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CLINICAL RESEARCH

Satisfaction of patients with amyotrophic lateral sclerosis with an oral appliance for managing oral self-biting injuries and alterations in their masticatory system: A case-series study

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ABSTRACT

Statement of problem. About 10% of patients with amyotrophic lateral sclerosis (ALS) are candidates for oral treatment specifically because of traumatic injuries in the lips, cheeks, or tongue due to self-biting. However, patients with ALS have a prevalence of temporomandibular disorder (TMD) similar to that in the general population.

Purpose. The purpose of this case-series study was to determine the degree of satisfaction of patients with ALS with an oral appliance for managing oral self-biting lesions or symptoms related to TMDs. This study also assessed the degree of improvement of the chief complaint and the compliance with and adverse effects of this treatment.

Material and methods. Eleven patients with ALS who sought oral treatment because of oral self-biting or TMD-related symptoms were included. A custom complete-coverage acrylic resin device was fabricated and fitted to each participant. A follow-up visit was planned for 3 months after the placement of the oral appliance, at which point the patients would rate the degree of improvement or worsening of the chief complaint and their degree of satisfaction with the treatment. A 1-sample *t* test was used to assess whether the degree of improvement of the chief complaint was significant.

Results. Participants reported a mean of 61% (95% confidence interval [CI] 38% to 84%) improvement of the chief complaint and a mean of 84% (95% CI 72% to 97%) satisfaction with the treatment. The mean rate of compliance was 62% (95% CI 40% to 84%) of the recommended time, and only a few adverse effects were reported.

Conclusions. Participants with ALS were highly satisfied with the use of an oral appliance to manage oral self-biting or TMD-related symptoms. Adherence to this treatment was high, and no major adverse effects were observed. (J Prosthet Dent 2019;121:631-6)

Amyotrophic lateral sclerosis (ALS) is a neurodegenerative disease characterized by progressive muscular paralysis reflecting the degeneration of motor neurons in the primary motor cortex, corticospinal tract, brainstem, and spinal cord.[1] Its incidence rate is approximately 1.4 and 2.1 per 100 000 persons/year in Catalonia and Europe, respectively.[2,3] Because ALS is rapidly progressive in nature, life expectancy is 3 to 5 years after diagnosis, although approximately 10% of patients with ALS survive for 10 or more years.[4] The typical clinical characteristics of ALS are variable and depend on whether the site of onset is spinal, bulbar, or respiratory. Most patients with ALS have a spinal onset, referring with weakness, muscle atrophy, and fasciculations due to lower motor neuron involvement and hyperreflexia and hypertonia due to upper motor neuron involvement. In about 20% of patients, weakness starts in the bulbar muscles, with dysarthria, dysphagia, and tongue fasciculations. Bulbar onset ALS has a poorer

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Clinical Implications

Patients with amyotrophic lateral sclerosis referred for oral self-biting or temporomandibular disorder symptoms can be managed efficiently by means of an acrylic resin device.

prognosis because of swallowing difficulties, weight loss, aspiration, and respiratory involvement, with poorer adaptation to noninvasive ventilation. About 3% to 5% of patients have a respiratory onset, referring with orthopnea or dyspnea and mild or even absent spinal or bulbar signs.[1]

About 10% of patients with ALS are candidates for oral treatment specifically because of their disease. The chief complaint may include traumatic injuries to the lips, cheeks, or tongue due to self-biting in the case of bulbar involvement.[5] However, patients with ALS have a prevalence of temporomandibular disorder (TMD) similar to that of the general population. Almost 50% report grinding and clenching, and 9% may be diagnosed with myalgia.[5] Furthermore, bulbar involvement is associated with the perception of functional limitation of the masticatory system, especially when masticating tough food or chicken or when swallowing or talking.[6]

The dentist should be part of a multidisciplinary team for the management of patients with ALS.[5-9] The treatment options available to the dentist include the use of a palatal lift and/or palatal augmentation prosthesis to improve dysarthria, as described in several patients with ALS.[10] Other types of oral appliances have been reported in clinical reports to assist with noninvasive ventilation or to control drooling.[11,12] However, the authors are unaware of a study that considered how to manage traumatic injuries to the lips, cheeks, or tongue because of self-biting in patients with ALS. Custom oral appliances, acrylic resin devices, or mouthguards have been used in other patients with oral self-injury.[13-28] For example, an acrylic resin device with a labial bumper for displacing the lower lip forward was effective in preventing traumatic lesions in the lip due to self-biting in an adult with severe neurological impairment.[13] Oral appliances are also recommended for the treatment of TMDs.[29-34]

