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HEAD AND NECK

Transoral robotic tongue base reduction and supraglottoplasty combined with maxillomandibular advancement: a new option for selected sleep apnea patients? Preliminary report

Iacopo Dallan[1] · Lodovica Cristofani-Mencacci[1] · Veronica Seccia[1] · Christina Cambi[1] · Giacomo Fiacchini[1] · Stefano Berrettini[1] · Bruno Brevi[2]

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Abstract

Purpose Transoral robotic surgery (TORS) and maxillo-mandibular advancement (MMA) are effective options for obstructive sleep apnea patients. Identification of the correct candidate is by far the most important item in achieving a succesful outcome. As a consequence, not all patients can be managed successfully via one or the other procedure. To overcome the limits of any single procedure we have combined, in a very selected population of patients, TORS tongue base reduction and MMA. Preliminary data are encouraging, in terms of both AHI and ESS.

Methods A retrospective cohort study was conducted on five patients treated with combined TORS-MMA surgery. Demographic and clinical data, pre-operative and post-operative PSG and ESS were collected.

Results Three of five patients were recruited. All patients presented severe OSAHS. Mean AHI and ESS went respectively from 48 and 12 pre-operatively to 19 and 4 post-operatively. Minor bleeding occurred in two patients. No significant sequelae have been reported.

Conclusions Combined TORS and MMA is feasible and safe. Our very preliminary data are encouraging, in terms of both AHI and ESS. Long-term follow-up and a larger amount of subjects are needed to confirm this surgical approach as a valuable option for selected OSAHS patient.

Keywords Obstructive sleep apnea syndrome · Robotic surgical procedure · Mandibular advancement · TORS · OSAHS

Introduction

Transoral robotic-assisted tongue base reduction (TORS TBR) for obstructive sleep apnea hypopnea syndrome (OSAHS) patients has been reported in 2010 [1] as a theoretical "evolution" of the Chabolle's technique [2] and it has been based on the experience in tongue base cancer management [3].

Since its introduction, the technique has gained worldwide popularity. Nowadays the number of patients treated with this approach is relevant (in 2016 more than 1200 cases were estimated [4] and a multi-institutional paper has already been published [5]).

Obviously clinical and polygraphic outcomes are not always optimal and management of TORS failures is a complicated and intriguing item. The improvement of selection criteria is probably the best way to reduce failures and increase success rates. Identification of predictive parameters is another critical point that has to be addressed in the future.

On the other hand, maxillo-mandibular advancement (MMA) has been historically considered the best surgical option in case of sleep apnea patients [6]. Data from literature report a high percentage of success with limited complications [7]. Furthermore, the procedure has been proved to be effective also in the long period. Anyway, as before,

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✉ Lodovica Cristofani-Mencacci
lodovicacristofani@yahoo.it

[1] Otorhinolaryngology Unit, Azienda Ospedaliero-Universitaria Pisana, Via Paradisa 2, Pisa, Italy

[2] Maxillo-Facial Unit, Azienda Ospedaliero-Universitaria Pisana, Pisa, Italy

not all cases are successful and not all patients are perfect candidates for this kind of treatment.

Based on these grounds, the understanding of the perfect patient's profile is the holy grail of any given procedure. There are patients in which soft-tissue surgery (the typical ENT surgery) or skeletal framework surgery alone do not seem to offer good chances of success. The management of these "grey-zone" patients represents a real challenge for the physicians, considering that most of these patients are young and do not tolerate CPAP.

With this in mind, we have started treating such complex cases combining TORS with MMA in the same setting. This paper focuses on our very preliminary experience on this topic outlining our preliminary feelings about pros and shortcomings.

Materials and methods

In this retrospective cohort study, five adult patients were treated with combined TORS and MMA for severe OSAHS at the University Hospital of Pisa between 2017 and May 2019 and were screened for study eligibility.

The diagnosis of OSAHS was made according to the American Academy of Sleep Medicine (AASM) criteria [8]. Demographic and clinical data were collected with special regards to Body Mass Index (BMI),symptoms related to the risk of OSAHS; previous conservative and/or surgical treatments. Pre- and post-operative polysomnography (PSG) and Epworth Sleepiness Scale (ESS) questionnaire were collected.

