



Article

Orthodontic Treatment in Pediatric Patients with Autism Spectrum Disorder: Compliance and Satisfaction: Pilot Study

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Abstract: Background: Most children with ASD require orthodontic treatment to correct a malocclusion, to restore masticatory function, and for cosmetic reasons. The use of orthodontic devices, however, could lead to a distortion of the child's routine, causing mood alterations, on the one hand, and a worsening of their oral hygiene status, on the other. The aim of this study is to evaluate the impact of orthodontic therapy on the daily management and level of collaboration of children with ASD determined using the Frankl scale. Materials and methods: An anonymous questionnaire was administered to 20 pairs of parents of children in which they were asked to evaluate any behavioral changes and/or difficulties encountered at the beginning of orthodontic treatment as well as their degree of satisfaction with the level of information received from medical personnel. Results: In about half of the sample, there were behavioral changes and difficulties in managing the treatment. Most of the responses received indicated parental satisfaction both with regard to the emotional impact on the children with respect to the introduction of the treatment and with regard to the attention received from the medical staff. Furthermore, there was a statistically significant increase (p = 0.001) in the Frankl scale score after treatment. Conclusions: Home management, which has always been the most difficult obstacle to overcome, with the support of a competent team can not only improve the quality of life of patients affected by ASD but also improve their collaboration and state of oral health without altering their emotionality.

Keywords: ASD; special needs; orthodontic treatment; satisfaction



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1. Introduction

Autism spectrum disorder (ASD) is a neurodevelopmental disorder with onset in the first three years of life and may be accompanied by mild, moderate, or severe intellectual disability [1].

It is a complex condition that involves persistent deficits in social interaction, verbal and nonverbal communication, and repetitive and stereotyped behaviors that occur at disparate levels and in extremely different ways [1].

The Centers for Disease Control and Prevention (CDC) in Atlanta has been conducting an epidemiological study in 11 states of the United States for two years on children who are turning eight years old. In 2016, it reached 18.5 per 1000, which is equivalent to 1 person with autism in every 54 people [2].

Studies in Asia, Europe, and North America have identified people on the autism spectrum at an average rate between 1% and 2% (CDC, Data & Statistics on Autism Spectrum Disorder).

A recent study conducted by Autism Europe on 631,619 children between the ages of 7 and 9 in different European countries determined an estimated average prevalence of 12.2 per 1000 (1 in 89). The values of various countries range from 4.4 to 19.7 per 1000

Appl. Sci. 2023, 13, 9189 2 of 11

(Autism Spectrum Disorders in the European Union (ASDEU) program summary report, 9/2018). There are no reliable data on the number of people with autism in Italy and scarce public data on the number of people affected. However, based on the same values, compared to 435,000 newborns in Italy in 2020, there would be over 4000 children on the autism spectrum each year. According to the Centers for Disease Control and Prevention (CDC), the prevalence of children with ASD has more than doubled, from 1 in 149 to 1 in 59 children with autism. The care of children with ASD and their families requires a particular engagement from health services. Like any other individual, a person with a disability has the right to receive the best health care possible without discrimination (Minister of Labour, Health and Social Policies; United Nations Convention on the Rights of Persons with Disabilities, September 2009). In the health care sector, therefore, to grant people with disabilities these rights, it is necessary to apply the principles of reasonable accommodation and universal design. Specifically, "reasonable accommodation" refers to the necessary and appropriate modifications and adaptations that, if adopted, do not impose a disproportionate or excessive burden to ensure people with disabilities enjoy and exercise, on an equal basis with others, all human rights and fundamental freedoms. "Universal design," in contrast, consists of creating products, structures, programs, and services that are suitable to all people, at the greatest extent possible, without the need for specialized adaptations or designs [3]. The American Academy of Pediatric Dentistry, in its guidelines, emphasizes how people with disabilities have an increased risk of developing oral diseases (American Association of Pediatric Dentistry—AAPD. 2008. Guideline on management of dental patients with special health care needs). Moreover, according to the World Health Organization, two-thirds of people with disabilities do not receive oral care. This is due to several factors, including the challenging management of their emotional reactions; the lack of attention that parents/guardians of patients have regarding the prevention of caries, considering oral health a secondary aspect in managing ASD (halo effect) [4,5]; and the unpreparedness of professionals in ambulatory management, as well as the use of sedation or general anesthesia (World Health Organization & World Bank (2011). World report on disability 2011. https://apps.who.int/iris/handle/10665/44575, accessed on 28 April 2023. Potential predisposing factors for poor oral health in patients with ASD, as well as the rest of the population, are mainly diet, oral hygiene, and low motivation/collaboration. A study conducted in Italy suggests that school-aged children (between 5 and 14 years old) with ASD are a high-risk population. A preference for soft and sticky foods and the use of sweet drinks make diet a significant risk factor for caries, while the difficult collaboration in domestic oral hygiene suggests a deterioration in periodontal health, as also evidenced in the existing literature for similar populations [6]. Therefore, it is easily understandable how it is more complex to maintain oral health or to subject individuals with this disorder to normal dental therapeutic maneuvers, if necessary. Intercepting dental problems early in young patients who are not able to univocally manifest a painful sensation from the oral cavity is undoubtedly more difficult [7,8]. The most influential risk factor in children with autism, however, remains oral hygiene, and special attention should be paid to the presence of visible plaque and gingivitis in individual patients. In most cases, oral hygiene maneuvers are not performed by the patients themselves but by their parents/guardians, for whom the success of the maneuvers is subject to numerous variables, especially the possibility of receiving indications and instructions from dental professionals [9–12].

