Use of alternative communication with pictures for outpatient periodontal treatment in fragile X syndrome patients

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Abstract:
The purpose of this article is to discuss the clinical management and behavior during periodontal treatment in two patients with fragile X syndrome (FXS) using alternative communication with pictures (ACP). Both patients had a history of previous dental care only possible under general anesthesia. The ACP was used to anticipate the activities to be carried out, promoting assistance, improving the professional–patient communication, and decreasing the stress of the patient. It was possible to carry out outpatient care without oral sedation and containment/physical restraint in both patients, being surpassed the communicative and behavioral difficulties. These case reports allow us to re-think dental care under general anesthesia or other invasive methods for patients with FXS. Therefore, the ACP is an important mediator tool that can facilitate the insertion and the management of patients with FXS, allowing the dental care outpatient clinic to promote oral health and quality of life for these patients, improving adherence to periodontal treatment and the periodontal maintenance for oral hygiene.

Key words:
Alternative communication, communication disorders, fragile X syndrome, periodontal maintenance

INTRODUCTION
Fragile X syndrome (FXS), also known as Martin–Bell syndrome, is a hereditary disorder of development linked to the X chromosome.¹ There is a change next to the final third of the long arm of this chromosome,² which gives an appearance of “fragile.”³ This occurs in the region of the fragile X mental retardation-1 gene.⁴ It presents repetitions (between 56 and 200 repetitions)⁵ of trinucleotide (cytosine-guanine-guanine),⁶⁷ leading to the silencing⁸ and the almost complete loss of protein (fragile X mental retardation protein).⁹

FXS is the most frequent cause of intellectual disability due to heredity, and estimates suggest a frequency between one in 1250–4500 in men and one in 2500–9000 in women.¹⁰¹¹ Most of the individuals with this syndrome have behavioral manifestations as well as other variables, including attention deficits, hyperactivity, impulsivity, anxiety, the behavior of self-aggression, and autism spectrum disorder (ASD).¹²

The behavioral issues promote a dental care complex,¹³ with the deficient and / or absent communication in these patients.¹⁴ In this case, one of the options for dental care is to submit the patient for general anesthesia.¹⁵ These difficulties of communication mean that these patients have less access to the dentist, which can result in worse dental conditions with subsequent loss of teeth.¹⁶¹⁷

However, behavioral approaches to promote and develop an alternative communication, as with the Treatment and Education of Autistic and related Communication-handicapped children (TEACCH),¹⁸ Son-Rise Program,¹⁹ and Picture Exchange Communication System (PECS)²⁰ are promising methods of intervention in patients with ASD. During dental care, these strategies can facilitate patient–professional contact²¹ and allow dental care ambulatory.²²

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The use of alternative communication with pictures (ACP) in dentistry for patients FXS was not reported in literature so far. Therefore, the aim of this article is reporting the clinical and behavioral management in the outpatient periodontal treatment of two patients with FXS using ACP with the support of parents and/or caregivers according to CARE guidelines for case reports.\[23\]

**CASE REPORT**

The following shall be dealt with clinical data, medical and dental history, as well as the clinical management of two patients with FXS treated in outpatient clinics [Table 1]. Before any procedure, parents/guardians of the patients were instructed about the actions involving the dental treatment, and they signed a term of free and informed consent and authorization for the publication of clinical data in this study.

The use of ACP adapted to dental treatment, and subsequent periodontal maintenance, was selected for treatment and intervention for both patients. The augmentative and alternative communication is a form used for patients with different degrees and levels of communication impairment, such as among them people with ASD.\[26\] It is an alternative communication because it is another option of communication and augmentative communication because it can increase the communicative repertoire.\[26\]

Zink *et al.*\[23\] initially described the use of alternative communication in dentistry, where pictures were used to anticipate the result of dental treatment of people with ASD. This communication strategy is to encourage the treatment in an outpatient environment by minimizing the use of physical restrictions, sedation, and forwarding for assistance under general anesthesia, promoting greater frequency of outpatient care and decreasing the indication and the queues for treatment at the hospital level. Zink *et al.*\[23\] found that the anticipation using images favors the acceptance and understanding of dental treatment for a person with ASD.

In clinical cases reported, we opted for choices involving the use of pictures associated with dental procedures,\[23\] which consist of the following three parts.

