

## ARTICLE

# Enablers and barriers in dental attendance in Rett syndrome: an international observational study

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## Abstract

**Aims:** Intellectual and developmental disabilities are heterogeneous in aetiology and presentation, and one cannot make assumptions about the oral health barriers of those with Rett syndrome (RTT) based on findings from generic studies. This study investigated caregivers' perceptions regarding access to dental care for those with (RTT), and associations of dental treatments received by those with RTT with their caregivers' perceived value of oral health and perception of their own as well as their daughter's dental anxiety.

**Methods and results:** Retrospective observational data of a subset of individuals with confirmed *MECP2* mutations in the *InterRett* database ( $n = 216$ ) were used to explore caregiver-related factors and their relationships with longitudinal data on dental service utilisation, using negative binomial regression. The main reported barriers to dental care access for individuals with RTT were primarily dentist-related in nature, regardless of dental service history. Those with reported dental nonattendance were of older age. Increasing levels of caregiver-reported dental fear were associated with less frequent dental check-ups or for any appointments for affected individuals.

**Conclusions:** Dentist-related barriers and caregiver-reported anxiety may both adversely affect dental attendance for those with RTT. Future research should explore caregivers' beliefs and oral health literacy.

## KEYWORDS

delivery of dental care, developmental disabilities, oral health, Rett syndrome

## 1 | INTRODUCTION

Rett syndrome (RTT) is a neurodevelopmental disorder which affects approximately 1:9000 live female births<sup>1</sup> and is caused by mutations in the *MECP2* gene, located on the X chromosome.<sup>2</sup> Relationships between geno-

type and phenotype have been investigated.<sup>3,4</sup> After a period of apparent normalcy, affected individuals show regression, and then multiple comorbidities appear, which include epilepsy,<sup>5-7</sup> problems with nutrition and growth,<sup>8</sup> scoliosis,<sup>9-11</sup> respiratory problems including breath holding,<sup>12,13</sup> sleep disorders,<sup>14,15</sup> gastrointestinal

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problems including reflux,<sup>16,17</sup> and susceptibility to bone fracture.<sup>18,19</sup> The current main diagnostic criteria include partial or complete loss of manual dexterity, spoken language, as well as abnormal gait and stereotypic hand movements.<sup>20</sup>

Bruxism, when awake, is one of the supporting diagnostic criteria for RTT.<sup>20,21</sup> The reported prevalence of bruxism ranges from 80% to 94%,<sup>22,23</sup> as discussed in our recent review of the literature.<sup>24</sup> Diurnal bruxism is the most frequently reported oral presentation,<sup>25,26</sup> and there are various management approaches reported in the published literature in RTT, ranging from tooth restorations for worn or fractured tooth structure to more novel approaches such as bite planes and even acupuncture.<sup>24</sup>

While bruxism in RTT has been explored and some studies of the overall oral health experiences of those with RTT have been completed, including oral disease incidence, type and modality of treatment received,<sup>25,27–29</sup> little is known regarding barriers faced by individuals with RTT for accessing timely and appropriate oral health care. An Australian population-based retrospective study<sup>27</sup> found that dental extractions were more likely to have occurred in individuals with RTT living at home in households with a low annual household income. This is the same as seen in the normal population, and suggests that social advantage may benefit oral health status,<sup>27</sup> notwithstanding the presence of targeted public health policy schemes that may be present in other countries. Drivers for dental treatment in RTT also include bruxism as well as dental caries. Those with full oral feeding receive more dental treatment than those with full tube feeding and may be attributed to the differences in both aspiration risk, and in tooth contact time and thereby opportunity for demineralisation to occur in the caries process. Moreover, higher maternal education levels may help drive better oral health outcomes.<sup>28</sup> A gap in past work is that it did not assess the impact of caregiver beliefs, nor did it assess barriers to accessing timely oral health care.