Another risk factor is represented by orthodontic malocclusions. The literature regarding studies analyzing the characteristics and prevalence of malocclusion in individuals with ASD is scarce; most articles focus on the prevalence of dental caries and periodontal disease. According to a recent review and meta-analysis, people with ASD are at higher risk of malocclusion than those without ASD. The most represented malocclusions are Angle's Class II, Angle's Class III, open bite, increased maxillary overjet, and crowding. Moreover, individuals with ASD have higher rates of persistent parafunctional habits, including mouth breathing, biting of various objects, and onychophagy, compared to those without ASD [13]. Based on scientific evidence, it could be argued that the influence of ASD

Appl. Sci. **2023**, 13, 9189 3 of 11

on malocclusion could be explained by behavioral factors [14,15]. For patients with ASD, as well as for healthy patients, the presence of malocclusions is associated with a higher risk of dental trauma, especially in patients who have self-injurious tendencies [16,17]. Early diagnosis of malocclusion could help to promptly intervene and improve the oral health of children with ASD throughout their lives, considering that the complexity of malocclusion and the need for orthodontic treatment are statistically significantly higher among children with ASD compared to those without, regardless of sex and age [18]. In Italy, the National Health Service (SSN) grants free access to early diagnosis and individualized treatment for people with ASD using methods and tools based on the most advanced scientific evidence and ensuring them all (Supplement to the "Gazzetta Ufficiale" No. 65, 18 March 2017). In addition, autism is included in the list of Essential Levels of Assistance (LEA) that exempts from health care fees people with chronic and disabling diseases, including orthodontic treatment. In recent years, the Paediatric Dentistry and Special-Needs Patients Unit of the Umberto I Polyclinic Hospital has seen an increase in requests from parents of children with autism spectrum disorder for the installation of orthodontic devices aimed at correcting their children's malocclusion. Starting orthodontic treatment in these patients is not easy, as it modifies important aspects of their daily routine and increases the individual risk factor for caries, as the orthodontic appliance can worsen oral hygiene and increase plaque retention [19,20]. There is a risk of losing collaboration and achieving failure in reaching both orthodontic and pedagogical relational goals [2,3]. Therefore, it is essential to select patients who are suitable for managing medium- to long-term orthodontic treatment so that it does not have a negative impact on their oral health [21]. For this reason, public orthodontic services have management characteristics that make them preferable in the choice of orthodontic treatment for patients with ASD [22,23]. The objective of this study is to determine malocclusion complexity and orthodontic treatment need in children with ASD and the impact of orthodontic therapy on the daily care and level of collaboration of children with ASD in a sample of patients referred to the Paediatric Dentistry and Special-Needs Patients Unit of the Umberto I Polyclinic Hospital in Rome, Italy. The secondary objective is to evaluate the degree of satisfaction of parents with the service offered and the proposed treatment plan.