The purpose of the present study was to determine the degree of satisfaction in patients with ALS after treatment with an oral appliance to manage oral self-biting or symptoms related to TMDs. This study also assessed the degree of improvement in the chief complaint, the change in the quality of life due to changes in the chief complaint, and other aspects of the treatment including compliance, side effects, and technical failures. The research hypothesis was that patients with ALS are satisfied with the use of an oral appliance to manage oral self-biting or symptoms related to TMDs.

Table 1. Clinical characteristics of patients with amyotrophic lateral sclerosis according to chief complaint

	Chief Complaint	
Clinical Characteristics	**TMDs (n=7)**	**Self-biting (n=12)**
Sex (% of male)	43	17
Median age (years)	61.8	61.7
Bulbar-onset ALS type (%)	43	58
Bulbar involvement (%)	71.4	100
Median time elapsed since symptom onset (mo)	27.2	24
Median time elapsed since ALS diagnosis (mo)	18.7	13.2
Botulinum toxin (%)	14	25
Noninvasive ventilation (%)	0	33
Oral feeding (%)	100	58
Use of tube feeding (%)	14	50
Median number of missing teeth	7	5.5

ALS, amyotrophic lateral sclerosis; TMDs, temporomandibular disorders.

MATERIAL AND METHODS

Nineteen adult patients diagnosed with ALS according to the revised El Escorial diagnostic criteria and who were referred with alterations of the masticatory system were invited to participate in this prospective case series.[35] All patients were attending the Motor Neuron Disease Unit of the Bellvitge University Hospital between September 2015 and July 2016 and had participated in previous studies.[5,6] Patients who could not be treated with an orofacial device because of the advanced stage of their disease, those with severe periodontal disease, or those without a sufficient number of teeth to hold an oral appliance were excluded. The nature of the study was explained in full to all participants, and all signed an informed consent form approved by the Bellvitge University Hospital Ethics Committee (Code PR259/15). All experiments were carried out in accordance with the principles of the Helsinki Declaration.

One dental clinician (N.R.-P.) recorded chronologic variables including the time elapsed since symptom onset and since the ALS diagnosis. Demographics such as sex, age, and a phenotypic classification according to the site of onset were recorded. Medication, use of mechanical ventilation, and gastrostomy were also recorded (Table 1).

All participants were examined by the same dental clinician (N.R.-P.) and answered the symptom questionnaire of the Diagnostic Criteria for Temporomandibular Disorders protocol.[36] The clinical examination included the measurement of maximum opening, protrusion, and laterotrusion; palpation and auscultation of the temporomandibular joints; and palpation of the masticatory muscles.[36-38] Participants were also assessed by means of a questionnaire about awareness of clenching/grinding and self-biting of the tongue, lips, or cheeks with dichotomous no/yes answers. Patients were

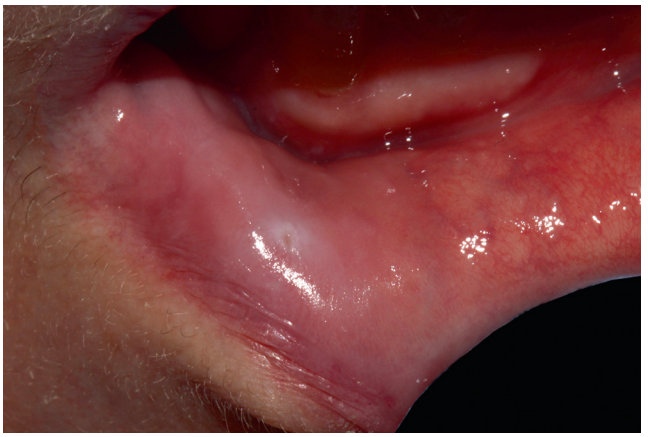

Figure 1. Lower lip self-biting lesion in patient with amyotrophic lateral sclerosis.

asked about the chief complaint because most had been referred with more than one. They were assigned to the TMDs or self-biting group accordingly. The most frequent complaints were lower lip self-biting, grinding/clenching, and masticatory muscle pain (Fig. 1).

