

Brief Report

Preliminary Report on the Efficacy of Music Therapy to Optimize the Compliance of Rett Syndrome Patients Attending Oral Hygiene Procedures

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Abstract: Background: Individuals with intellectual developmental disability (IDD) often face barriers in accessing dental care. The main aim of the present study was to test the feasibility and the outcome of oral hygiene (OH) procedures in Rett syndrome (RS) patients, and a secondary aim was to test the utility of music therapy during dental hygiene procedures. Methods: A case group (Group 1) of female patients with RS (stage 3 of disease, age > 5 years and <18 years) and a control group of age-matched healthy females (Group 0) were formed, and subgroups received the intervention of oral hygiene combined with a music therapy (Subgroup 1M and Subgroup 0M) or standard oral hygiene (subgroup 1W and subgroup 0W). The Simplified Oral Hygiene Index (OHI-S) was used to assess the effectiveness of oral hygiene measures. Results: The OHI-S at T0 was 1.41 in Group 1 and 2.58 in Group 0 (p value < 0.001). The mean duration of the oral hygiene treatment was 7018'' (SD 3272'') in Group 1 and 873'' (SD 161'') in Group 0 (p value < 0.001). The mean duration of OH was significantly higher in Subgroup 1M and Subgroup 0M. Discussion: Based on the results of our case-control study, OH procedures can be successfully performed in RS patients with good compliance and efficacy. In addition, our study provides preliminary evidence to suggest that music therapy may be a useful adjunct, as it appears to improve patient cooperation.

Keywords: Rett syndrome; intellectual developmental disability; music therapy; oral hygiene



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1. Introduction

Rett syndrome (RS) is a rare genetic neurological disorder that primarily affects girls. It is typically caused by a mutation in the methyl-CpG binding protein 2 (MECP2) gene located on the X chromosome, which encodes for the homonym protein that is crucial for brain development and function [1]. RS patients usually begin to show symptoms of development regression in early childhood, around 6–18 months of age. Loss of functional use of the hands, loss of acquired speech and language skills, coordination problems, breathing difficulties, and intellectual developmental disability (IDD) are the main characteristics of RS. Individuals with RS may also experience seizures, scoliosis, and gastrointestinal disease [2]. While there is currently no cure for RS, the treatment is focused on managing symptoms and providing supportive care to improve quality of life [3].

Individuals with IDD often face barriers in accessing dental care due to a lack of specialized training among dental professionals and inadequate resources to accommodate their needs [4–6]. Studies have shown that dental professionals may be hesitant to treat patients with IDD due to a lack of training or experience, misconceptions about their ability to comply with dental procedures, and fear of legal liability [7]. Additionally, some dental offices may not have the necessary equipment or accommodations, such as specialized chairs or additional staff, that would enable patients with IDD to be effectively treated.

For RS patients, annual dental health screening and cleaning every 6 months have been recommended by international guidelines [8,9]. Nonetheless, a recent multicentric survey by Lai et al. (2022) reported barriers to dental attendance in patients with RS, mainly due to difficulties with transportation, lack of awareness and understanding of the syndrome among dental professionals, and the high cost of dental treatment [4].

Music therapy can be an effective tool for improving the quality of life of individuals with intellectual and developmental disabilities (IDD). Music therapy is a form of expressive therapy that uses music to address physical, emotional, cognitive, and social needs. It is a non-invasive and non-pharmacological approach that can be used to support individuals with IDD in various settings, such as schools, hospitals, and community centers [10,11]. Research has shown that music therapy can improve communication and social skills, reduce stress and anxiety, improve mood and emotional regulation, and enhance cognitive abilities in individuals with IDD. Music therapy can also be used to support physical therapy and rehabilitation as well as to promote relaxation and sleep [12,13]. Music therapy approaches have been reported on RS patients [14,15], but, to the best of our knowledge, there are no reports on the feasibility of music therapy to enhance the compliance of these patients during dental oral procedures.

The main aim of the present study was to test the feasibility and the outcome of oral hygiene procedures in RS patients, and a secondary aim was to test the utility of music therapy during dental hygiene procedures.

2. Materials and Methods

2.1. Study Design

This preliminary study was developed according to a quasi-experimental case-control design. The research study was performed in accordance with the ethical principles that are outlined in the Declaration of Helinski. The examination of the data was conducted in compliance with Italian laws regarding privacy and sensitive data, as well as the internal regulations of the U.O.C. of Odontostomatology, Oral and Maxillofacial Surgery of the Santa Maria alle Scotte Hospital in Siena, University of Siena. Written consent for the publication of their clinical data was obtained from all of the participants.