2. Materials and Methods

2.1. Study Design

A specific questionnaire was developed to evaluate the impact of orthodontic treatment on patients with ASD who were referred to the Paediatric Dentistry and Special-Needs Patients Unit of the Umberto I Polyclinic Hospital in Rome. The questionnaire consisted of 8 questions. The questions aimed to evaluate the overall satisfaction level related to the service provided and the path followed, as well as any mood or behavioral changes and the management of home hygiene resulting from the use of orthodontic devices.

The questionnaire was created using Google Forms, a web-based software program that allows the development and publication of online surveys using a web-based interface. Before being administered online, the questionnaire was submitted to a focus group made up of dentists, hygienists, and caregivers to evaluate the correct understanding of the questions.

In June 2022, the form was sent via an email link to the parents of patients undergoing orthodontic treatment at the time of the survey, for a total of 20 families, inviting them to participate and complete the anonymous online questionnaire. A reminder email was sent approximately two weeks after the initial invitation to participate, and a third and final reminder was sent in September 2022. All data collected were anonymous.

The pilot study consisted of an exploratory qualitative/quantitative investigation through an analysis relative to the answers of parents with special-needs children and clinical data collection performed by 2 specialized and calibrated dentists using the Frankl scale for evaluating the degree of collaboration and the IOTN scale for evaluating malocclusion, or misalignment, of the teeth and jaws.

Appl. Sci. 2023, 13, 9189 4 of 11

The study underwent a formal review and received approval from the Council of the Department of Oral and Maxillofacial Sciences, Paediatric Dentistry and Special-Needs Patients Unit, Umberto I Polyclinic Hospital, Rome, Italy (No. 15/2020 Prot. n. 0000216 dated 6 February 2020). This study was performed in line with the principles of the Declaration of Helsinki.

The Frankl scale, a reliable behavior-rating system in common use in both clinical dentistry and research on ASD patients, was used to measure the level of cooperativeness at the first dental visits. Each child was classified by a dentist experienced in treating ASD children into one of the four categories of the Frankl scale: definitely negative—refusal of treatment, crying forcefully, being fearful, or any other evidence of extreme negativism; negative—reluctance to accept treatment, being uncooperative, or some evidence of a negative attitude but not pronounced, i.e., sudden withdrawal; positive—acceptance of treatment, at times cautiously, willingness to comply with the dentist, at times with reserve, but following the dentist's directions cooperatively; and definitely positive—good relationship with the dentist, interested in the dental procedures, or laughing and enjoying the situation [24].

The Index of Orthodontic Treatment Need (IOTN) scale is a commonly used system for evaluating malocclusion, or misalignment, of the teeth and jaws. It is designed to help orthodontists and dental professionals assess the severity of a patient's malocclusion and determine the appropriate level of orthodontic treatment required. The IOTN scale takes into account both dental health and aesthetic factors. The IOTN scale consists of two components: Dental Health Component (DHC) and Aesthetic Component (AC) [25]. Two orthodontists performed clinical evaluation and collected all raw data.

2.1.1. Dental Health Component (DHC)

The DHC assesses the clinical need for orthodontic treatment based on certain dental health factors. It considers aspects such as overcrowding, spacing, crossbites, overjet (horizontal distance between upper and lower incisors), and reverse overjet. Each factor is assigned a specific grade from 1 to 5, with 1 indicating no or a slight need for treatment and 5 indicating a severe need for treatment.

2.1.2. Aesthetic Component (AC)

The AC assesses the impact of the malocclusion on the patient's dental appearance and smile. It evaluates factors such as the position and prominence of the front teeth, visible gaps, and irregularities. The AC grade is determined based on a set of standardized photographs or visual assessments. It ranges from 1 to 10, with 1 indicating no aesthetic need for treatment and 10 indicating a severe aesthetic need.

The overall treatment need is determined by combining the DHC and AC grades. Generally, if a patient receives a DHC grade of 4 or 5 or an AC grade of 7 or above, they are considered to have a significant treatment need and may benefit from orthodontic treatment.

2.2. Study Population

The study included children between the ages of 5 and 18 years who were referred to the Paediatric Dentistry and Special-Needs Patients Unit of the Umberto I Polyclinic Hospital in Rome, were diagnosed with autism spectrum disorder according to the criteria of the *International Classification of Diseases* (ICD-11), and were undergoing orthodontic treatment at the time of the survey. The exclusion criteria included patients whose parents did not provide consent for the study, patients with a definitely negative Frankl scale value, and patients assigned IOTN grade 1.