All patients underwent pre-operative drug induced sleep endoscopy (DISE) and radiological evaluation with cephalometric teleradiograph and dental radiograph. They were all submitted to a pre-operative multidisciplinary evaluation, and adequately counselled about their condition, at the presence of the ENT, the oral and maxillofacial surgeon and a dedicated anesthesiologist. All patients were provided with informed written consent for the procedure. Patients were selected for a combined procedure when the senior surgeons (BB-MXF and ID-ENT) felt not confident to offer an adequate chance of success to these patients with their own stand-alone procedure. Patients were considered eligible for a combined procedure when tongue base hypertrophy was associated with an evident lateral wall collapse during DISE; when MMA patients presented an associated significant tongue base hypertrophy or when expected bony advancement in MMA was considered inadequate by the senior MXF surgeon.

Hospitalization days, time from surgery to oral feeding, complications' rate were all collected and analyzed. No statistical analyses were performed due to the low number of patients. This retrospective analysis has been approved by the local ethical committee of the Azienda Ospedaliero-Universitaria Pisana.

Results

Five patients, affected by OSAHS, were treated with a combined TORS and MMA procedure. The TORS procedure consisted in tongue base reduction (TBR) and supraglottoplasty (SGP). Only three patients were considered eligible for the study: one patient was excluded because of the TORS procedure limited to the supraglottoplasty. Another patient was excluded for the very short follow-up.

Our study cohort was formed by male patients with a mean age of 41 ± 6 years. All patients were affected by severe OSAHS with a mean Apnea–Hypopnea Index (AHI) of 48 ± 7.2. All patients already tried a positive airway pressure medical approach, eventually associated with a mandibular advancement device, but were either intolerant or refused the treatment. Mean pre-operative BMI and ESS were, respectively, 29 and 12. Mean post-operative BMI and ESS were, respectively, 28 and 4. Post-operative AHI obtained 6 months after the surgical procedure was 19.7 ± 5.4 (Table 1).

All but one patients underwent a prophylactic tracheotomy, that was removed after a mean period of 19 ± 1 days. A nasogastric feeding tube was placed during the surgery in all patients and removed after 12 ± 5 days. Two cases of minor post-op bleeding from the tongue base region occurred and

Table 1 Clinical details of our OSAHS patients cohort

Patient number	Sex, age (years)	Pre-op AHI	Pre-op CPAP	Pre-op MAD	Pre-op ESS	Pre-op BMI	Post-op AHI	Post-op ESS	Post-op BMI
1	M, 47	40	Yes	No	17	31	14	9	28.2
2	M, 39	50	Yes	No	14	31.25	24.9	2	33.7
3	M, 35	54	Yes	Yes	7	25	20.4	1	21.8

M male, *Pre-op AHI* pre-operative Apnea–Hypopnea Index, *Pre-op CPAP* pre-operative use of continuous positive airway pressure, *Pre-op MAD* pre-operative use of mandibular advancement device, *Pre-op ESS* pre-operative Epworth Sleepiness Scale, *Pre-op BMI* pre-operative Body Mass Index, *Post-op AHI* post-operative Apnea–Hypopnea Index, *Post-op ESS* post-operative Epworth Sleepiness Scale, *Post-op BMI* post-operative Body Mass Index

Table 2 Perioperative details, complications and sequalae in our cohort

Patient number	Prophylactic tracheotomy	Time of decannulation (days)	NGFT placement	NGFT removal (days after surgery)	Post-operative bleeding, days after surgery	Long-term sequelae
1	Yes	18	Yes	12	Yes, 2	No
2	Yes	19	Yes	6	No	Inferior lip hypoaesthesia
3	Yes	20	Yes	16	Yes, 7	No

NGFT naso-gastric feeding tube

were managed transorally in the O.R, respectively ,at day 2 and day 7 post-op. Mild inferior lip hypoaesthesia occurred in one patient as long-term consequence. No significant sequelae have been reported (Table 2).