**First part**

Preparation of the images: Images can be either photograph, as pictures and/or pictograms (representations of objects and concepts translated into a graphical form extremely simplified), addressing the dental context. For example, it was represented at low speed along with the brush of Robinson as a reference to professional prophylaxis. Presentation of dental instruments, such as the triple syringe, high-speed dental handpiece, clinical mirror, exploratory probe, gloves and others. The pictures from the dental office, as with images of the dental chair, reflectors and a photograph of the dentist with and without a mask, images of dental auxiliaries, were obtained. These images can be plasticized or pasted on another role (such as a card). Velcro® can be added or magnets to facilitate the handling by parents and/or caregivers and also by the patient.

**Second part**

The pictures are given to parents and/or caregivers. They are instructed on how to set up a panel of routine to the patient (also a panel of daily activities). These pictures can be present on a panel as a photo album, attached to a metallic structure (in the case of magnets), or in electronic devices (cell phones or tablets). Pictures were presented to anticipate each procedure: initially, sit in the chair, open the mouth, high-speed dental handpiece, anesthesia, and isolation of the operative field.

Nevertheless, this does not mean that you must submit all images in all consultations; in most cases, the skills learned do not need to be presented. The whole image must be complemented with verbal information and positive reinforcement (stimulation) by parents on the way to the dental office. This procedure should be done in the day and on the eve of care dental care. The positive reinforcement should be motivated with words such as “very good!”

**Table 1: Characteristics of the patients in this study**

|                         | Patient 1                                      | Patient 2                                      |
|-------------------------|-----------------------------------------------|-----------------------------------------------|
| General data            | 29 years old, male gender, leukoderma         | 19 years old, male, melanoderma               |
| Diagnosis               | ICD Q.99.2 and association with ICD F84 (fragilex syndrome associated with autism spectrum disorder) | Breathers and atypical swallowing              |
| Systemic condition      | Oral breathing, atypical swallowing, and astigmatism | Absent                                        |
| Medication              | Tegretol® 100 mg/day                          | Without verbal communication                  |
| Communication           | Features verbal communication                 |                                               |
| Global behavior         | Very anxious, but not aggressive. Difficulty in social interaction, communication and repetitive movements and stereotyped with their hands and echolalia |                                               |
| Motor behavior          | Regular fine motor coordination               | Poor fine motor coordination                  |
| Tooth brushing          | Own, with parental supervision                | Performed by the mother three times per day, however unsatisfactorily |
| Previous dental visits  | Only under general anesthesia                 | Poor dental condition (DMFT=10), with gingivitis and calculus full mouth |
| Oral condition          | Clenching and bruxism spontaneous, pseudomacroglossia, regular dental condition (DMFT=3), with gingivitis and calculus full mouth |                                               |
| Radiographic examination| Presence of 3rd and 4th molar included. Periapical lesions in 15 | Cavity lesion in 36 and 46, both with furcation lesions |
| Behavioral management   | ACP, technique talk-show-do and ludic aid with puppets | ACP countries and talk-show-do technique |
| Proposal for dental care | Outpatient dental treatment carried out from the use of ACPs for dental care and subsequent periodontal maintenance |                                               |

ICD – International Classification of Diseases; DMFT – Index for decayed, missing, filled teeth; ACP – Alternative communication with pictures; 15 – Number corresponding to the second premolar tooth right upper; 46 – Number corresponding to the first lower right molar
**Third part**

Complementation with visual support (talk-show-do). The patient can take the images on the day of dental care. The dentist can show the physical object, such as the triple syringe among others, to facilitate the understanding of the relation of images with the context. The dentist needs to observe if the patient is associating the specific object in the image displayed, i.e., if he/she believes that by showing the image of the chair, the patient understands that it is the dental chair where you need to sit down. If you do not understand, you will need to strengthen the image to the concrete object.

To check if there is evolution/adoption of the proposed treatment with the use of ACPs, a scale was used in the reported cases to assess the behavior of the patients’ faces in different situations during clinical practice.\(^{25}\)

In both cases, 13 items were evaluated that correspond to the actions of communicative intent and interaction with the dentist during the dental treatment: scores of 0, 1, and 2 were applied when the patient did not present any action/reaction (score 0), or restricted way (score 1), or clear (score 2). Each patient could have a minimum score of 0 and a maximum of 26 points: the values close to zero show great loss of communication, whereas those close to 26 imply greater understanding by the patient.