One qualitative study has described parents' perceptions of potential barriers to accessing oral health care for patients with intellectual disability,<sup>30</sup> including behavioural problems, issues around obtaining relevant information on services, and concerns about the use of general anaesthesia. Other parents could recount positive experiences in their child's receiving oral health care.<sup>30</sup> These themes are similar to those from studies of access to dental services for other populations.<sup>31–33</sup>

While dental attendance rates may differ for patients with special needs compared with counterparts in the general population,<sup>34,35</sup> it seems likely that maintaining oral health is valued by most individuals with special needs, or

by their caregivers.<sup>28,36</sup> As well, oral health status impacts the quality of life of those with disability.<sup>37</sup>

Possible barriers to utilising dental services by individuals with disabilities include inadequate training of providers, the challenging behaviours of the individual,<sup>38</sup> inadequate reimbursement and unprofitable care provision due to the additional time required,<sup>39</sup> and practitioner perceptions that dental problems may not be of major importance.<sup>40</sup> Other barriers to oral health care include inadequate clinical facilities, problems gaining informed consent, distrust of dentists, treatment costs and anxiety.<sup>41</sup>

Intellectual and developmental disabilities (ID/DD) are heterogeneous in aetiology and presentation, and one cannot make assumptions about the barriers of those with RTT based on findings from generic studies of individuals with other ID/DD. Hence, the present study was undertaken to explore barriers to accessing oral health care for individuals with RTT.

Using data collected in the *InterRett* database, we investigated the following research questions:

1. What are caregivers' perceptions regarding access to dental care for individuals with RTT?
2. What are the associations of dental treatments received by individuals with RTT with their caregivers' perceived value of oral health and perception of their own as well as their daughter's dental anxiety?

## 2 | METHODS

Ethics approval for this study was received from the Human Research Ethics Committee of the University of Western Australia (Approval No. RA/4/1/7449).

### 2.1 | Data source and selection criteria

Established in 2002, the International Rett Syndrome Phenotype Database (*InterRett*) is the world's only ongoing international database of families with Rett syndrome, and on registration, families are invited to complete a baseline questionnaire about their daughter.<sup>42</sup> Two follow-up questionnaires in 2015 and 2018 were administered to explore the natural history of RTT.<sup>43,44</sup> English speaking families who participated in the 2015 (first) follow-up study were also the invited to participate in the 2018 (second) follow-up study. This second follow-up questionnaire included a dental component designed and piloted at Rett syndrome family conferences and with a consumer reference group comprising families of individuals with Rett syndrome. Families who returned a completed questionnaire were included in the present study.

## 2.2 | Variables

In considering the available literature on Rett syndrome and oral health, the following directed acyclic graph (DAG) (Supplementary Figure 1), illustrates relationships, of salient predisposing factors to the various exposure and outcome variables to illustrate potential sources of bias and which shaped the approach to treatment of confounding factors in this study, which are described herein.

White nodes denote adjusted variables in the current study. It is noted that household income, an unobserved variable in this study, was unable to be adjusted as income data was not available in the international dataset. Proposed proxy variables for financial status, included in the piloting process, were excluded from the final questionnaire version approved by the ethics committee, and this was related to burden on families in view of the overall length of the 2018 Wave 2 follow up family questionnaire, which approached 300 questions in total.

## 2.3 | Outcome

**Dental visits:** Families were asked whether their daughter with RTT had visited the dentist in the preceding three years; those who did provided information on the approximate timings of each dental visit in a three-year period, including the nature of procedures performed (clinical examination, radiographs, tooth debridement, restorations, stainless-steel crowns or extractions).

## 2.4 | Covariates

**Demographic/Socioeconomic factors:** The mother's highest level of education, and parental employment status were captured as categorical variables. The country of residence was identified from data collected at registration.

**Child factors:** Age groups were defined to best to approximate the primary dentition (0–6 years), mixed dentition (6–12 years), and permanent dentition (12–19 years; 19 years and above) phases of dental development.