2.2. Population

The study involved a case group (Group 1) of female patients with RS, with the following inclusion criteria: (i) diagnosis according to International Criteria [16]; (ii) stage 3 “plateau” [17]; and (iii) age > 5 years and <18 years. The control group was composed of age-matched healthy females (Group 0). A match to disease severity was not possible due to the poor oral hygiene of the study group, which is not a common feature to find in an age-matched health population. A subgroup of each group received the intervention of oral hygiene combined with music therapy (Subgroup 1M and Subgroup 0M) or oral hygiene alone without music therapy (Subgroup 1W and Subgroup 0W).

2.3. Interventions

Two different interventions have been evaluated in the present study according to primary and secondary aims. The guardians of the patients were present in the clinic for the whole time of the procedure for both of the groups, sitting at a distance of 3 m from the operating chair.

2.3.1. Oral Hygiene

The oral hygiene session was performed with the same professional equipment for all the included patients. Patients sat on a standard dental chair (Anthos 7, Imola, Italy). The Cavitron Select SPS (Dentalsply, Charlotte, NC, USA) magnetostrictive scaler was used, and a Powerline FSI-10 Grip was used to remove supragingival plaque and tartar deposits. Manual instruments, such as scalers and curettes, were not used.

2.3.2. Music Therapy

The music was “delivered” as ambient music in the clinic through a Wireless Soundlink Speakers system (Bose, Framingham, MA, USA) and directly to the patients using Quiet-Comfort 45 high-definition wireless headphones (Bose, Framingham, MA, USA), and this was to isolate the patients as much as possible from external noise. The diffusion of music started two minutes before the beginning of the hygiene maneuvers in order to induce relaxation. The musical genre played was “Classical Instrumental” (e.g., *Romance from Piano Concerto n20 in D Minor, K. 466*, Wolfgang Amadeus Mozart; the complete list of music pieces can be seen in Supplementary Materials).

2.4. Outcomes

The total duration of the service was timed from “T0” (beginning of the session) to “T1” (the end of the procedure and/or the moment in which the patient was no longer collaborative). The Simplified Oral Hygiene Index (OHI-S) was used to assess the effectiveness of oral hygiene measures [18], which is based on the amount of plaque and calculus present on the teeth, and is scored on a scale of 0–3.

2.5. Statistical Analysis

The study employed Jamovi (version 2.3) computer software (The Jamovi Project 2022, Sidney, Australia) for macOS Big Sur [19]. Owing to the scarcity of the literature on the subject and the unavailable mean values for the target population, power analysis was not conducted. As our quantitative data were not normally distributed and the sample sizes for the compared cohorts were relatively small, the statistical significance of any differences between medians was determined using a non-parametric measure, the Mann–Whitney U test, as previously described [20]. The p -values < 0.01 were considered significant, and they are reported in the results section below.

3. Results

Twenty-four patients were enrolled in the present case-control study. Group 1 was composed of 12 female patients affected by RS (mean age 11.8 years, SD 2.54) and Group 0 was composed of 12 female healthy patients (mean age 11.6 years, SD 2.68). There were no missing values. As shown in Table 1, the OHI-S at T0 was 1.41 in Group 1 and 2.58 in Group 0, with a statistically significant difference (p value < 0.001). The mean duration of the oral hygiene treatment was 7018'' (SD 3272'') in Group 1 and 873'' (SD 161'') in Group 0 (p value < 0.001). The OHI-S at T1 was 0.33 in Group 1 and 0.0 in Group 0.

Table 1. Descriptive values of the patients included in this case-control study (Group 0 = healthy females; group 1 = RS patients).

	Group	N	Mean	Median	SD	Minimum	Maximum	M-W p -Value	M-W ES																																																
Age	0	12	10.9	11.0	2.968	6	15	0.907	0.0347																																																
	1	12	11.1	11.5	2.778	7	15			OHI-S T0	0	12	1.41	1.00	0.515	1	2	<0.001	0.826	1	12	2.58	3.00	0.515	2	3	Procedure time (")	0	12	873	872	161	624	1140	<0.001	1.000	1	12	7018	7026	3272	2521	14,520	OHI-S T1	0	12	0.0833	0.00	0.289	0	1	0.151	0.250	1	12	0.333	0.00
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Abbreviation: ES: Effect Size; M-W: Mann–Whitney U Test; N: Number of Subjects; SD: Standard Deviation. p -values < 0.01 were considered significant and highlighted in bold.

We further analyzed the results for each subgroup as described in Section 2.2. The mean duration was 4744'' (SD 1777'') in Subgroup 1M and 9292'' (SD 2827'') in Subgroup 1W; (p value = 0.026, ES 0.777); the mean duration was 765'' in Subgroup 0M and 981'' in Subgroup 0W (p value = 0.026, ES 0.7778), as shown in Table 2.

Table 2. Descriptive values of the subgroups of the present case-control study (Group 0 = healthy females; group 1 = RS patients; W = without music therapy; M = music therapy).

