyond formal education), hours worked by mother, mental health symptoms (DASS-21 total score), sleep (PSQI total score) and fatigue (Total score on IFS). In the regression analysis for parenting stress stigma was included as a factor and in the regression analysis for perceived stigma parenting stress was included in modelling. In the regression analysis for the total sample additional factors included were epilepsy status (epilepsy/neurodisability). The three epilepsy factors were not included in this regression analysis.

Only factors statistically significant at the $p < 0.10$ level on univariable analysis were included in multivariable modelling.

The alpha level for all analyses was $p < 0.05$. All analyses were performed with IBM SPSS version 23.0 (Armonk, NY, USA).

Ethics Approval

The study was approved by the Westminster Research Ethics Committee and was registered with the collaborating hospital primary care organization: The Sussex Community NHS Trust.

3. Results

The prevalence of epilepsy during the study period was 2.7 per 1000 (1 in 370 95% CI 285-476). During the study period 53 children with epilepsy met eligibility criteria. 49 parents returned an interest form and 48 children subsequently underwent psychological assessment. Of these 48 children with epilepsy, 47 mothers completed the PSI-4 and the perceived stigma scale (see figure 1). In the non-epilepsy related neurodisability group 56 parents returned an interest form and 48 agreed to their child's participation and all 48 mothers completed the PSI-4 and the perceived stigma scale in the neurodisability group (see figure 1). Table 1 shows the characteristics of the mothers in both groups and Table 2 the characteristics of the children in the two groups. In the non-epilepsy related neurodisability group, 35 children had a concern about global development noted, seven had a motor concern without reference to developmental delay noted, and 15 had social communication difficulties noted (six of whom did not have developmental delay mentioned as a referral concern). There were no statistically significant differences between the groups with respect to child age ($p=0.139$), Developmental Quotient ($p=0.626$), CHSQ total ($p=0.092$) or SDQ total ($p=0.494$).

3.1 Maternal Parent characteristics - Between group analyses

No statistically significant differences were found between maternal age ($p=0.230$), maternal hours worked ($p=0.902$), maternal educational level ($p=0.477$), maternal paid employment status ($p=0.765$) or maternal diagnosis of epilepsy ($p=0.213$) in the two groups. However,

significantly more mothers of children with neurodisability had been diagnosed with a mental health condition than mothers in the epilepsy group ($p=0.042$). Additionally mothers in the epilepsy group had significantly higher levels of socioeconomic deprivation ($p=0.045$) than mothers in the neurodisability group.

3.2 At-risk scores on PSI-4

For both groups, the percentages of scores in the at-risk range are shown in figure 2 for the three subscales and the total score of the PSI-4.

The Difficult Child subscale was the subscale where most mothers scored in the at-risk range in both groups. The difference between the two groups of mothers was statistically significant for Parental Child Dysfunction ($p=0.013$; $\chi^2=6.143$) but not for Parental Distress ($p=0.325$; $\chi^2=0.967$), Difficult Child ($p=0.257$; $\chi^2=1.282$) or Total Stress ($p=0.062$; $\chi^2=3.485$). After correction for multiple comparisons the difference between the two groups on Parental Child Dysfunction was no longer significant. Eight of 47 (17%) mothers of children with epilepsy and 10 of 48 (21%) mothers of children with neurodisability scored 10 or less on the Defensive Responding scale of the PSI-4 indicating that they have responded in a biased manner.

3.3 Mean scores on the PSI-4 and stigma scale

Table 3 shows the mean scores on PSI for all parents.

There were no significant differences between groups in any of the subscales and all effect sizes were small. ((Difficult Child subscale ($p=0.183$; $d=0.28$ 95%CI -0.13 to 0.68), Parent-Child Dysfunctional Interaction subscale ($p=0.135$; $d=0.31$ 95% CI -0.10 to 0.71), Parental Distress subscale $p=0.094$; $d=0.35$ 95% CI -0.06 to 0.75), Total Stress subscale ($p=0.087$; $d=0.35$

95% CI -0.05 to 0.76)). There was also not a statistically significant difference between the groups with respect to Defensive Responding ($p= 0.133$; $d= 0.31$ 95% CI -0.09 to 0.72). Mean scores on the stigma scale for each item and total score are in Supplement 2. There was not a statistically significant difference between the two groups on the stigma scale ($p=0.508$; $d= 0.14$ 95% CI -0.27 to 0.54).