**MECP2 mutation types:** They were categorised as C-terminal deletions, p.Arg106Trp, p.Arg133Cys, p.Arg168\*, p.Arg255\*, p.Arg270\*, p.Arg294\*, p.Arg306Cys, p.Thr158Met, early truncating deletions<sup>3</sup> and large deletions.<sup>4</sup> All other *MECP2* mutations were categorised as 'other.'

**Enabling/predisposing factors to accessing oral health service:** Families were asked whether their daughter had visited the dentist in the preceding three years, and

to rate the following parameters on a five point scale: the value they placed on oral health; the ease of getting their daughter with RTT to dental clinic visits; the standard of dental care provided; and the perceived levels of dental anxiety of the daughter and of the caregiver. Variables were aggregated if subcategories contained fewer than ten percent of the total sample size, in order to facilitate more appropriate analysis. Thus, the perceived levels of dental anxiety of the daughter and of the caregiver were then aggregated into a three-point scale. Families were also asked to indicate the barriers to their daughter seeing a dentist from a list of factors including: the caregiver's own dental fear/anxiety, high cost, 'the dentist does not understand my daughter,' 'it is hard to find a dentist to treat my daughter who has special needs,' 'my daughter can't get inside the building,' 'the waiting list for treatment is too long,' 'the waiting list to see the dentist is too long,' 'there is a lack of expertise to treat my daughter,' 'lack of time,' 'my daughter has more important issues that need to be dealt with.' Free text fields were provided as appropriate.

## 2.5 | Statistical analysis

The incidence figures of all dental treatments, dental restorations ("fillings" and "stainless steel crowns"), extractions (number of teeth extracted, including third molar extractions), dental examinations and all other dental appointments over were calculated by dividing the number of episodes of treatment/examination/appointment during the previous three-year period by the person-time at risk of experiencing the event. Confidence intervals of incidence rates were determined using the exact Poisson method. Negative binomial regression was used to estimate the incidence rate ratio of the association of dental treatments with caregiver beliefs on oral health and reported dental fear of the caregiver and their daughter, with adjustment for confounding factors, namely age group, mother's highest level of education, and the mother's and parental employment status respectively. All analyses were carried out using Stata/IC version 16.0.<sup>45</sup>

## 3 | RESULTS

There were 232 invited families for this study, and 216 completed the entire questionnaire, giving a response fraction of 93.1%. For these families included in this study, the mean age of their daughter at the *time of original baseline* recruitment to the *InterRett* database itself was 10.9 years

TABLE 1a Caregivers' perception of access to dental care in 216 individuals with RTT

Parameter	Rating, n (%)					
	Excellent	High	Good	Poor	Very poor	Missing
Standard of dental care	93 (43.1)	57 (26.4)	52 (24.1)	8 (3.7)	1 (0.5)	5 (2.3)
Ease of getting to dental visits	Very easy	Fairly easy	Somewhat difficult	Difficult	Very difficult	Missing
	72 (33.3)	84 (38.9)	24 (11.1)	12 (5.6)	18 (8.3)	6 (2.8)
Dental anxiety	Very low	Low/moderate	High/very high	Missing		
(Perceived) daughter	48 (22.2)	120 (55.56)	43 (19.91)	5 (2.3)		
Caregivers	66 (30.6)	109 (50.46)	35 (16.20)	6 (2.8)		

(range 1.7–45.0 years), with the *date of original recruitment* spanning from 2003 to 2015.

Characteristics of the study population are reported in Supplementary Table S1. The age range of individuals with RTT was 6.2–51.9 years (mean 20.5, SD 9.5). There were no individuals under 6 years of age in the study cohort. All cases were female.

