

# Is Atypical Swallowing Associated With Relapse in Orthognathic Patients? A Retrospective Study of 256 Patients

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**Purpose:** To determine the association of atypical swallowing (AS) with relapse in patients undergoing combined orthodontic and orthognathic treatment.

**Materials and Methods:** In this retrospective cohort study, the clinical data of all patients who underwent combined surgical and orthodontic treatment from 1998 through 2015 at the University Hospital of Geneva (Geneva, Switzerland) were reviewed. The primary outcome variables were 1) AS and 2) post-treatment relapse. Other variables included, age, gender, dentofacial deformity (facial asymmetry, mandibular deficiency, mandibular excess, maxillary retrusion, open bite), surgical procedure performed (bilateral sagittal split osteotomy [BSSO] with or without Le Fort I osteotomy), and dental interocclusal relations according to Angle.

**Results:** Of the 256 patients investigated, 23 (9%) showed relapse at 1 year after treatment. Multivariate analysis showed that AS status (before treatment, odds ratio [OR] = 9.44,  $P = .005$ ; after treatment, OR = 56.17,  $P < .001$ ; before and after treatment, OR = 20.49,  $P < .001$ ) was significantly associated with relapse. The presence of a pretreatment open bite also was associated with an increased risk of relapse (OR = 0.04,  $P < .001$ ) regardless of AS status.

**Conclusion:** The present study showed that in orthognathic patients 1) AS was associated with relapse regardless of dentofacial deformity or surgical procedure performed and 2) AS can appear after treatment in patients who were initially free of it.

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The success of combined orthodontic and orthognathic management of dentofacial deformities relies on obtaining a balanced and stable dental occlusion while attaining harmonious facial esthetics. Assessment of stability over time requires that postoperative follow-up be continued not only after surgery, but also after removal of orthodontic appliances. Indeed, relapse of certain malocclusions is sometimes

observed several years after carrying out orthognathic surgery, despite acceptable initial postoperative results.<sup>1,2</sup>

Postoperative stability after orthognathic and surgical procedures depends on numerous factors. After comparing different surgical movements with correct malocclusions, Proffit et al<sup>2</sup> proposed a hierarchy of stability and found that some movements are more

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stable and others are more unstable over time. On the one hand, maxillary impaction and maxillary and mandibular advancements are particularly stable; on the other hand, mandibular setback and maxillary expansion are unstable. Nevertheless, the stability of surgical outcomes is multifactorial. Facial balance largely depends on functions that could be the cause of unstable results, despite good anatomic correction. Breathing, swallowing, and chewing disorders can be sources of instability and a potentially important cause of relapse.<sup>3</sup>

Atypical swallowing (AS) is recognized as a potential relevant risk factor for the development of malocclusion in preschool children.<sup>4</sup> This has led to the recommendation to rehabilitate functional orofacial parameters after orthodontic and surgical treatment to favor long-term success.<sup>5</sup>

Normal adult swallowing is performed with the teeth in contact, the tongue pushing the food toward the pharynx, with an ascent of the oropharynx as the food passes to the esophagus. The occlusion is in maximum intercuspation, the elevating muscles of the mandible are contracted, and the entire lingo-hyoid mass is lifted. There is no interposition of the tongue between the dental arches.<sup>6</sup>

Although there are no consensual diagnostic criteria, infantile swallowing (tongue thrust or AS) has been described by many investigators as a thrust or interposition of the tongue between the dental arches, the absence of dental contact, and the contraction of the muscles of the lips and cheeks. Given that the circumoral contraction also has been reported in normal swallows, the main diagnostic criterion retained is the forward movement of the tongue against or between the anterior teeth.<sup>7-9</sup> Infantile swallowing disappears at approximately 3 years of age. If an infantile swallowing pattern persists into adulthood, this is referred to as AS and is considered by some investigators as a risk factor for relapse after the correction of certain malocclusions.<sup>10</sup> This particular form of AS also has been described as “detrimental tongue thrust.”<sup>8</sup> The mechanism put forward is that of the continuous tongue thrust on the teeth, although it is likely that the resting position of the tongue is more important than its activity during swallowing. Phonetic disorders represent the other deleterious consequence of AS and can lead to a true communicative disorder because of difficulties in correctly pronouncing particular phonemes, such as /s/ and /z/, in patients who develop a frontal lisp.<sup>8</sup>

Because the physiopathology of relapse might be similar to the mechanisms that established the dentofacial deformity in the first place, the authors sought to describe whether the presence of AS in patients who underwent combined orthodontic and orthognathic treatment was related to relapse of the malocclusion.