Appl. Sci. **2023**, 13, 9189 5 of 11

2.3. Treatment Protocol [18]

To improve the response to dental treatments in patients showing "uncooperative" behaviors with an educational approach, a "sensory education" method has been established. It focuses on the unique attitudes of each patient and how these can be selectively modulated to facilitate the operation, management, and maintenance of dental care both in the hospital and at home.

First meeting with parents: The parents are informed about the educational approach adopted in each phase of the dental treatment, and information is collected.

First visit: All the ideas regarding the environment, times, and methods of the first approach should be taken from the list of information received from the parents. However, it is important that everything remain in a perspective of flexibility, reversibility, and adaptability to meet the needs of children and to be prepared for the unexpected during dental therapy.

Environment: The environment must be designed to avoid distractions during therapy. It should be safe for the child and the operations team.

Time: The time is adapted to the needs of the patient in relation to the behavior manifested. Approach: In the first observation, the patient's ability to understand and react is verified through a sequential representation of the therapy, according to the method of augmentative and alternative communication (AAC).

Sensory approach: Following the sensory approach, the educator constantly observes the child to identify their needs or specific reactions that may require the adoption of a different strategy. The sensory approach may require several sessions, and its duration depends on the patient's level of confidence in the dentist and in the dental instruments.

The method was applied to all patients included in the sample for the entire duration of treatment.

Patients in orthodontic therapy were seen with a frequency of one appointment every three weeks.

2.4. Statistical Analysis

Twenty questionnaires were collected, each one of them adequately filled. The questionnaire data were collected and analyzed using the Microsoft Excel 16 database. Descriptive statistics were calculated for quantitative and qualitative variables, including mean \pm SD values and percentages. The Wilcoxon signed-rank test was used to evaluate differences between the values of the Frankl scale pre- and posttreatment.

Data were evaluated using standard statistical analysis software (Statistical Package for the Social Sciences, version 20.0, IBM Corporation, Armonk, NY, USA). A *p*-value of <0.05 was considered statistically significant.

3. Results

The response rate was 100%, and a total of 20 children with ASD undergoing orthodontic treatment at the time of the survey were included in this study. The mean age of the sample was 10 years, with a male predominance of 80% compared to 20% females. Of the 20 patients, 8 were undergoing treatment with fixed appliances, 9 were undergoing treatment with removable appliances, and 3 had been treated with both. In general, the orthodontic and orofacial treatment was based on four different basic treatment concepts: (1) removable orthodontic appliance (RA) in 20% of the treatments (N = 4); (2) palatal expander (PE) (with/without a facemask (FM)), with/without a lower appliance (e.g., a lingual arch or lip bumper) in 45% of the treatments (N = 9); (3) orthodontic preformed functional device (Occluso-o-Guide® by Sweden & Martina SpA, Carrare, Italy) in 25% of the treatments (N = 5); and (4) fixed appliance (braces) in 10% of the treatments (N = 2).

Successfully, 95% of all treatments (N=19) led to a favorable outcome by reaching the pre-set goal in relation to treatment progress times, while only 5% (N=1) had partial success due to a combination of non-compliance and lack of oral hygiene, which prevented the treatment from being completed.

Appl. Sci. 2023, 13, 9189 6 of 11

The average duration of orthodontic treatment at the time of the survey was 12 months. The prevalent IOTN grade in the sample was grade V, followed by grades III and IV, while few patients were classified as IOTN grade I or II. The treatment need was estimated for the majority of patients both for the Dental Health Component and for the Aesthetic Component. The IOTN-DHC revealed that a substantial portion (60%) of the ASD group had an indicated treatment need.

According to the IOTN-AC, nearly all of the ASD group had a score of 8–10, indicating the need for treatment. Tables 1 and 2 show first the distribution and description of the IOTN among the sample and then the actual treatment need according to the scoring, respectively.

Table 1. IOTN distribution among the sample.