Discussion

The oropharyngeal area, in OSAHS patients, should be considered as a complex anatomo-functional entity, named subglosso-supraglottic region. Histology and geometry of the enlarged tongue base differ widely among different patients [9]. In terms of anatomy the lateral walls of the upper airway are mainly formed by constrictor muscles and, to a lesser extent, by extrinsic muscles of the tongue and muscles of the soft palate. One of the major tenets of upper airway surgery is to understand the concept of skeletal framework and the soft tissue within [10]. TORS TBR is today included in the surgical armamentarium for the treatment of sleep disordered breathing in a great number of ENT departments. Data from literature show good outcomes and limited complications [5, 11]. On the other hand, it is well clear that obstructive sleep apnea is not only influenced by anatomy, but also by other functional phenomena that could play a fundamental role in its genesis [12]. It is not surprising that not all patients are treated successfully via TORS TBR and patient selection represents the most critical factor in predicting post-op success. The ideal TORS candidate is a patient with BMI < 25 and a huge and lymphatic tongue base. Based on actual knowledge and failures analysis we learnt that a

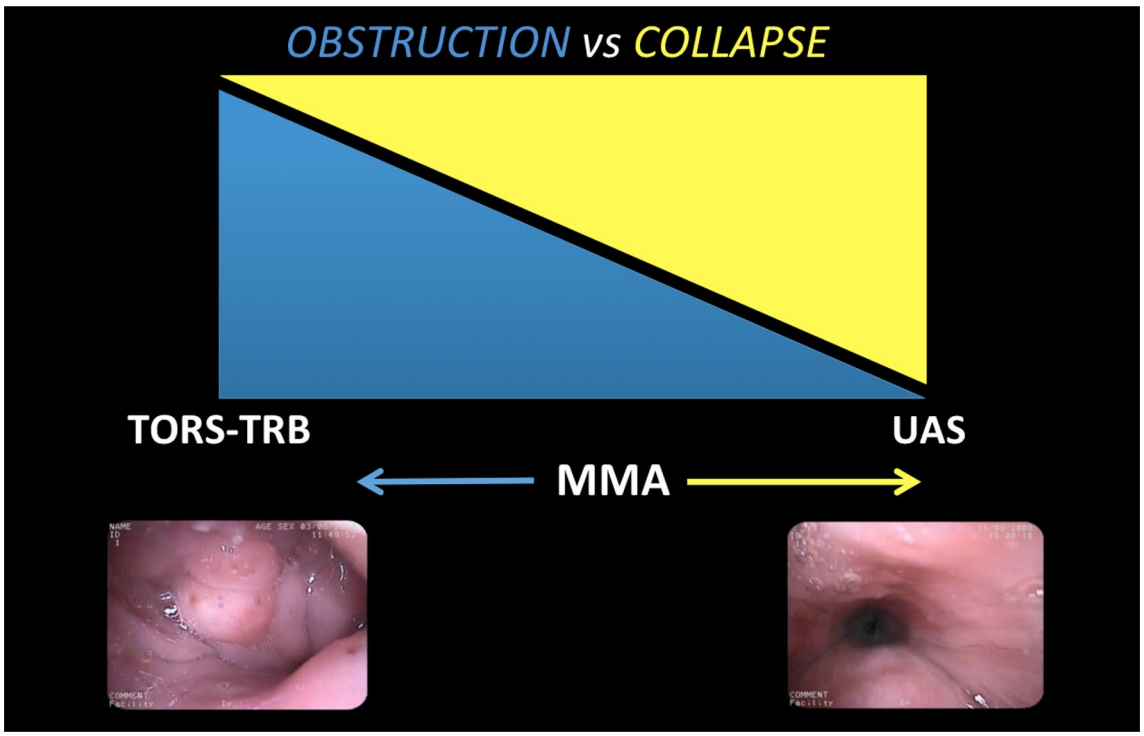

Fig. 1 Schematic drawing showing the potential role of upper airways stimulation (UAS), maxillo-mandibular advancement (MMA) and tongue base reduction via TORS (TORS-TRB) on upper airways patency. If TRB is able to manage obstruction by removing lymphatic tissue (pure "volumetric" action), the UAS is effective on the pharyngeal wall collapse. MMA is more effective on pharyngeal lateral wall collapse and tongue base position and has less impact on lymphatic tongue base obstruction