In addition to the association between pretreatment AS and post-treatment relapse, an association between surgery and future AS was postulated. MacAvoy et al<sup>11</sup> studied the effects of acute change in occlusal vertical dimension on intraoral swallowing patterns. Their results support the existence of a central control mechanism for swallowing, which could be modified by peripheral inputs. In other words, orthodontic and orthognathic treatment can induce the development of an AS pattern that was not present before treatment. Thus, the cause-and-effect relation between swallowing and malocclusion remains highly controversial.

This study investigated the relation between AS and the stability of 1-year post-treatment results in a population of patients who underwent combined orthodontic and orthognathic treatment. The main objective of this study was to ascertain whether the presence of AS is associated with post-treatment relapse. The secondary objective was to analyze the cases in which orthodontic and orthognathic treatment induced the development of an AS pattern that was not present before treatment.

## Materials and Methods

### STUDY DESIGN

To address the research purpose, the authors designed and implemented a retrospective cohort study. This study adhered to the Declaration of Helsinki on medical protocol and was approved by the regional ethical review board.

### STUDY SAMPLE

The study population was composed of all patients with dentofacial deformities treated by combined surgical and orthodontic treatment at the Hôpitaux Universitaires de Genève (Geneva, Switzerland) from 1998 through 2015.

Patients were excluded from the study if they had a history of facial surgery, trauma, or treatment for temporomandibular joint disorders, had undergone genioplasty alone, or were lost to follow-up before the 12 month post-treatment period.

### STUDY VARIABLES

The primary outcome variables were AS and post-treatment relapse. Other variables included age, gender, type of dentofacial deformity (facial asymmetry, mandibular deficiency, mandibular excess, maxillary retrusion, open bite), surgical procedure performed (bilateral sagittal split osteotomy [BSSO] with or without Le Fort I osteotomy), and dental interocclusal relations according to Angle.

The diagnosis of AS was based on protrusion of the tongue against or between the anterior teeth, which

was clinically assessed by asking the patient to swallow saliva 3 consecutive times during the same visit while separating the patient's lips manually (Fig 1).<sup>7</sup>

Relapse was defined clinically by a return to the initial malocclusion 12 months after completion of the combined orthodontic and orthognathic treatment, when consolidation of the osteotomies would have been completed. Thus, the definition excludes immediate postoperative complications leading to immediate relapse, such as problems related to consolidation or pseudarthrosis that are clearly unrelated to the presence or absence of AS. Relapse was assigned a binary value of present or absent. In cases with pretreatment open bite, the presence of a post-treatment open bite of at least 0 mm was considered relapse. In cases with pretreatment asymmetry, the presence of a post-treatment midline deviation of at least 2 mm was considered relapse. In cases with pretreatment mandibular deficiency, the presence of a post-treatment change in the molar relation of more than one fourth of a cusp toward an Angle Class II relation was considered relapse. In cases with pretreatment mandibular excess or maxillary retrusion, the presence of a post-treatment change in the molar relation of more than one fourth of a cusp toward an Angle Class III relation was considered relapse. These definitions of relapse are based on data presented by Profit et al.<sup>12</sup>

Patients' medical records were analyzed preoperatively and 12 months after the end of treatment. It was decided to follow the patient up to 1 year after removal of orthodontic appliances, because, according to the literature, most relapses occur during the first year after surgery.<sup>13</sup> Moreover, even patients presenting with relapse at the 1-year follow-up visit refused further follow-up visits, because they were not bothered by the condition and they did not want any further treatment.

#### STATISTICAL ANALYSIS

First, the association of AS before and after surgery was examined to assess the causality of the relation between surgery and AS using the McNemar test. Second, the association between presurgery AS and post-treatment relapse was assessed using univariate logistic regression. Because of the relatively small proportion of cases in which relapse was evident, the number of covariates used to adjust the analyses was limited. The main confounding factors of the association between AS status and relapse were dentofacial deformity (open bite vs others) and surgical procedure. Thus, in the final step of the analysis, a multivariable logistic regression model was used to assess the adjusted association between AS and relapse. Statistical significance was set at a *P* value less than .05.



**FIGURE 1.** Clinical assessment of atypical swallowing (tongue thrust) by asking the patient to swallow saliva while separating the patient's lips manually.