	Sub-Groups	N	Mean	Median	SD	Minimum	Maximum	M-W <i>p</i> -Value	M-W ES																																																																																																																				
Age	0W	6	11.0	10.5	2.53	8	14	0.197	0.472																																																																																																																				
	0M	6	10.8	12.0	3.60	6	15			OHI-S T0	0W	6	1.17	1.00	0.408	1	2	0.112	0.500	0M	6	1.67	2.00	0.516	1	2	Procedure time (")	0W	6	765	780	113	624	906	0.009	0.889	0M	6	981	993	129	780	1140	OHI-S T1	0W	6	0.167	0.00	0.408	0	1	0.282	0.333	0M	6	0.000	0.00	0.000	0	0	Age	1W	6	12.2	12.5	2.48	8	15	1.000	0.0278	1M	6	10.0	10.0	2.83	7	14	OHI-S T0	1W	6	2.33	2.00	0.516	2	3	0.112	0.500	1M	6	2.83	3.00	0.408	2	3	Procedure time (")	1W	6	4744	4410	1777	2521	7521	0.026	0.777	1M	6	9292	8580	2827	6667	1452	OHI-S T1	1W	6	0.167	0.000	0.408	0	1	0.405	0.166	1M	6	0.500	0.500
OHI-S T0	0W	6	1.17	1.00	0.408	1	2	0.112	0.500																																																																																																																				
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Abbreviation: ES: Effect Size; M-W: Mann-Whitney U Test; N: Number of Subjects; SD: Standard Deviation. *p*-values < 0.01 were considered significant and highlighted in bold.

4. Discussion

Treating children with IDD is challenging and needs the collaboration of a plethora of medical specialists. According to the Royal Australian and New Zealand College of Psychiatrists, there is a significant need for subspecialty training in the field of IDD Mental Health services as well as opportunities for the creation of new training programs [21]. The authors of the study also suggest that there is a need for increased collaboration between academic institutions, professional organizations, and government bodies. Individuals with IDD have difficulty understanding the importance of oral hygiene and communicating their oral health needs, and they also have physical limitations that can make it challenging for them to perform oral hygiene practices effectively [9]. To address these challenges, it is important for clinicians to provide tailored dental care that takes into account an individual's specific needs and abilities. Special programs for IDD patients targeting healthy behaviors, such as good oral hygiene, need continued development according to the WHO [22].

In this preliminary study, we found that the OHI-S at T0 was significantly higher in the RS group (2.58) than in the control group (1.41), indicating that the RS group had more plaque and more calculus on their teeth at baseline than the healthy controls. These results are in line with the literature: studies of periodontal disease in IDD reported increased accumulation of dental plaque and secondary augmented prevalence and severity of gingivitis and periodontitis when compared with the general population [23]. A recent review of the literature on oral pathology in RS patients concluded that, to date, no specific guidelines of the dental treatments that could be used on these patients have been reported, and, therefore, that the role of prevention is essential [9]. In our study, the mean duration of the oral hygiene treatment was significantly longer in the RS group (7018") compared to the control group (873"). This tenfold difference cannot be entirely justified by the OHI-S mismatch between the groups at T0, which suggests that the oral hygiene procedures were more challenging to perform on patients with RS. The difficulty of treating patients with IDD and especially RS patients has been reported in the literature. A retrospective chart review of the dental records of 242 individuals with RS who had attended a specialist dental clinic in the United Kingdom between 2001 and 2017 showed that many individuals with RS required specialist dental treatment, such as sedation or general anesthesia, in order to receive dental care, and that this was due to their complex needs and difficulties with cooperation during the dental procedures [7]. The study stressed the significant oral health challenges that are faced by individuals with RS, and the consequent importance of providing tailored dental care that takes into account their unique needs and challenges [7]. Regarding equipment, we used ultrasonic dental scalers in this study, which are generally considered to be more efficient and comfortable for the patient compared to manual

scalers [24]. Nowadays, these instruments are widely available and commonly used in dental clinics and practices worldwide, but it should be noted that not all dental clinics may have them. In fact, the availability and quality of dental equipment varies depending on the location, the size, and the resources of the dental practice [24].