The oral appliance was a custom complete-coverage acrylic resin occlusal device with a flat occlusal surface in contact with all antagonistic teeth at habitual closure and providing anterior guidance in lateral and protrusive movements (Fig. 2).[29,32] It was placed on the maxillary or mandibular arch depending on the chief complaint and on the dental conditions (Table 2). Alginate impressions of the maxillary and mandibular arches were used to make gypsum casts. Among the 19 participants, difficulties making the impression were encountered in 5 individuals, mainly because these patients were not able to open their mouth wide enough or keep it sufficiently open while the impression material was setting (Table 2). If the chief complaint was self-biting the lower lip or cheeks, the acrylic resin device could include a buccal bumper to move the lower lip forward (Fig. 2). If the chief complaint was self-biting the tongue, the acrylic resin device could include a lingual bumper to move the tongue backward. All oral appliances were fabricated by the same dental technician and fitted and adjusted by the same dental clinician (N.R.-P.). The patients were instructed to use the oral appliance every night during sleep and/or during the day at times they considered helpful.

During treatment, patients with technical complications involving the oral appliance and/or adverse effects involving the masticatory system were seen by the same dental clinician (N.R.-P.). The number of additional dental visits, the reason for each extra visit, and the number of oral appliance repairs in the dental laboratory were recorded.

The follow-up visit was planned for 3 months after the patient had worn the oral appliance normally. This follow-up consisted of a questionnaire to assess compliance as the percentage of time the oral appliance was used with respect to the recommended time and the adverse effects reported by the patients. This questionnaire also assessed the degree of improvement or worsening of the chief complaint after 3 months of oral appliance treatment by means of a visual analog scale (VAS), for which the patient made a mark on a 10-cm line anchored by "extreme worsening" (-100%) or "completely improved" (+100%) at either end and "no change" in the center of the line (0%).[39] The change in quality of life because of changes in the chief complaint after 3 months of treatment was assessed by using a similar VAS. Finally, patients also rated their degree of satisfaction with treatment by using a VAS anchored by "extremely dissatisfied" (0%) or "completely satisfied" (+100%) at either end.

The outcome variables were the degree of improvement of the chief complaint, the change in quality of life because of changes in the chief complaint, and the degree of satisfaction with the treatment. The degree of improvement of the chief complaint and the change in quality of life because of changes in the chief complaint were assessed by using a 1-sample *t* test. The degree of satisfaction with the treatment was expressed as mean (95% confidence interval [CI]) (α=.05).

RESULTS

Among the 19 treated participants, 8 were excluded because they did not attend the 3-month evaluation (7 from the self-biting group and 1 from the TMD group). Of these 8 participants, 4 did not attend the evaluation because their disease had worsened, 1 because the individual considered the oral appliance no longer necessary, 1 because she had developed hypersalivation and had stopped using the oral appliance, and 2 because they preferred not to attend the clinic, even though the treatment had apparently improved the chief complaint. Therefore, 11 participants were included in the study and performed their evaluation for a mean of 4.4 months after being fitted with the oral appliance.

The participants reported a mean of 61.2% (95% CI 38% to 84.4%) improvement in the chief complaint (P<.001, 2-tailed 1-sample *t* test) and a mean of 84.3% (95% CI 72% to 96.6%) satisfaction with the treatment. Because of changes in the chief complaint, quality of life improved by a mean of 58.6% (95% CI 23.5% to 93.7%) (P=.004, 2-tailed 1-sample *t* test). Of the 11 participants, only 1 reported a reduction in quality of life because the chief complaint had not improved (Table 3).

Of the participants who attended the 3-month evaluation, 5 had needed at least 1 extra visit because of technical complications with the oral appliance, 3 of them for adjustment and 2 for loosening. Only 1 oral appliance