3.4 Regression analysis

Supplement 3 shows factors significantly associated with PSI-4 total score and stigma on multivariable analyses in the epilepsy sample. The only factors significantly associated with total stress on the PSI-4 on multivariable analysis in the epilepsy sample were child behavior ($p<0.001$) and maternal sleep ($p=0.015$). Increased child behavior problems and increased maternal sleep difficulties were both independently associated with increased parenting stress. In the total sample the factors associated with total stress on multivariable analysis were child behavior ($p<0.001$), parental mental health difficulties ($p=0.001$) and developmental quotient ($p=0.034$).

The results of regression analysis to identify factors significantly associated with perceived stigma are shown in Supplement 4. The child's developmental level ($p=0.008$) was the only factor significantly associated with perceived stigma on multivariable analysis in the epilepsy sample. Lower developmental level was associated with increased stigma. In the total sample (i.e. epilepsy and neurodisability groups combined) lower developmental quotient was also associated with stigma ($p=0.002$). Additionally increased maternal fatigue was also significantly associated with increased perceived stigma ($p=0.001$). Furthermore, epilepsy status (i.e. epilepsy/not epilepsy) was not a significant predictor of perceived stigma on

multivariable analysis indicating that even after considering other variables there was not a significant difference between the epilepsy and neurodisability group.

4. Discussion

Many mothers of children with epilepsy in the current study reported high levels of parenting stress although not at a significantly different rate to mothers of children with non-epilepsy related neurodisability. Parenting stress in these mothers is primarily a function of behavioral difficulties in the child in conjunction with poor maternal sleep. It is of interest that epilepsy related factors are not independently associated with maternal parenting stress, although mothers of children with epilepsy have more difficulty with parent-child interactions. The lack of difference in stigma between the two groups, and the fact that the association between stigma and developmental level was significant, suggests that stigma in young children with epilepsy is related more to the child's developmental level than the child's seizures. These findings add to our understanding how epilepsy in young children impacts on maternal wellbeing and suggest that broad based epilepsy approaches that target not only seizures but also concentrate on approaches to developmental and behavioral impairments have the greatest potential to maximize the quality of life of families.

The high proportion of mothers scoring in the 'at-risk' range on a measure of parenting stress shows that having a young child with epilepsy and associated neurodevelopmental comorbidities can have a significant negative impact on a mother's perception of her role as a parent. Epilepsy may distort the mother's view of her role due to the potential additional caring responsibilities arising both from seizure management concerns and the child's neurodevelopmental needs. The subscale where mothers reported most difficulties was the

'Difficult Child' subscale where over half of all mothers of children with epilepsy scored in the 'at-risk' range, similar to the frequency in mothers with non-epilepsy related neurodisability. Scores in this range on this subscale indicate that the mother is having difficulties gaining the child's cooperation or perceives the child's behavior as difficult to manage. The only subscale where significantly more mothers of children with epilepsy scored in the at-risk range was the parent child-interaction subscale indicating that more mothers of children with epilepsy experience unmet expectations of the child, feel rejected by the child or have not properly bonded with the child. Parent-child interaction difficulties are associated with an increased risk for both externalizing and internalizing emotional and behavioral problems in the general pediatric population^{33,34}. Therefore, the high levels of difficulties reported by parents and significantly higher levels than the comparison group are concerning and indicate that epilepsy in young children can contribute to suboptimal parent-child-relationships. These suboptimal relationships could have long-term negative consequences for the child's psychological development and wellbeing.

Child behavioral and parental sleep difficulties were both independently associated with parenting stress in mothers of young children with epilepsy and non-epilepsy related neurodisability in the current study. We found no relationship between epilepsy factors and parenting stress in mothers in the current study which has also been noted in other studies which have included a consideration of age of seizure onset, illness severity, and seizure frequency^{13,35}. A significant association between parenting stress and child behavioral difficulties has previously been noted^{13,14} but the significant association between parental sleep and parenting stress has not previously been reported. Parental sleep is often compromised in childhood epilepsy³ and efforts to improve sleep may contribute to reducing parenting stress.

