EDITORIAL

Dent. Med. Probl. 2011, **48**, 4, 459–463
ISSN 1644-387X

Katarzyna Olszewska, Izabella Dunin-Wilczyńska

Orthodontic Management of Children with Autism – Review of the Literature

Leczenie ortodontyczne dzieci z autyzmem – przegląd piśmiennictwa

Department of Jaw Orthopaedics, Medical University of Lublin, Poland

Abstract

Autism spectrum disorder (ASD) is a life-long neurodevelopmental disorder characterized by qualitative abnormalities in reciprocal social interactions and patterns of communication, and by a restricted, stereotyped, repetitive repertoire of interests and activities. ASD is a heterogeneous disorder with a wide range of expression, and is categorized into autism (Autistic Disorder – AD), pervasive developmental disorder – not otherwise specified (PDD--NOS) and Asperger Syndrome (AS). Patients with ASD will exhibit wide variation in their level of understanding and ability to cooperate during orthodontic treatment. The spectrum of methods used for pain and anxiety control during orthodontic treatment of the autistic child may be divided into conscious methods (such as oral, intramuscular, inhalation with nitrous oxide and oxygen and intravenous sedation) and unconscious methods. This article presents a review of the possible problems and current methods of the behavior management used in the orthodontic treatment of children with autism (**Dent. Med. Probl. 2011, 48, 4, 459–463**).

Key words: autism, orthodontic treatment.

Streszczenie

Spektrum autystyczne (ASD) jest zaburzeniem neuropsychiatrycznym charakteryzującym się znacznym upośledzeniem interakcji społecznych i komunikacji, jak również występowaniem ograniczonych, stereotypowych i powtarzających się czynności oraz zainteresowań. Stanowi heterogenną grupę zaburzeń o szerokim zakresie objawów i obejmuje następujące jednostki chorobowe: autyzm, całościowe zaburzenie rozwoju nie zdiagnozowane inaczej oraz zespół Aspergera. Pacjenci ze spektrum autystycznym wykazują dużą różnorodność zachowań i zdolności do współpracy podczas leczenia ortodontycznego. Metody stosowane w kontroli lęku i bólu podczas leczenia ortodontycznego pacjentów autystycznych obejmują metody z zachowaniem świadomości pacjenta oraz metody bez świadomości pacjenta. W artykule zaprezentowano przegląd problemów pojawiających się podczas leczenia ortodontycznego dzieci autystycznych i współczesnych metod postępowania z pacjentem ze spektrum autyzmu (**Dent. Med. Probl. 2011, 48, 4, 459–463**).

Słowa kluczowe: autyzm, leczenie ortodontyczne.

Autism spectrum disorder (ASD), introduced in 1988, is a life-long neurodevelopmental disorder characterized by qualitative abnormalities in reciprocal social interactions and patterns of communication, and by a restricted, stereotyped, repetitive repertoire of interests and activities.

ASD is a heterogeneous disorder with a wide range of expression, and is categorized into autism (autistic disorder), pervasive developmental disorder – not otherwise specified (PDD-NOS) and Asperger syndrome. Autism, PDD-NOS, and Asperger syndrome are included in the broader category of pervasive developmental disorders, along with Rett's disorder, and childhood disintegrative disorder [1–6]. The diagnosis of ASD is based on two major systems of classification: International Statistical Classification of Diseases and Related Health Problems (ICD) and Diagnostic and Statistical Manual of Mental Disorders (DSM – IV, 4th ed.) [7, 8].

Autism was first described in 1943 by the American child psychiatrist Leo Kanner. He presented 11 children whose behavior was obviously different from others. Kanner suspected that they

had an inborn feature which prevented their forming regular social contacts. Autism is now recognized as an organic disorder characterized by abnormalities in the brain, especially the limbic system and cerebellum [7].

Diagnosis of autism is based on 4 criteria: early onset (prior to age 3 years), severe abnormality of social reciprocity, severe abnormality of communication development (often including spoken language), restricted, repetitive and stereotypical patterns of behaviour, interest, and imagination. The spectrum of disorders in autism includes deficits in speech development, social skills and patterns of behavior (so called autistic triad), moreover cognitive, emotional and motoric functions.

The other two ASDs, PDD-NOS and Asperger Syndrome (AS), are less severe developmental disorders. PDD-NOS is a diagnosis of exclusion for those with problems similar to autism but insufficient to meet the criteria for autism in number, severity or age of onset. Individuals with PDD-NOS have more social activity, higher empathy and greater interaction than those with autism [1–6, 9–12].