These data were collected in the first consultation and after three consultations for patient 1 (P1) and two consultations for patient 2 (P2), using this interval for alternative communication [Table 2].

We observed an exponent growth in communication, especially with P2. The use of pictures enabled better expression and interaction with the patients, mainly for acceptance of the activities proposed by the professional and reduction in anxiety to a signal (exchange of shifts). The pictures promoted anticipation of activities and enveloped the patient with the context of the treatment and dental environment, improving the interaction with the dentist. The patient 2 had improved gestural reproduction (mimicry) after using the ACP, and such movements are important for the assimilation of hygiene and periodontal treatment at home. P1 had initially scored 6 and after the use of the ACP, scored 19, and P2 had an initial score of 2 which rose to 16 after the use of the GPA.

However, the use of the ACP has proved to be effective for the development of dialogs and also for the understanding of abstract thinking, possessing a low effect on verbal communication (such as the production of words or phrases in preparation) for both patients. This is in agreement with the objective of this ACP form. Therefore, outpatient treatment was possible with periodontal maintenance to promote oral health for both patients.

Despite the various benefits of using the ACP, some limitations of this case report were as follows: there is a need for parental/caregiver support to reinforce alternative communication at home. Depending on the cognitive capacity of the patient, more or fewer consultations may be required to adapt to the ACP. Even in patients with advanced degrees of intellectual disability or early / high age, the use of ACP can promote benefits, as evidenced by a systematic review in preschoolers and ASD children.\(^{23}\) More clinical trials are necessary to elucidate better how ACP should be addressed during dental treatment in an outpatient clinic.\(^{23}\)

**DISCUSSION**

FXS is one of the genetic changes that most often leads to cognitive impairment, which can affect and derail outpatient dental care. FXS patients may have behavioral manifestations that are also compatible with ASD.\(^{13}\) The use of ACP promoted an exponential growth in the degree of understanding and communication in both cases [Table 2], being that P1 presented an evolution of 316% and P2 of 800%. These data signaled a satisfactory resolution of the clinical cases that allowed dental care and periodontal maintenance in an outpatient clinic, which was possible only in the surgical center previously.

The use of communication with a picture can be used for any person with changed cognitive development.\(^{24}\) However, for some patients, especially those that require more concrete references, it may be necessary to associate visual support or physical contact to allow the assimilation with the object or with the situation to be presented, for example, the dental care context. The complementation with the use of the technique speak-show-do helps in clinical management and shows great additional benefits to the use of alternative communication with the images.\(^{23}\) It should be emphasized that individuals

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**Table 2: Observation of communicative behavior during dental care**

| Evaluated item                          | Examples of situation                                                                 | P1 | A | B | P2 | A | B |
|----------------------------------------|---------------------------------------------------------------------------------------|----|---|---|----|---|---|
| Interaction with the evaluator         | Does the patient maintain that (in) difference before the information passed?         | 1  | 2 | 0 | 2  |    |    |
| Communicative intention                | Does the patient try to communicate? Does he communicate with interpreters and/or speech? | 1  | 2 | 0 | 1  |    |    |
| Eye contact                            | Does the patient remain with eye contact before information?                          | 1  | 2 | 1 | 2  |    |    |
| Production of words                    | Does the patient produce words?                                                      | 0  | 1 | 0 | 1  |    |    |
| Exchange of shifts                     | Was the patient awaiting the completion of information and then responded/spoke?      | 0  | 2 | 0 | 2  |    |    |
| Participation in dialog                | Did the patient have a question/doubt?                                              | 0  | 0 | 0 | 0  |    |    |
| Keep dialog                            | After the answer to a question, is the patient able to keep the conversation going? | 0  | 2 | 0 | 1  |    |    |
| Logical-temporal sequence              | Is the patient able to construct sentences properly?                                 | 0  | 1 | 0 | 0  |    |    |
| Understanding of concrete situations   | The patient understands concrete situations, such as: “Show your teeth for me“       | 1  | 2 | 1 | 2  |    |    |
| Understanding of abstract thinking     | The patient understands abstract situations, such as, “Have you ever had a toothache?" | 0  | 0 | 0 | 0  |    |    |
| Time of attention                      | The time that the patient can stay in the dental chair, without dispersing           | 1  | 2 | 0 | 2  |    |    |
| Acceptance of activities               | Does the patient allow the realization of the proposed activities?                   | 0  | 2 | 0 | 2  |    |    |
| Mimicry                                | Is the patient able to assimilate and repeat movements, such as toothbrushes?        | 1  | 1 | 0 | 1  |    |    |
| Total                                  |                                                                                        | 6  | 19| 2 | 16 |    |    |