patient with a vertical tongue base with poor lymphatic tissue is not a good candidate for TORS. From a surgical point of view robotic-assisted procedures on tongue base are able to effectively remove bulky tissues, but cannot improve tongue shape or change/improve muscle "pretension". In other words, TORS TBR increases retrolingual space and nothing more. To be clear, TBR via TORS should not be considered the "robotic" transposition of the Chabolle's technique, simply because in this last procedure the lingual complex is anteriorly placed (via hyoid bone attachment to the mandible) and a reshaping of the tongue base (linguoplasty) is obtained. This explains why, in the event of significant lateral wall collapse observed during DISE, TORS has been proved as not so effective [9]. This observation can be easily explained with the lack of activity of TORS-TBR on the lateral pharyngeal wall and on the function of the genioglossus muscle. Therefore, the Achille's wheel of all TORS TBR procedures is the lack of management on both the pharyngeal lateral wall collapse and tongue base position. With this in mind it becomes clearer how pure "volumetric" patients respond well to the procedure while "muscular and vertical tongue base" or multiple-collapse-sites patients are really poor candidates (Fig. 1). On the other hand, MMA has been proved to be really an effective and stable procedure able to cope with even severe OSAHS cases, regardless of the specific DISE pattern. The mechanical aspects of MMA activity are mainly related to the stiffening of the pharyngeal lateral wall and to the "reshaping" of the tongue base. But, as for any other procedure, not all patients are good candidates for MMA and it is not difficult to understand that, in case of limited advancement, the expected outcomes are not so good. To overcome these limits and to offer a reasonable surgical option to our patients we have combined, in the same surgical procedure and, in very selected cases, TORS TBR-SGP and MMA. Our preliminary data are absolutely encouraging, since the procedure was successful in two out of three patients (defined as a decrease in the AHI by 50% of baseline and AHI < 20) (Fig. 2). One patient has been considered improved (improvement defined as > 50% reduction in pre-op AHI). It should be noted that BMI was > 30 in two out of three patients and that in one patient BMI also increased after surgery. No patient has been cured in