We explored the impact of music therapy on the duration of the oral hygiene procedure. The results showed that the mean duration of the oral hygiene treatment was significantly longer in the RS subgroup that received the music therapy (Subgroup 1M) compared to the subgroup that did not receive the music therapy (Subgroup 1W). A similar trend was observed in the control group subgroups (see Table 2), with better outcomes in terms of OHI-S after treatment (0.167 vs. 0.000), although the difference was not statistically significant. These results can be interpreted as an augmented compliance of the RS patients related to listening to the music during the oral hygiene procedure. There is a growing body of scientific literature on the use of musical interventions for reducing dental anxiety. In their review of the subject matter, for example, Bradt et al. (2008) recommend that dental practices should consider incorporating music therapy services administered by trained music therapists in order to cater to highly anxious patients and children. Furthermore, to optimize the treatment's impact, music interventions should be initiated before the commencement of dental procedures [25]. Some randomized controlled trials (RCT) have been published on this topic. For instance, Lai et al. (2018), in a RCT study on 22 subjects, found that the patients who listened to music during their dental treatments experienced less anxiety and were more satisfied with their experience compared to those who did not listen to music [26]. Tan et al. (2023) recently published the first meta-analysis that pools data from multiple studies in order to investigate the efficacy of music therapies in treating patients with dental anxiety disorders. Based on their comprehensive analysis of 18 clinical randomized controlled trials, the authors concluded that music therapy has the potential to facilitate normal dental care by mitigating a patients' pain and anxiety, reducing their blood pressure and heart rate during procedures, and thereby enhancing their cooperation. However, due to the study's limitations, further multicenter, large-sample, high-quality randomized controlled trials are warranted in order to validate these findings and minimize the risk of confirmation bias [27]. Studies have also shown that dental professionals may be hesitant to treat patients with IDD due to a lack of training or experience, misconceptions about the ability of the patients to comply with dental procedures, and fear of legal liability [4,5,28]. Additionally, some dental offices may not have the necessary equipment or accommodations, such as specialized chairs, that would enable the patients with IDD to be effectively treated [5,6]. It is important for dental and oral maxillo-facial professionals to receive specialized training on how to work with patients with IDD in order to ensure that these patients are able to access necessary dental care [29,30]. This can help to reduce the barriers that patients and their families face when seeking dental treatment. Overall, our study suggests that music interventions can be an effective and easily feasible way to reduce dental anxiety in RS patients undergoing dental procedures. The application of tailored care to RS patients could ameliorate oral health, improve quality of life, and reduce the need for more complex intervention under general anesthesia [31]. However, more research is still needed in order to determine the optimal type of music and the best way to implement music interventions in dental settings. In this context, it would be feasible to use different forms of stimulation (e.g., environmental music, binaural music, etc.) or apply cognitive behavioural music therapy and/or group music therapy under the supervision of trained music therapists.

Limitations and Future Perspectives

Our study has some limitations, such as the small sample size and the lack of blinding of the personnel performing the oral hygiene procedures. However, our findings provide some insights into both the feasibility and the potential benefits of using music therapy during dental hygiene procedures for patients with RS. The use of music therapy during oral hygiene procedures was well tolerated, and it may have contributed to the positive

outcomes in terms of patient cooperation and relaxation. To the best of our knowledge, no such reports are present in the international literature, and further research is needed to confirm our preliminary findings in multicentric, well-designed clinical trials. Moreover, the aim is to implement further tailored oral procedures for RS patients. For instance, virtual reality has been utilized to reduce dental anxiety in children, and the application of VR technologies on RS patients could be tested in future studies [32].

5. Conclusions

Based on the results of our case-control study, we can conclude that oral hygiene procedures can be successfully performed in RS patients with good compliance and efficacy, taking into account a prolonged procedure time. In addition, our study provides preliminary evidence to suggest that music therapy may be a useful adjunct to oral hygiene procedures in this patient population, as it appears to improve patient cooperation. However, further larger multicentric research is needed in order to confirm these findings and to determine the optimal use of music therapy in this context. Overall, our study highlights the importance of tailored approaches to oral healthcare in patients with special needs, such as those with RS, and the potential benefits of complementary interventions, such as music therapy, in improving patient outcomes.

Supplementary Materials: Following supporting information can be downloaded at: <https://www.mdpi.com/article/10.3390/psychiatryint4030023/s1>.

Author Contributions: Conceptualization, M.T. and M.V.; methodology, M.T. and A.F.; software, M.T. and A.F.; validation, M.T. and A.F.; formal analysis, M.T. and A.F.; investigation, M.T., S.P. (Sara Pianigiani) and A.F.; resources, M.T. and A.F.; data curation, M.T. and A.F.; writing—original draft preparation, M.T. and A.F.; writing—review and editing, M.T., A.F., F.C., M.V., P.G. and G.G.; visualization, M.T. and A.F.; supervision, S.P. (Stefano Parrini), F.C., M.V., P.G. and G.G.; project administration, F.C., M.V., P.G. and G.G. All authors have read and agreed to the published version of the manuscript.

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Institutional Review Board Statement: The study was conducted in accordance with the Declaration of Helsinki, and approved by the Clinical Ethic Committee of the Siena University Hospital in date 26 July 2023, protocol n7.

Informed Consent Statement: Informed consent was obtained from all subjects involved in the study.

Data Availability Statement: The data that support the findings of this study are available from the corresponding author, [A.F.], upon reasonable request.

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Conflicts of Interest: The authors declare no conflict of interest.

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