The term Asperger Syndrome was coined by Lorna Wing in 1981 and it derives from Hans Asperger's name, who first described the disorder in 1943 [13]. The criteria for a diagnosis of Asperger Syndrome (AS) are impaired social interaction, restricted, repetitive and stereotypical patterns of behaviour, interest and activities, clinically significant impairment in social, occupational or other functioning; and no clinically significant delay in language, cognitive development, adaptive behaviour or in curiosity about the environment. Therefore, individuals with Asperger Syndrome have many autistic-like symptoms, but relatively normal language skills and an average or even high intelligence [13–17]. People with Asperger Syndrome have problems with interpersonal communication and adjusting to specific places, people and situations. AS people are similar to infantile autism patients in regard to the kind of impairment and difficulties in communication, but they stand out with better social adaptation, a presence of narrow and specific intellectual interests that make them good at one field. They are good at remembering dates and people, they have mathematical abilities. The feature that distinguishes AS patients from autistic patients is proper development of language in the range of vocabulary and grammar [18, 19].

Behavioural disturbances associated with ASD include self-injurious behaviour, aggression, temper tantrums, psychiatric symptoms, and pica. Other conditions associated with ASD are mental retardation, seizure disorders, cerebral palsy, fragile X chromosome, tuberous sclerosis, untreated phenylketonuria, neurofibromatosis, and congenital rubella [2, 3].

The incidence of AD varies between 2–15 per 10 000 births, depending on the criteria used for diagnosis. Males are four to five times more affected than females, but females are more likely to exhibit more severe mental retardation. The disease has been identified internationally with no ethnic propensity [5].

The onset of AD usually occurs before three years. The expression of symptoms varies widely. To be diagnosed as autistic, a patient must exhibit a specified number of symptoms, although not all of them must necessarily be present at the same time or to the same degree. The criteria described in the DSM encompass qualitative impairments in social interaction and communication, as well as deviant patterns of behavior, interest, or activities. Parents are important aids in diagnosing AD, as they are usually the first to be concerned about disturbed development of their child: impaired communication lack of social relationships and imaginative play, and to a lesser extent, hearing impairment and delay in attaining milestones. The mean age noted for these deviations is 17 months and the mean age for final diagnosis is 44 months [5, 7, 20].

Strong evidence suggests that autistic disorder (AD) is an organically based neurodevelopmental disorder associated with abnormalities in brain structure and function. Characteristic findings are a reduced number of Purkinje cells in the posterior inferior regions of the cerebellar hemispheres, truncation in the dendritic tree development of neurons in the limbic system and hypoplasia of cerebellar lobules VI and VII [14–17]. Multiple indices support a genetic basis for AD. Twin studies find a high concordance rate in monozygotic twins. Although the recurrence risk for AD following birth of an autistic child is only 3%, this risk is 60–100 times greater than the base rate for AD in the general population. The higher prevalence in males suggests an X-linked mode of inheritance, but a study by Hallmeyer et al. could not verify any moderate to strong AD gene effect on the X chromosome [21–25].

Orthodontic Management

The main challenge to the orthodontic team may be the reduced ability of autistic patients to communicate and relate to others. Further problems include uneven intellectual development, peculiar repetitive body movements, hyperactiv-

ity, limited attention span, and a low frustration threshold that may lead to temper tantrums or bizarre vocalization. Although there appears to be no experimental verification, several publications described autistic individuals as having a higher threshold to pain, concluding that short procedures may be carried out without local anesthesia. On the other hand, there is agreement that patients with AD exhibit tactile and auditory hypersensitivity, and may have exaggerated reactions to light and odors. Patients with AD tend to dislike changes in their environment and need sameness and continuity; they may react with tantrums over small environmental changes [25–30]. A higher degree of lateral vision in autistic individuals is mentioned by Kopel [26] who concluded that all lateral movements toward the patient are potential distractions and should therefore be avoided. Some autistic individuals prefer using their peripheral vision, because they get more reliable information when they look from the corners of their eyes [5].

It has already been reported that malocclusions occur more often in physically or mentally handicapped children than in healthy individuals. Specific handicapped groups, such as those with Down's syndrome or cerebral palsy, have been associated with increased frequencies of particular dental features [31]. Oreland et al. [32] found a higher prevalence of dental malocclusion in the severely mentally retarded compared with physically handicapped children, and concluded that the mental condition is more important for the orthodontic status than the medical diagnosis. In addition to a displeasing appearance, malocclusion may compromise all aspects of oral function and generate adaptive alterations in chewing, swallowing or speech.