### 3.1 | Caregivers' perception of access to dental care

The majority of families favorably rated the standard of dental care received, with 93 (43.1%) indicating 'excellent' care, and approximately one quarter giving 'high' (26.4%) and 'good' (24.1%) scores, respectively (Table 1A). Most families reported access to dental clinic visits as 'very easy' (33.3%) or 'fairly easy' (38.9%). About one tenth (11.1%) of respondents had some difficulty getting to the clinic, and a very small portion finding it 'difficult' (5.6%) or 'very difficult' (8.3%) to access care (Table 1A). When asked to indicate specific barriers to their daughter with RTT seeing a dentist, 127/216 (58.8%) caregivers cited no barriers. Among the 88 who responded to perceived barriers, 55 (62.5%) stated that 'finding a dentist who will treat [my] daughter' was a hurdle to seeking care. Thirty two (36.4%) cited 'a long waitlist to see the dentist' as a barrier to dental care. Less frequent barriers included other more important issues requiring attention (13.6%), lack of understanding on the part of the treating dentist (11.4%), high cost of dental care (10.2%), poor building access (9.1%), lack of time/own fear (6.8% respectively) and long waitlists for treatment (5.7%) (Table 1B). Twelve families indicated that their daughter had not attended the dentist in the preceding three years. In this subgroup, the most common barrier was the long waitlist to see the dentist ( $n = 9$ ), followed by 'finding a dentist who will treat my daughter' ( $n = 8$ ) and 'the dentist does not understand my daughter' ( $n = 5$ ). Three families indicated that their 'daughter has more

important issues to deal with,' and two cited poor building access as a barrier. Three caregivers cited 'other' barriers including one whose daughter had all teeth removed in her 20s and had not seen a dentist since then. The caregiver's own fear, cost, lack of expertise, and long waitlist for treatment, were listed also ( $n = 1$ , respectively). Interestingly, where participants had no reported dental visits in the preceding three years, most of the individuals with RTT were over 17 years of age ( $n = 10$ ).

### 3.2 | Caregivers' dental anxiety in relation to frequency of dental care

The majority of families reported that their daughter had low/moderate, or very low levels of dental anxiety. There were similar ratings for the caregiver's own levels of dental anxiety (Table 1A). The incidence rate ratios are shown in Table 2, while Supplementary Table S2 reports the incidence rates of dental treatment by different variables reflecting the caregiver's beliefs. Compared with those caregivers with very low reported dental anxiety, those caregivers with low/moderate and high/very high levels of dental anxiety had daughters with RTT who less frequently attended dental care, including both dental examinations (low/moderate caregiver dental fear: aIRR = 0.752, 95% CI 0.603–0.939;  $p = 0.012$ ) (high/very high caregiver dental fear: aIRR = 0.615, 95% CI 0.446–0.846;  $p = 0.003$ ) and overall visits regardless of appointment type (low/moderate caregiver dental fear: aIRR = 0.753, 95% CI 0.614–0.924;  $p = 0.007$ ) (high/very high caregiver dental fear: aIRR = 0.663, 95% CI 0.494–0.891;  $p = 0.003$ ) (Table 2). Similarly, compared with the reference group, dental examinations were less frequent when the patients with RTT themselves had perceived increased levels of dental anxiety (low/moderate: aIRR = 0.748, 95% CI 0.586–0.955;  $p = 0.020$ ) (high/very high: aIRR = 0.751, 95% CI 0.552–1.022;  $p = 0.069$ ), as well as overall dental appointments for those with low/moderate dental fear (aIRR = 0.785, 95% CI 0.624–0.987;  $p = 0.038$ ).