A – After; B – Before the use of an adapted form of alternative communication with pictures; P1 – Patient one; P2 – Patient two
with higher cognitive ability are adhering more quickly to the understanding of images. This is one of the difficulties that may be encountered from a lack of interest or understanding of the image by the patient. Each person with ASD is unique, and the difficulties will have to be analyzed individually and new strategies to approach should be outlined. A barrier that can be developed is the nonmembership of this communication strategy by parents, and this should be analyzed with caution because it may prevent/hinder the achievement of periodontal treatment. With the use of ACP as a strategy of dental approach, it is expected to improve the communication between professionals and patients, decreasing anxiety through the predictability (anticipation of facts). This anticipation employing pictures makes the action more predictable, and patients can adopt it into the daily routine, assisting in the implementation of the outpatient periodontal treatment.

Other professionals who maintain contact with the patient (such as speech pathologists, psychologists, and occupational therapists) can also assist in anticipation for a visit to the dentist, inserting activities that relate to dentistry in the eve of dental care, for example. FXS patients may present ASD characteristics, which necessitates multidisciplinary treatment that also works with the development of an alternative communication. Therefore, the predictability of acceptance and development by individuals with this form of communication is favorable.

We have demonstrated that the use of ACP during outpatient periodontal treatment was effective for patients with FXS. The dental biofilm control of patients with FXS is a clinical challenge and should be the focus of future work. During periodontal maintenance, measures for effective control of biofilms are extremely important for these patients. A systematic review did not demonstrate the superiority of electric toothbrush over manual toothbrush (for people with disabilities in general), and modified brushes and chlorhexidine (in concentrations of 0.12% or 0.2% as mouthwashes and/or spray) are more effective than conventional brushes and placebos. For the mechanical control of dental biofilm, a toothbrush with three heads can be more effective instead of a single head. For all these steps, the ACP can be used.

The initial approach through ACP in public health and health supplement points to be a problem-solving strategy and proposes to reduce costs and risks associated with surgical periodontal treatment in surgical centers under general anesthesia. However, further studies that prove the effectiveness of this technique, as well as the development of other tools for integration and inclusion of the patient with a drip in outpatient periodontal treatment and periodontal maintenance, are needed.

Implications for research The use of ACP during periodontal care in a patient with FXS can be a broad field of research:
1. Another syndrome may be considered and applied to the treatment of other dental needs
2. Assessing the impact of such treatment on private practices and the public network maybe the target of future studies
3. New research is also suggested that can optimize both the use of ACP and also the periodontal care of patients with FXS.

Implications for clinical practice The ACP approach during patient care with FXS allows several benefits in clinical practice, such as:
1. Favors the reception and makes the periodontal treatment of these patients feasible
2. Avoids performing periodontal procedures (as with other dental needs) in the operating room for patients with FXS or other disabilities
3. Promotes oral health and quality of life for patients with FXS from nonsurgical periodontal treatment in an outpatient setting
4. Expands the strategies of periodontists to care for patients with cognitive deficits, such as patients with FXS.

CONCLUSIONS
We conclude that the use of ACP led to dental care and periodontal treatment in an outpatient clinic for patients with FXS and ASD, even with a history of dental care at the surgical center under general anesthesia by their behavioral changes. This form of behavioral interventions is a viable and satisfactory alternative for outpatient periodontal treatment. It is a low-cost method that favors acceptance and adherence of patient’s and promotes success in the preventive and curative treatment.

Declaration of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form, the patients/parents have given their consent for their images and other clinical information to be reported in the journal. The patients/parents understand that names and initials will not be published, and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest
There are no conflicts of interest.

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