In the current study, no epilepsy variables were associated with stigma and stigma was not greater in the epilepsy group compared to the neurodisability group. This finding suggests that in young children with epilepsy, seizures in and of themselves are not significant contributors to stigma suggesting that minimizing seizure frequency will have a limited impact on reducing stigma. In the epilepsy sample, the child's developmental level was the main driver of stigma. Intellectual disability is associated with significant stigma both in affected individuals but also in mothers of affected individuals³⁶. Child developmental difficulties would appear to influence maternal perceived stigma more than epilepsy perhaps because of the pervasive impact of global development on child functioning.

Given that parenting stress can be associated with negative psychological outcomes for the parent and child and even impact on management of the child's condition¹² maternal parenting stress is an important factor to consider when planning comprehensive management of epilepsy in young children. It has been recommended that mothers of children with epilepsy be screened for depression³⁷. The results of the current study suggest that they also experience high levels of parenting stress and thus asking about stress directly related to the role of parenting will be important. As well as screening for child behavioral difficulties in epilepsy clinics it will also be important to observe parent-child interactions and offer support via parent-training where necessary. The significant associations between child behavior and parenting stress and parental sleep and parenting stress suggest that successful interventions to improve child behavior or parental sleep are likely to impact positively on parenting stress. In relation to reducing stigma in childhood epilepsy, stigma reduction initiatives will need to focus on reducing stigma associated with child neurodevelopmental difficulties as well as stigma associated with epilepsy.

Future research efforts are likely to benefit from a consideration of parenting stress and perceived stigma in fathers as well as mothers. There is also a need to further explore the trajectory and development of parenting stress via qualitative methods (e.g., focus groups, interviews) to better understand the impact of parenting stress on both the child and wider family. Previous research suggests that parenting stress may be highest at, or close to time of diagnosis and reduce over time¹⁹ and factors associated with changes to parenting stress need further exploration. There is a need to understand better the possible difficulties in parent-child interaction in childhood epilepsy and directly focusing on observing mothers and fathers interacting with children may be useful. There is evidence that parenting stress reduces but does not normalize after pediatric epilepsy surgery³⁸ and there is a need to consider the impact of other epilepsy treatments (e.g., ketogenic diet) as well as parental psychoeducational interventions and interventions that address child behavior and developmental difficulties. However, it appears that it is critically important to consider a child's developmental or cognitive level in research on stigma in epilepsy. Interventions that show promise in adults³⁹ should be adapted for the childhood epilepsy population.

There are number of limitations that need to be considered when interpreting the results of the current study. Although our epilepsy sample was population-based the sample size is small and replication with larger samples is needed. The comparison group was heterogeneous in terms of etiology. It will be useful to compare parenting stress and stigma in epilepsy with less heterogeneous groups to better understand possible differences in the nature of stigma and parenting stress between epilepsy and other neurodevelopmental and neurological conditions. Fathers were not included and it is important to include fathers in studies of parenting stress as their perspectives and needs may differ from that of mothers. The measure used to assess stigma was designed for the epilepsy population and had not been used in a non-epilepsy population

before. The study was based in a defined geographical areas of the UK and findings may not be generalizable to other parts of the world. There were significant differences between mothers in the epilepsy group and mothers in the neurodisability group with respect to previously diagnosed mental health problems which may have influenced perceived stigma.

5. Conclusion

Epilepsy in young children can have a very significant impact on maternal parenting stress largely as a function of the associated developmental and behavioral comorbidities. When considering the impact of epilepsy on maternal wellbeing it will be important to include a measure of parenting stress. There is a need to not only consider child behavior problems but also mother-child interaction and provide supports where necessary in order to reduce stress and maximise maternal and child quality of life. Perceived stigma associated with epilepsy in young children is largely due to non-epilepsy variables including child developmental level. This needs to be taken into account when developing stigma reducing initiatives.

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Disclosure

Professor Brian Neville was a Trustee of the George E Neville Foundation. None of the remaining authors have any conflict of interest to declare. 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.

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