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## Results

Of the 277 patients who underwent a mandibular BSSO with or without a Le Fort I osteotomy, 256 had a 12-month follow-up (140 women and 116 men; mean age,  $24.5 \pm 9.6$  yr). The most frequent dentofacial deformities were mandibular deficiency (30.9%), open bite (27.0%), and maxillary retrusion (23.0%). Sixty-five patients had been diagnosed with AS before surgery. In 46 of these patients (71%), signs of AS after surgery had disappeared. However, of the 191 patients who did not have AS before surgery, 25 developed AS after treatment (Table 1). Based on these data, developing AS after treatment was less frequent than the disappearance of AS after treatment (odds ratio [OR] = 1.8, *P* = .02).

Categorizing patients by their AS status before and after treatment yielded 4 groups: no AS before and after treatment, AS before but not after treatment, AS after but not before treatment, and AS before and after surgery. Table 2 presents patients' characteristics in

**Table 1. AS STATUS BEFORE AND 12 MONTHS AFTER TREATMENT**

	AS After Treatment	No AS After Treatment	Total
AS before treatment	19	46	65
No AS before treatment	25	166	191
Total	44	212	256

*Note:* The risk of AS disappearing after surgery was higher than the risk of developing AS after treatment in patients without AS before treatment (odds ratio = 1.8, *P* = .02 by McNemar test).

Abbreviation: AS, atypical swallowing.

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each group. Most patients ( $n = 186$ ) did not have AS before or after treatment, whereas only 19 (7.4%) of the 256 patients had AS before and after treatment. Patients with and without AS were similar in gender distribution, type and severity of dentofacial deformity, and surgical procedure. However, patients without AS before and after surgery were older. Relapse (primarily the reappearance of open bite after surgery) was more frequent in those with a positive AS status, with patients without AS presenting the lowest rates of relapse and patients with AS before and after treatment having the highest rates of relapse (Fig 2).

Of the 256 patients investigated, only 23 (9%) showed relapse at 1 year after treatment. Univariate analysis showed that baseline predictors of relapse were AS status (before treatment,  $P = .002$ ; after treatment,  $P = .001$ ; before and after treatment,  $P < .001$ ), type of dentofacial deformity, and type of surgical procedure. Specifically, patients with open bite had a greater risk of relapse than patients with other deformities ( $P < .001$ ). Furthermore, double-jaw surgery (Le Fort I and BSSO) increased the risk of relapse ( $P = .02$ ) compared with only BSSO or Le Fort I (Table 3).

A multivariate analysis was performed to assess whether AS was associated with relapse. This analysis was performed after adjusting for gender, dentofacial deformity, and surgical procedure. After adjustment, AS status (before treatment, OR = 9.44,  $P = .005$ ; after treatment, OR = 56.17,  $P < .001$ ; before and after treatment, OR = 20.49,  $P < .001$ ) was shown to be significantly associated with relapse. Multivariate analysis also showed that regardless of AS status, the presence

of an open bite at baseline was associated with an increased risk of relapse ( $P < .001$ ; Table 4).

## Discussion

This study sought to describe the links between AS and relapse after combined orthodontic and orthognathic treatment. In the present patients, AS was associated with post-treatment relapse. Moreover, the combined treatment, regardless of the specific procedure, could lead to the disappearance or the appearance of AS.

### PREVALENCE OF PRE- AND POST-TREATMENT AS

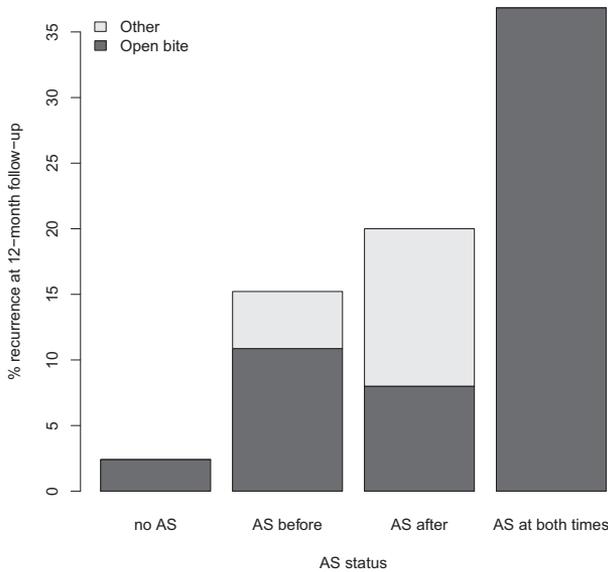
AS present before treatment persisted in approximately one third of cases and disappeared in the remaining two thirds after treatment. In contrast, almost 10% of patients developed AS after treatment, although AS was not present initially. Swallowing perturbations after orthognathic surgery have been related to the reciprocal displacement of the buccal floor musculature stretched between the mandible and hyoid bone<sup>14</sup>; however, these changes are normally transient and would be corrected 3 to 6 months postoperatively.<sup>15</sup> The interpretation of these data can be put in perspective with the hypothesis of MacAvoy et al,<sup>11</sup> that the swallowing patterns of an individual is under central control and can be influenced by peripheral changes induced by orthognathic surgery. The causal links between an excessive lingual volume and the development of a malocclusion have long been recognized, and this could justify the