**TABLE 1b** Caregivers' perceived barriers in access to dental care in 216 individuals with RTT

	Overall (n = 216)	Age group <sup>a</sup>		
		7–12 years (n = 33)	13–19 years (n = 88)	>19 years (n = 95)
<b>Perceived barrier</b>	n (%)			
No	127 (58.8)	15 (45.5)	56 (63.6)	56 (58.9)
Yes	88 (40.7)	18 (54.5)	32 (36.4)	38 (40.0)
Missing	1 (0.5)	0	0	1(1.1)
<b>Type</b>	n (%) <sup>b</sup>			
Own fear	6 (6.8)	2 (11.1)	4 (12.5)	0
High cost	9 (10.2)	2 (11.1)	4 (12.5)	3 (7.9)
Dentist does not understand daughter	10 (11.4)	2 (11.1)	6 (18.8)	2 (5.3)
Finding dentist who will treat daughter	55 (62.5)	10 (55.6)	20 (62.5)	25 (65.8)
Lack of expertise	2 (2.3)	0	0	2 (5.3)
Poor building access	8 (9.1)	0	4 (12.5)	4 (10.5)
Long waitlist for treatment	5 (5.7)	1 (5.6)	1 (3.1)	3 (7.9)
Long waitlist to see dentist	32 (36.4)	8 (44.4)	12 (37.5)	12 (31.6)
Lack of time	6 (6.8)	1 (5.6)	3 (9.4)	2 (5.3)
Daughter has more important issues to deal with	12 (13.6)	5 (27.8)	5 (15.6)	2 (5.3)
Other	10 (11.4)	1 (5.6)	4 (12.5)	7 (18.4)

n, number of individuals.

<sup>a</sup>Age range of individuals with RTT was 6.2–51.9 years (mean 20.5, SD 9.5).

<sup>b</sup>Denominator is the number of caregivers who reported any perceived barriers.

## 4 | DISCUSSION

This study provides several insights into dental treatment patterns of patients with RTT in countries other than Australia, the latter of which is captured in the ARSD and the subject of another ongoing study and therefore will not be discussed in relation to this study. In those families where the daughter with RTT was attending the clinic for care, the overall satisfaction with this from the perspective of the caregivers was high in the majority of cases. The overall level of satisfaction of 93.5% is higher than the 85% reported in the a recent UK national patient survey.<sup>46</sup>

In terms of barriers to care, in the sample population, 58.8% reported no barriers to their daughter seeing the dentist, regardless of whether or not they had attended a dental clinic in the preceding three years. Existing cross-sectional studies of those with IDD or disability have not reported the proportion of survey respondents with no perceived barriers to dental care access, so direct comparisons are not possible.

For the families in the present study where there had not been any dental attendance in the preceding three years, their reported barriers could be compared with those of non-attenders in other settings. In a U.S. survey, only 9% of dental non-attenders in the preceding 12-month period cited 'no barriers' to dental attendance.<sup>47</sup> In the UK, one fifth of non-attenders in a two year period cited 'no need',

while 26% 'never tried' to get a dental appointment.<sup>48</sup> In contrast, for the RTT group, all families with non-attendance reported there being at least one barrier to attendance.

In other studies evaluating dental access barriers for patients with IDD, cost was reported as a major barrier in several studies.<sup>36,49–52</sup> In comparison, for the RTT cohort, few cited 'cost' as a barrier, regardless of their recent dental attendance history- which could be explained by sociodemographic factors as 60% of the sample had a university education and it has previously been suggested that this group may be more advantaged compared with the population-based Australian cohort in the ARSD,<sup>42</sup> as distinct from this *InterRett* database. This finding contrasts with cost being a major obstacle for 59% of adult dental non-attenders in the US in 2015.<sup>47</sup> A 2019 study reported that 9.9% of the overall US population did not seek dental care due to cost,<sup>53</sup> with the figure being 4.4% for those under 19 years of age. Furthermore, in the US, children with special health care needs are more likely to have cost as a barrier to dental care, than healthy children.<sup>54</sup>

In Canada, where most of the general population (74.7%) were recently reported to have seen a dentist in the preceding year, some 22.4% will have avoided care because of cost.<sup>55</sup> Issues stemming from the cost of dental services will invariably be linked to the model of care and coverage by health insurance. This will vary between