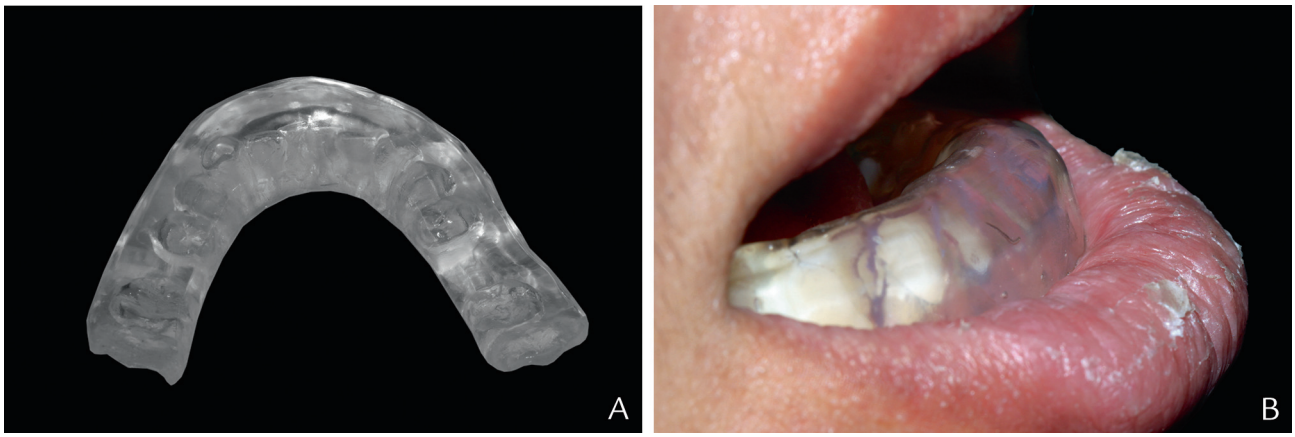

Figure 2. A, Mandibular acrylic resin occlusal device with buccal bumper. B, Device inserted in participant with lip biting.

Table 2. Characteristics of oral appliance and degree of difficulty when making impressions according to chief complaint

Characteristics of the Oral Appliance	Chief Complaint	
	TMDs (n=7)	Self-biting (n=12)
Maxillary oral appliance (%)	29	0
Mandibular oral appliance (%)	71	100
Presence of buccal bumper (%)	14	67
Presence of lingual bumper (%)	0	17
Difficulty making impressions (%)	14	33

TMDs, temporomandibular disorders.

Table 3. Treatment success perceived by participants according to chief complaint

Treatment Success	Chief Complaint	
	TMDs (n=6)	Self-biting (n=5)
Mean (95% CI) improvement of chief complaint (-100 to 100)	56.8 (16.4-97.2)	66.4 (25.7-100)
Mean (95% CI) satisfaction with treatment (0 to 100)	83.8 (62.0-100)	85.0 (64.5-100)
Mean (95% CI) improvement in QoL (-100 to 100)	55.8 (0-100)	62.0 (25.4-98.7)

CI, confidence interval; TMDs, temporomandibular disorders; QoL, quality of life.

needed to be repaired at a dental laboratory because of the lack of retention. The mean rate of compliance was 62.3% (95% CI 40.3% to 84.2%) of the recommended time. The main reason for not having used the oral appliance 100% of the recommended time was discomfort, as reported by 4 participants (36%). Only 3 participants reported no side effects at the evaluation, and the most reported side effect was excessive salivation, affecting 64% of the participants (Table 4). The participant who reported worsening of the chief complaint and had stopped using the oral appliance reported all types of side effects (Table 4).

Table 4. Number of participants (percentage) who reported side effects related to use of oral appliance at 3-mo evaluation according to chief complaint

Side Effects	Chief Complaint	
	TMDs (n=6)	Self-biting (n=5)
Excessive salivation	3 (50%)	4 (80%)
Dry mouth	1 (17%)	0 (0%)
Tooth discomfort or pain	2 (33%)	1 (20%)
Mucosal irritation	1 (17%)	0 (0%)
Muscular discomfort	1 (17%)	0 (0%)
TMJ discomfort or pain	1 (17%)	0 (0%)
TMJ sounds	1 (17%)	0 (0%)
Bite change	2 (33%)	0 (0%)
Other	1 (17%)	0 (0%)
At least 1 side effect	4 (67%)	4 (80%)

TMDs; temporomandibular disorders; TMJ, temporomandibular joint.

DISCUSSION

The results of this study suggest that patients with ALS are satisfied with the use of an oral appliance to manage oral self-biting or symptoms related to TMDs, and therefore, the hypothesis was not rejected. The effectiveness of this treatment can be demonstrated by a mean of 61% improvement of the chief complaint, implying improved quality of life, and by a mean of 84% degree of satisfaction with the treatment. Furthermore, compliance was high, and few and nonrelevant side effects or technical complications were detected.