**Table 2. BASELINE SAMPLE CHARACTERISTICS BY AS GROUP**

	AS 00 (n = 166)	AS 10 (n = 46)	AS 01 (n = 25)	AS 11 (n = 19)	P Value
Gender					.67
Women	89 (53.6%)	26 (57.8%)	16 (64.0%)	9 (47.4%)	
Men	77 (46.4%)	19 (42.2%)	9 (36.0%)	10 (52.6%)	
Age (yr)	26.1 (10.6)	21.8 (6.5)	21.8 (6.6)	21.1 (6.5)	.005
Dentofacial deformity					.11
Facial asymmetry	10 (6.0%)	4 (8.7%)	2 (8.0%)	1 (5.3%)	
Mandibular deficiency	63 (38.0%)	7 (15.2%)	6 (24.0%)	3 (15.8%)	
Mandibular excess	17 (10.2%)	8 (17.4%)	5 (20.0%)	2 (10.5%)	
Maxillary retrusion	38 (22.9%)	12 (26.1%)	6 (24.0%)	3 (15.8%)	
Open bite	38 (22.9%)	15 (32.6%)	6 (24.0%)	10 (52.6%)	
Surgical procedure					.11
BSSO	50 (30.1%)	8 (17.4%)	3 (12.0%)	4 (21.1%)	
Le Fort I osteotomy	33 (19.9%)	8 (17.4%)	9 (36.0%)	2 (10.5%)	
Le Fort I + BSSO	83 (50.0%)	30 (65.2%)	13 (52.0%)	13 (68.4%)	
Open bite after surgery	4 (2.4%)	7 (15.2%)	5 (20.0%)	7 (36.8%)	<.0001

Note: For AS group, 0 denotes the absence of AS and 1 denotes the presence of AS, such that group AS 10 is composed of patients who presented with AS before treatment that disappeared after treatment.

Abbreviations: AS, atypical swallowing; BSSO, bilateral sagittal split osteotomy.



**FIGURE 2.** Proportion of relapse at 12-month follow-up according to AS status. AS, atypical swallowing.

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undertaking of a glossectomy before orthognathic surgery to avoid treatment failure or relapse linked to excessive force of the tongue position on the teeth and their bony bases.<sup>16</sup> Similarly, the relation between orthognathic surgery and AS can be explained by the impact of maxillary or mandibular movements on the volume of the oral cavity, thus affecting intraoral proprioception, especially of the tongue (the oral cavity is sometimes described as a “tongue box”).<sup>17</sup> Orthognathic surgery also alters the physiology of masticatory muscles. Takeda and Saitoh<sup>18</sup> established a direct link between the contraction of the masseter muscles and swallowing. Thus, AS can appear as a possible consequence of the static changes in the tongue box and the dynamic changes in masticatory function induced by orthognathic surgery. In the present 256-patient sample, orthognathic surgery was nearly twice as likely to make AS disappear than to appear (OR = 1.8).

None of the patients perceived the AS or complained of it. Nevertheless, swallowing therapy was proposed to all patients with AS, but surprisingly none of them accepted this proposition, preoperatively or postoperatively, even in relapsed cases.

#### AS, GENDER, AND AGE

In the present sample, AS did not present any statistical association with gender. In contrast, patients without AS were notably older than those with AS. This could indicate that patients in this sample who had AS at the outset also had a malocclusion severe

enough to justify orthodontic and surgical management at a younger age. Ovsenik<sup>19</sup> reported the link between AS and the early development of crossbite, and Hebling et al<sup>20</sup> established a link between AS and open bite, with AS being a major risk indicator to the development of an open bite before 5 years of age (OR = 7.4), although causality here is unclear because AS could be an adaptation of the swallowing pattern to the presence of an open bite.