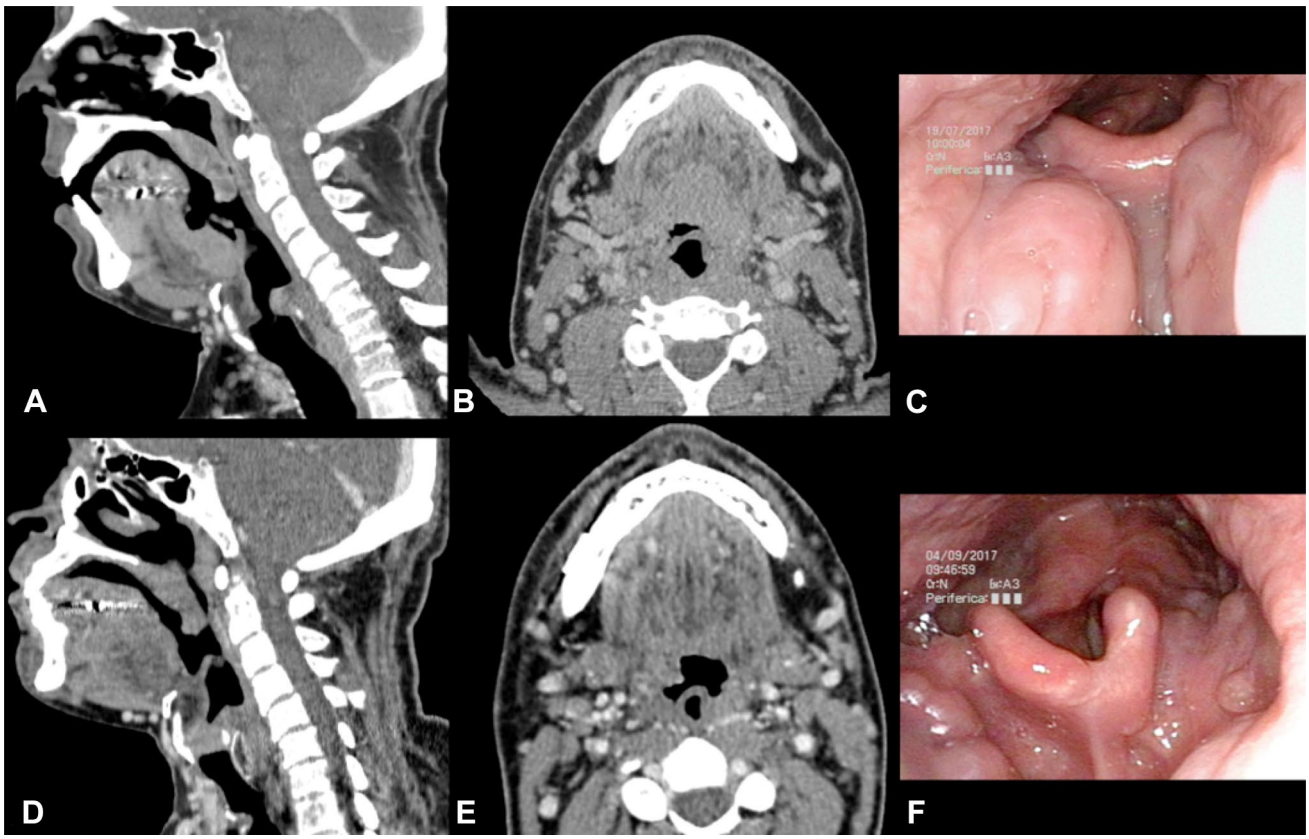

Fig. 2 Pre and post-operative CT images and endoscopic examination of one of our patients. **a**, **b**, **d**, **e** Sagittal and axial CT images show respectively the pre and post-operative radiological features of the tongue base region and lateral pharyngeal wall. In **c** and **f** the pre and post-operative endoscopic examination of the tongue base region. To notice the modification of the shape and position of the tongue base, related to removal of the exceeding lymphatic tissue by TORS, and the stiffening of the pharyngeal lateral wall and the "reshaping" of the tongue base by maxillo-mandibular advancement

our small series, but the authors did really not expect this outcome. Adequate oral intake was obtained between day 6 and 16, but we think that, with increased experience, we will run for an earlier introduction of oral feeding. Regarding complications, two out of three patients presented a minor post-op bleeding from the tongue base region that was easily managed transorally in the OR. This rate is by far higher than expected from literature's data [13] and also comparing with our overall case series of TORS (as a whole, around 12% of our cases). We do not have a specific explanation for this and it is more probably a by-chance association, since we do not feel that MMA-TORS combination could be related to this rate. Considering the virtual lack of realistic alternative and the willing of the patient against CPAP we do feel that these data are absolutely worthy to be shared. We stress the concept that our patients were not considered good candidates for a "single" procedure, in other terms, the two senior surgeons (BB and ID) did not feel confident to offer an adequate chance of success to these patients with their own stand-alone procedure. In other words, patients presented one of the following: tongue base hypertrophy associated with an evident lateral wall collapse during DISE,MMA candidates that presented an associated significant tongue base hypertrophy or expected bony advancement in MMA was considered inadequate by the senior MXF surgeon. As a consequence we think that the implementation of a combined procedure, reducing tongue base volume, stiffening lateral pharyngeal wall and remodeling tongue base shape may overcome the limits of "single surgeon" approach and offers a valuable option to very selected patients. And this concept of combining different procedures and treatments, is actually the best option to this set of patients, before we really enter in the era of precision medicine also in obstructive sleep apnea [14]. Anyway, our final aim is not to present our solution as the gold standard in these complex cases, but simply to show our preliminary experience and demonstrate the feasibility and safety of such combined procedure. Last but not least, these kinds of approaches should be performed in a tertiary referral center with adequate competence and facilities. The role of all the medical and nurse staff cannot be underestimated in managing these complex patients.

Conclusion

Combined TORS and MMA is feasible and safe. Our very preliminary experience is interesting and shows good outcomes. Complications' rate is acceptable and, although practically complex, this blended solution should be considered a valuable option for really selected OSAHS patient CPAP not tolerant.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Informed consent Informed consent was obtained from all individual participants included in the study. Additional informed consent was obtained from all individual participants for whom identifying information is included in this article.

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