The effectiveness of an oral appliance in preventing self-biting has also been reported in other neurological diseases but not in ALS and only in case reports.[13,14,17-24] However, the authors are unaware of previous prospective case series studies that evaluated effectiveness in patients with ALS. In some individuals, the increased vertical dimension produced by the oral appliance was sufficient to avoid oral self-biting because the lips, tongue, or cheeks did not invade the interocclusal space. In others, a bumper was needed to separate the lower lip from the teeth because the increased vertical dimension was not sufficient to stop the soft tissues encroaching on

the interocclusal region. Before fabricating the oral appliance, the dentist should explore the placement of the tissues being traumatized in several mouth opening increments, from the intercuspal position to the resting position, to determine the required increase in vertical dimension increase and whether a bumper is required in the oral appliance.

The degree of patient satisfaction with the acrylic resin device and the compliance rate of participants with ALS were similar to those reported in those without ALS but with TMD symptoms.[29] Oral appliance therapy is a common approach to manage patients with TMDs. Although the mechanism of action of this approach remains unclear, multiple effects may be present, including allowing an orthopedically comfortable jaw position, reducing masticatory muscle activity and joint loading, and increasing patients' awareness and ability to reduce bad oral habits.[30-34] Therefore, patients with ALS with the chief complaint of clenching/grinding and/or muscular pain can be managed using an acrylic resin occlusal device.

The most common complication encountered in these patients during treatment was difficulty in making impressions due to the evolution of their disease. A poor impression can compromise the quality of the cast and therefore the acrylic resin device. Only one impression of one arch without the antagonist and/or the anterior region of this arch can be made in those who cannot be fed orally and when the dental occlusion has lost its normal function.[13] Intraoral scanning could be an alternative to traditional impression procedure in cases where patients are not able to keep their mouth sufficiently open while the impression material was setting.[40] Another option could be a removable lip bumper fabricated at the chairside without making an impression.[17] Similarly, custom mouthguards have been described as an option for the treatment of self-inflicted oral trauma.[16,25] A self-modeled mouthguard was reported to protect against cheek biting in a patient under orthodontic treatment,[26] although the self-modeled mouthguard is worn on the maxillary arch and this could be inconvenient.

Although the prevalence of ALS is low and the percentage of patients with ALS who require an oral appliance treatment is only about 10%, this disease is highly disabling; patients will appreciate any help that improves their quality of life. The use of an oral appliance permits daily oral care; its maintenance is straightforward, it can be placed and removed easily by the patient or the caregiver, it can be repaired or modified, and treatment can be conducted by a general dentist. Using an oral appliance can help the patient avoid the more extreme solution of extraction of all teeth.[27,28]

Side effects were generally the same as those reported in other studies of oral appliances.[29,37,41] However, excessive salivation could exacerbate the problem of drooling, which is common in ALS. Moreover, sialorrhea itself is already common in ALS and can be treated with amitriptyline, oral or transdermal hyoscine, or sublingual atropine drops.[7]

This study has several limitations. First, no control group was used, and a cause-and-effect relationship between the improvement of the chief complaint and the use of an oral appliance should be assumed with caution. The present findings encourage further studies with appropriate controls to demonstrate the effectiveness of the oral appliance in patients with ALS. Moreover, the initial sample size and the high proportion of dropouts could reduce the validity of these results. The fact that patients with ALS may have difficulty traveling from their homes to the clinic, mainly because of the evolution of their disease, could make it difficult to monitor treatment for longer periods. In the 4 patients excluded because of worsening of the disease, it is not known whether the acrylic resin device was effective or not. This suggests that patients with ALS should use the acrylic resin device in the first phase of their disease and not wait until their condition deteriorates, at which point compliance might decline. An early diagnosis and appropriate dental approach are indispensable to avoid severe injuries to the oral mucosa.[26]

CONCLUSIONS

Within the limitation of this case-series clinical study, the following conclusions were drawn:

1. Patients with ALS were highly satisfied with the use of an oral appliance to manage oral self-biting or symptoms related to TMDs because of the improvement in the chief complaint, which increased their quality of life.
2. Compliance regarding the use of an oral appliance was high, and few side effects and technical failures were observed.

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