Variables		Mean \pm SD or Number (%)
Age (years)		10 ± 3.24
Sex	Male	
	Female	
IOTN I		0
IOTN II		1
	Displacement of teeth > 1 mm but \leq 2 mm	1 (5)
IOTN III		7 (35)
	Anterior or posterior crossbites with >1 mm but \leq 2 mm discrepancy between the retruded contact position and the intercuspal position	6 (30)
	Increased and incomplete overbite without gingival or palatal trauma	1 (5)
IOTN IV		4 (20)
	Increased overjet > 6 mm but \leq 9 mm	3 (15)
	Reverse overjet > 1 mm but <3.5 mm with recorded masticatory and speech difficulties	1 (5)
IOTN V		8 (40)
	Reverse overjet > 3.5 mm with reported masticatory and speech difficulties	1
	Impeded eruption of teeth (apart from 3rd molars) due to crowding, displacement, the presence of supernumerary teeth, retained deciduous teeth, and any pathological cause	6 (30)
	Defects of cleft lip and palate	1 (5)

Table 2. IOTN Dental Health Component and IOTN Aesthetic Component.

Variables		Number (%)	
IOTN-DHC			
	No treatment need (score 1–3)	8 (40)	
	Treatment need (score 4–5)	12 (60)	
IOTN-AC			
	No treatment need (score 0-7)	2 (10)	
	Treatment need (score 8–10)	18 (90)	

Regarding the values of the Frankl scale at the beginning of treatment, only 2 of the 20 patients had a negative value of 2, while most of the sample had positive values of 3 and 4. At the end of treatment, all patients with a Frankl scale value of less than 4 improved by one degree of their starting value.

Appl. Sci. 2023, 13, 9189 7 of 11

Of the 20 participants recruited to the study, the treatment elicited an increase in the Frankl scale score of 12 participants compared to the pre-treatment values, whereas 8 participants saw no increase. The Wilcoxon signed-rank test determined that there was a statistically significant increase (p = 0.001) in the Frankl scale score after treatment.

Regarding the degree of satisfaction with the service offered and the benefits obtained with respect to the problems encountered, the majority of the sample provided positive and satisfactory answers and affirmed that it was worthwhile to undergo the therapy, even though just over half had problems again compared to the management of the orthodontic appliance, 35% reported a worsening of home oral hygiene, and 30% said they had found a worsening in the patient's mood (Table 3).

Table 3. Descriptive statistics for quantitative and qualitative variables.

	Frequency	Percentage
Frankl Scale		
2	2	10.0
3	11	55.0
4	7	35.0
Total	20	100.0
Are you satisfied with the orthodontic service?		
Scant	2	10.0
Good	3	15.0
Excellent	15	75.0
Total	20	100.0
Did you receive an adequate response to your child's needs		
during the duration of the orthodontic treatment?		
Scant	1	5.0
Good	2	10.0
Excellent	17	85.0
Total	20	100.0
Was the information received from the medical staff clear		
and exhaustive during your child's orthodontic treatment?		
Good	2	10.0
Excellent	18	90.0
Total	20	100.0
How satisfactory was the orthodontic treatment? (0 = worst		
to 5 = excellent)		
3	3	15.0
4	9	45.0
5	8	40.0
Total	20	100.0
Was it worth it?		
Yes	1	5.0
No	19	95.0
Total	20	100.0
Have you encountered difficulties in managing your		
child's braces at home?		
Nothing	3	15.0
Little	8	40.0
Enough	7	35.0
A lot	2	10.0
Total	20	100.0

Appl. Sci. 2023, 13, 9189 8 of 11

Table 3. Cont.

	Frequency	Percentage
Have you experienced a deterioration in the home		
management of your child's oral hygiene during		
treatment?		
Yes	7	35.0
No	13	65.0
Total	20	100.0
Have you experienced a change in your child's		
mood/collaboration due to the orthodontic appliance?		
Yes	6	30.0
No	14	70.0
Total	20	100.0

4. Discussion

This study aimed to determine malocclusion complexity and orthodontic treatment need in children with ASD and the impact of orthodontic therapy on the daily care and level of collaboration of children with ASD. As mentioned before, studies on malocclusion traits in children with ASD are scarce. In line with other similar studies, we found a higher prevalence for at least one malocclusion trait in children with ASD as a malocclusion Angle's Class II, a malocclusion Angle's Class III, an open bite, an increased maxillary overjet, and crowding [21,26–28].

A recent study among Indonesian children with ASD found that 20% of the tested children showed an IOTN-DHC score of ≥ 4 (n=4), indicating an orthodontic treatment need, while a study conducted in the lowlands reported 79.2%. In our study, the percentage of the IOTN-DHC score of ≥ 4 was 60%, remaining within the range of values found in the literature.