TABLE 2 Incidence rate ratios of dental treatment by caregiver beliefs

Covariate, Subcategory	n (%) <sup>a</sup>	Dental treatment <sup>b</sup> adjusted IRR (95% CI), p value	Dental restorations	Dental extractions	Dental check-ups	Dental appointments-all
<b>Value of oral health n = 216</b>						
Not important at all	0 (0)					
Not important	0 (0)					
Neutral	17 (7.87)	Baseline	Baseline	Baseline	Baseline	Baseline
Important	51 (23.61)	1.90 (0.31, 11.78), 0.492	0.30 (0.01, 6.19), 0.437	0.62 (0.13, 2.92), 0.543	0.98 (0.62, 1.55), 0.926	0.90 (0.59, 1.38), 0.641
Very important	148 (68.52)	4.18 (0.81, 21.58), 0.088	2.41 (0.20, 29.82), 0.492	0.89 (0.23, 3.38), 0.862	1.19 (0.78, 1.82), 0.413	1.07 (0.72, 1.58), 0.739
<b>Caregiver's dental fear n = 210</b>						
Very low	66 (30.56)	Baseline	Baseline	Baseline	Baseline	Baseline
Low/Moderate	109 (50.46)	1.52 (0.77, 2.97), 0.226	1.13 (0.32, 4.01), 0.852	1.09 (0.45, 2.64), 0.847	0.75 (0.60, 0.94), 0.012	0.75 (0.61, 0.92), 0.007
High/Very high	35 (16.20)	1.11 (0.45, 2.75), 0.814	1.00 (0.16, 6.13), 0.996	0.80 (0.27, 2.36), 0.688	0.61 (0.45, 0.85), 0.003	0.66 (0.49, 0.89), 0.006
<b>Perceived daughter's dental fear n = 211</b>						
Very low	48 (22.22)	Baseline	Baseline	Baseline	Baseline	Baseline
Low/Moderate	120 (55.56)	0.96 (0.46, 1.98), 0.904	1.55 (0.38, 6.36), 0.546	0.70 (0.28, 1.76), 0.448	0.75 (0.59, 0.95), 0.020	0.78 (0.62, 0.99), 0.038
High/Very high	43 (19.91)	1.14 (0.49, 2.67), 0.760	2.24 (0.48, 10.43), 0.306	0.76 (0.26, 2.20), 0.610	0.75 (0.55, 1.02), 0.069	0.83 (0.62, 1.11), 0.204

n, number of individuals; IRR, incidence rate ratio; CI, confidence interval; LL, lower limit; UL, upper limit.

<sup>a</sup>figures account for missing values.

<sup>b</sup>IRR adjusted for age group, mother's employment status, parental employment status and mother's highest level of education.

jurisdictions. In the US, dental service affordability and access may be influenced by the breadth of coverage of the insurance provider, be that private insurance, a self-funded plan or Medicaid.

A small proportion of the families in the present study were from the U.K. Barriers in the UK to care within the public sector include waiting list issues (14.1%) and cost (5%).<sup>48</sup> One reason why cost may not have been perceived as a major barrier for most families in the present study was that the families placed a high value placed on oral health and therefore on dental attendance, as noted in a previous study.<sup>28</sup> Another reason why cost may not have been perceived by many as a major barrier for dental access may again be sociodemographic factors. In the absence of household income information being available, socioeconomic status could perhaps be best approximated by parental education and employment status, as at least half of families had at least one parent in full-time employment and that over 60% of mothers had university education.

Most of the families in the present study reported some difficulty finding a dentist who would treat their daughter, or long waitlists, regardless of their dental attendance history. This has been a common issue in studies of dental patients with IDD, namely the dentist's unwillingness to treat the case, the dentist's availability, a lack of sufficient appointment [dentist's] time, and a lack of experience of the dentist.<sup>49–51,56,57</sup> In the current RTT sample, the most important dentist-related barriers were a long waiting list, 'finding a dentist who will treat my daughter' and that a concern that the 'dentist does not understand my daughter.'