#### AS AND DENTOFACIAL DEFORMITY

Patients treated for open bite in this sample were at greater risk of relapse than patients treated for other types of deformity, thus corroborating the literature that open bite is an “unstable” malocclusion whose development is often related to an abnormal positioning of the tongue.<sup>21-23</sup> However, according to the present results, AS appears to be associated with relapse independent of the type of deformity (including open bite). Looking at the results in more detail, the association of open bite with AS might potentiate the effects of AS and open bite when considered separately. In other words, the relative risk of relapse in the combined presence of AS and open bite seems higher than the sum of the relative risks of AS and open bite, despite this result not reaching statistical significance ( $P = .07$ ). This needs

**Table 3. UNIVARIATE LOGISTIC REGRESSION ANALYSES OF OPEN BITE RELAPSE ACCORDING TO AS GROUP**

	Univariate Model		
	OR	95% CI	P Value
<b>AS status</b>			
No AS	—		
AS before treatment	7.27	2.09-28.92	.002
AS after treatment	10.13	2.49-43.95	.001
AS before and after treatment	23.62	6.28-101.63	<.001
<b>Gender</b>			
Women	—		
Men	1.64	0.69-3.99	.26
Age	0.94	0.87-1.00	.07
<b>Dentofacial deformity</b>			
No open bite	0.08	0.02-0.21	<.001
<b>Surgical procedure</b>			
BSSO	—		
Le Fort I osteotomy	1.00	0.88-1.07	.99
Le Fort I + BSSO	5.61	1.58-35.76	.02

Abbreviations: AS, atypical swallowing; BSSO, bilateral sagittal split osteotomy; CI, confidence interval; OR, odds ratio.

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**Table 4. MULTIVARIATE LOGISTIC REGRESSION ANALYSIS OF OVERBITE RELAPSE ACCORDING TO AS GROUP, ADJUSTED FOR DENTOFACIAL DEFORMITY (OPEN BITE VS OTHER) AND SURGICAL PROCEDURE**

	Multivariate Model		
	OR	95% CI	P Value
<b>AS status</b>			
No AS	—	—	—
AS before treatment	9.44	2.11-52.12	.005
AS after treatment	56.17	7.49-622.6	<.001
AS before and after treatment	20.49	3.93-138.1	<.001
No open bite	0.04	0.01-0.15	<.001
<b>Surgical procedure</b>			
BSSO	—	—	—
Le Fort I osteotomy	0.98	0.73-1.29	.99
Le Fort I + BSSO	1.68	0.30-1.38	.58

Abbreviations: AS, atypical swallowing; BSSO, bilateral sagittal split osteotomy; CI, confidence interval; OR, odds ratio.

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to be confirmed in a larger sample. Although 17.2% of the overall patient sample still had AS after treatment, this percentage was 23.2% in patients treated for open bite. The percentage of relapse in these patients was statistically larger than in patients without AS. Overall, AS status and open bite at baseline showed a strong association with relapse. These associations became even stronger when adjusting for type of surgery.

There are 2 main limitations to this study. The primary weakness is inherent in the retrospective design (ie, limited control over the data gathered over an 18-yr period and the involvement of many different surgeons in the treatment of these patients). However, these data were collected during normal clinical examinations, and the physicians at that time were blinded to the goal of the study. In addition, baseline AS was assessed before relapse and thus would not be influenced by knowledge of the outcome. Given these factors, a cautious interpretation of AS as a cause of relapse is appropriate. A second limitation concerns the detection of AS, which should be interpreted with caution given the degree of subjectivity in how it is assessed. Another criticism that can be raised about this study design is that the authors did not consider the possible association of the amount of skeletal movements with relapse and AS. Although a possible deficiency, the main objective was to determine whether clinically diagnosed AS was associated with relapse in orthognathic patients.

Despite these limitations, this is the only large study that has attempted to assess the association of AS with relapse in orthognathic patients.

Based on the present results, the following should be considered for patients undergoing combined orthodontic and orthognathic treatment. 1) AS was associated with relapse after orthognathic surgery regardless of dentofacial deformity or surgical procedure performed. 2) AS can appear after treatment in patients who were initially free of it. These results justify 1) the systematic screening of AS before orthognathic surgery but also during postoperative follow-up; 2) the provision of information to patients with AS on the increased risk of relapse, and possibly 3) the systematic proposal to these patients that they undergo speech therapy as part of their postoperative rehabilitation should AS remain after the surgery. Individualized swallowing therapy techniques can be proposed to promote the upward and backward repositioning of the tongue, the approximation of the upper and lower molars, the de-emphasis on use of the lower lip, and greater emphasis on the use of the upper lip during mastication and swallowing. A remedial program involving phonetic placement also could be helpful in frontal lisp cases to correctly produce /s/ and /z/ sounds.<sup>8</sup>

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