The sample of our study aligns to other orthodontics samples found in the literature [22,26–28], both for the IOTN scoring and also for the orthodontic therapies performed, the obstacles, and the successes.

From the collected data, it is evident that there is a need to make specialized health care services for diagnosis and treatment easily accessible and to ensure an adequate approach that is specific to each age group and indispensable for addressing the complexity and heterogeneity of autistic syndromes. Going to the dentist can be a problem for these patients: They are children who have other medical conditions, often more severe than dental ones, which is why parents/caregivers become concerned about the oral cavity when the situation has already deteriorated, thus having to intervene on a non-trivial oral problem that is already complex [5]. Relational difficulties and difficulties in managing people with ASD in the dental field often lead to a late diagnosis of malocclusions and preclude interceptive treatment [23]. To provide dental care to such sensitive and complex patients, appropriate tools and personnel who are truly prepared not only from a purely dental point of view but also from a psychological point of view are needed [24]. Studies conducted on samples of orthodontists show that most of them are not interested in treating patients with special needs; time constraints and poor cooperation are the most commonly cited reasons [29].

The creation of a specialized structure for the care of patients with special needs within the Paediatric Dentistry and Special-Needs Patients Unit, and the training of an adequately prepared team, has made it possible to overcome this barrier by offering PhD students and postgraduates adequate preparation to acquire the skills necessary for treating this patient population [21,29]. The team, composed of dental hygienists and orthodontists, was trained in managing ASD patients using pedagogical sensory techniques that allowed for the proper management of orthodontic treatment issues in these patients and the acquisition of cooperation from caregivers [23,26,27,30–32]. It is important to note, in our study, the evaluation of the child's collaboration through the Frankl scale, which in most of the sample

Appl. Sci. 2023, 13, 9189 9 of 11

returned to a positive value at the end of the treatment, thanks to the sensory approach used in all phases. The number of patients in our study is a limitation in establishing the effectiveness of the approach used and the predictability of the orthodontic treatment outcome. However, since there are no similar experiences in the literature, our study is the first in Italy, the results obtained are encouraging, and we aim to increase the sample size.

Some studies in the literature have examined the outcome of orthodontic treatment, reporting less satisfactory results in patients with special needs [30]. The reasons include the fact that both families and orthodontists focus more on functional outcomes and are more willing to compromise on aesthetics, also considering the timing as well as oral hygiene issues [31]. The absence of an adequate level of oral hygiene in patients with special needs and the need to not subject patients to long sessions and long treatment periods to avoid compromising their emotional stability may require premature interruption of orthodontic treatment, leading to suboptimal success [30,32]. Such issues were also found in our sample. The difficulty in maintaining correct oral hygiene, the changes in mood, and, above all, the difficulties in home management of the orthodontic appliance are problems that were overcome by correct management by the team that was able to provide the right support and the right information to caregivers and manage the emotional components of patients in treatment. Despite these limitations, the results of orthodontic treatment can still be considered satisfactory. The only patient whose treatment was partially successful was a patient whose Frankl scale score was 2 = negative and who was treated with an orthodontic preformed functional device; unfortunately, the partial failure of the therapy was due to a lack of collaboration of the patient related to his complex behavior and consequently to inadequate oral hygiene and insufficient wearing time of the device (the indication was all night plus 4 h during the daytime). As the behavior of autistic patients varies and is within a broad spectrum, the different therapeutic and psychological approaches—even when customized—are not always effective for all patients, even if they are in the same range of the Frankl scale score.

It has been shown that orthodontic treatment in the special-needs population improves not only the facial appearance but also masticatory function, speech, reduction in dentoalveolar trauma, and control of salivation [33–35]. In our sample, caregivers also found the treatment path taken to be satisfactory, despite the difficulties encountered, as providing orthodontic treatment to these children can result in a benefit in these social aspects [36–40].

5. Conclusions

In conclusion, it can be stated that although orthodontic treatment may seem complex and difficult to achieve in children with ASD, by selecting the appropriate appliance based on the child's needs and issues, good results can be obtained. Home management, which has always been the most difficult hurdle to overcome, with the support of a competent team can not only improve the quality of life of patients with ASD without altering their emotions but also improve their collaboration and oral health status. Future studies could further explore this approach.

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Appl. Sci. 2023, 13, 9189 10 of 11

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, [D.C., I.C.C.], upon reasonable request.

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

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