The willingness of a dentist to care for a patient with RTT will be influenced by the available systems for care. In the US, over 90% of dentists work in the private sector.<sup>58</sup> This explains why, in 2012, of the \$85 billion spent on dental care, the majority (\$76 billion) was spent through private dental insurance or through out-of-pocket expenses, and only some six percent (\$5.5 billion) was spent on dental care through Medicaid/Medicare. Hence, less was spent per patient through the public schemes.<sup>59</sup> Those with Medicaid insurance were less likely to have attended the dentist, and this is seen in statistics for children with special health-care needs.<sup>54</sup>

A further relevant consideration is the difficulties experienced in the transition from pediatric to adult-based health services. This challenge is well documented in both the medical and dental literature.<sup>60–66</sup> Once again, jurisdictional differences will have an influence. Countries with a larger public healthcare system, such as Australia and Canada, may tackle this challenge in less complex ways. The relationship between insurance coverage and dental access is complex in that it involves the interplay between

multiple factors, including not only out of pocket costs, but also health literacy and service availability.

The presence and amount of dental fear in caregivers of, and patients with RTT seemed to influence the relative dental attendance in general, and for examination appointments, in this RTT sample. In the general population, dental anxiety is known to reduce dental attendance, however in the RTT cohort, less than two percent of caregivers indicated that dental fear was a major barrier to their daughter seeing the dentist, despite 16.2% of the caregivers themselves having high or very high self-ratings of dental anxiety. Other observational studies have reported child and caregiver fear as a barrier to dental care access in those with IDD<sup>36,50</sup> as well as in the general population.<sup>67–70</sup>

There are several limitations to the present study. First, the database was not derived from a population-based sample. The study population was confined to English speaking families and did not have girls less than 6 years of age due to the recruitment criteria. Additionally, it is likely that more pro-active families may be likely to participate. Thus, the present findings may not be applicable to younger children with RTT. Additionally, the relative social advantage of the *InterRett* sample<sup>42</sup> may limit applicability of findings relating to other populations with a lower SES. Additionally, the severe disabilities of the population necessitated use of parent-proxy questionnaires, including that of dental service provision over the preceding three years. However, the team has found that this time frame for recalling past events has been a reasonable one for families answering the questionnaires based on administration of successive longitudinal overall family questionnaires to those in their Australian population-based database every three or so years. Finally, wider confidence intervals in some findings, despite the high participation fraction, may reflect the rare prevalence of the disorder.

The findings of this study highlight the interplay of caregiver attitudes and dental attendance for patients with RTT. In those with IDD as highlighted, both carers and dentists are likely to underestimate their dental treatment needs.<sup>71</sup> This is especially so in RTT, where pain sensitivity is also decreased,<sup>72</sup> and where affected individuals are severely impaired and thus unable to communicate that they are experiencing pain. This underscores the importance of recognising barriers to optimal dental care access in RTT. Targeted oral health promotion at the times when caregivers attend dental appointments could be beneficial. This may help to solidify their role as advocates for their daughters and increase their awareness of dental conditions requiring care, as well as the importance of preventive care.

In conclusion, overall, this study shows that the major reported barriers to dental care access for individuals with RTT are dentist-related. Increasing levels of caregiver

self-reported dental fear also reduces attendance for both dental examinations and for visits regardless of treatment rendered. Future research should examine the extent to which the caregivers' oral health literacy affects dental preventive practises and health outcomes, in order to shape future efforts to identify realistic strategies to maintain and improve the oral health of those with RTT.

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## CONFLICTS OF INTEREST

The authors have no conflicts of interest to declare.

## ETHICS APPROVAL STATEMENT


Ethical approval for this study, which conforms to the Declaration of Helsinki, was received from the Human Research Ethics Committee of the University of Western Australia (Approval No. RA/4/1/7449; 5 October 2015, amendment 7 June 2018).

## PATIENT CONSENT STATEMENT

Consenting families who returned a completed questionnaire were included in the study.

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## SUPPORTING INFORMATION

Additional supporting information may be found in the online version of the article at the publisher's website.

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