The spectrum of methods used for pain and anxiety control during orthodontic treatment of the autistic child may be divided into conscious methods (such as oral, intramuscular, inhalation with nitrous oxide and oxygen and intravenous sedation) and unconscious methods which include intravenous or inhalation deep sedation and general anesthesia (GA) with endotracheal intubation [33].

Becker and Shapira [34] have defined the main problems that may occur during orthodontic treatment and have shown how, using these methods, treatment delivery is possible for many disabled children. Experience shows that most disabled children approach treatment with exaggerated levels of apprehension, far more than normal orthodontic patients. Accordingly, these patients must be approached with understanding and compassion to gain their trust.

The first several visits are directed towards raising the patient's confidence and determining the maximum level of compliance that is achievable. Far more important and critical in the long-term is the level of parental motivation and the ability, willingness and availability of the parent to carry out the new responsibilities posed by the orthodontic treatment of their child. Only after these assessments have been made during the first few visits, can the orthodontist establish reasonable individual goals on a modular, stage-by-stage basis, which need to be reassessed at the completion of each stage, being prepared to change these goals if needed. At the same time, an estimate of the most suitable way (behaviour management, sedation or GA) to perform the more difficult procedures, such as impressions or bracket bonding may be made. Perfection is not often achievable in these patients, but this need not deter an attempt to improve their occlusion. The practitioner should understand the particular relevance in this context of the concept that the patient may benefit substantially from each treatment goal achieved, even when some may be beyond his/her reach [34].

The placement of bonded fixed appliances demands strict control of the intra-oral environment and any adverse behavioural tendency must be overcome in order to permit its successful conclusion. The means by which the adverse behavior was overcome – the adjunctive modality, in order to place the appliances, was recorded as: GA, general anesthesia; SED, sedation; BM, behavioural modification techniques alone [31].

Jackson believed that children with a learning disability 'should not be turned away merely because a really good result from an orthodontic standpoint is not possible'. Should more severely disabled children be discounted? These children and by association many of their parents, already suffer from a social stigma and a poor dental appearance accompanied by impaired oral functions may lead to a further negative social response [35].

Becker and Shapira [34] used conscious sedation or GA for the most difficult procedures needed during the orthodontic treatment of disabled patients. In the same article, they reported that it was policy to limit the use of GA, preferring to work on a fear-free child with unaltered normal reflexes. The use of Midazolam in the form of nose drops combined with nitrous oxide was the preferred sedation modality.

Jackson [35] was the first to suggest using GA for the placement of orthodontic bands. Chadwick and Asher-McDade claimed that the majority of the more profoundly disabled patients are able to tolerate the adjustment of an appliance, but are unable to keep still for long enough to place brac-

kets and bands. They presented two cases of mentally retarded patients whose bonded appliance were placed under GA, but whose further maintenance was provided in the dental chair by behavioural modification techniques [occ. 36].

The present authors, in agreeing with this approach, strongly emphasize that the decision to perform the most difficult procedures under conscious sedation or GA should be taken solely on condition that the patient has proved to be able to undergo short visits, such as those needed for the appliance adjustments, by BM techniques alone [33].

The autistic patient's need for continuity may require several visits to the dental office prior to the treatment appointment to familiarize the autistic patient with the facility and to establish a routine. Gradual and slow exposure to the dental environment with nonthreatening contacts is recommended. Parental presence in the clinic area is usually discouraged.

It was shown that an ascetic physical environment effectively decreased those negative behaviors and it was therefore speculated that austerity and order in the surrounding setting would have a soothing effect on the patient. When translating this into an orthodontic visit it is rather unrealistic to demand a specially designed operatory for patients with AD; however, it may be feasible to treat the patient in a quiet, shielded single operatory versus an open-bay arrangement, with reduced decoration and dimmed lights [26, 30].

Because of the autistic patient's limited attention span, short, well-organized appointments should be planned and the waiting time should not exceed 10–15 minutes to avoid upset. To address the autistic individual's preference for sameness and aversion to change, a routine should be established by maintaining days, times, and personnel for each visit.

Discussions of any aspect of the actual work should be avoided during its course. Light background music might be beneficial. Anyone participating in the procedure should minimize movements because the autistic child is easily distracted [27, 30, 36, 37].

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Address for correspondence:

Katarzyna Olszewska
Ułanów 21/28
20-554 Lublin
Poland
E-mail: catieol@interia.pl

Received: 17.08.2011
Revised: 28.11.2011
Accepted: 30.11.2011

Praca wpłynęła do Redakcji: 17.08.2011 r.
Po recenzji: 28.11.2011 r.
Zaakceptowano do druku: 30.11.2011 r.