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ORIGINAL ARTICLE

Trans-Oral Versus Trans-Cervical Submandibular Gland Removal in Benign Submandibular Gland Swelling Patients

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ABSTRACT

Background: The main advantages of trans-oral (TOA) removal of submandibular gland (SM) are to eliminate the potentiality of remnant duct disease since the entire duct and papillae are removed and avoid cervical scar. The aim of this work was to assess the feasibility and outcome of TOA for removal of the submandibular gland (SMG) in comparison to the standard trans-cervical (TCA) approach.

Methods: This study was applied on 18 patients with benign SMG swelling indicated for gland excision.9 patients were operated via the standard TCA and 9 patients were operated via the recent TOA. Then the patients were followed up and assessed for postoperative pain, edema, nerve deficits, scar and complication.

Results: All patients complained of intermittent pain and swelling in the submandibular area. TOA take longer operative duration than TCA.TOA led to more severe early postoperative symptoms than the TCA, including postoperative pain in all patients, salivary pooling in six cases (66.7%), temporary tongue numbness and hypoesthesia that was reported in all cases, tongue deviation on protrusion was documented in one case (11.1%), and mouth floor infection was detected in 2 cases. All these symptoms resolved after one week and did not leave permanent sequela. Non-disfiguring visible neck scar was seen in all the TCA cases without reported complication of the scar.

Conclusion: Even though, the TOA for excision of the SMG is not easy and takes a longer duration and most patients temporarily complained of neurologic problems of the lingual nerve, but these were completely resolved within 2 months after surgery.

Keywords: Submandibular Gland; TOA trans- oral removal of sub mandibular gland; TCA trans-cervical removal of sub mandibular gland; Lingual Nerve and Hypoglossal Nerve
Suckling.

INTRODUCTIONS

The submandibular gland (SMG) is situated in the SM region of the neck, lying under the platysma muscle [1,2]**.** The SMG is divided into a superficial and a deep part by the mylohyoid muscle, and to gain complete gland access, the mylohyoid must be anteriorly retracted [2] .The SM (Wharton's) duct emerges the deep part of the gland to pour anteriorly in the mouth floor, lateral to the tongue frenulum [2 **] .**The lingual nerve (LN) crosses the duct twice (anteriorly and posteriorly) on the way to provide sensory innervations to the ipsilateral tongue anterior two-thirds. The marginal mandibular nerve of the facial nerve (MMN) locates in the subplatysmal plane under the mandibular angle, supplying motor innervations to the mouth corner, and this nerve damage might cause permanent drooling from the mouth corner [2]SMG removal is indicated for numerous SMG disorders, neoplasia, sialolith or sialadenitis, and plunging ranula being the most common causes of SMG removal. SMG removal is typically done using a trans-cervical approach (TCA); which is considered a safe method, but complications like visible or complicated scar, MMN injury and injury to different nerves in the surgical field could happen**[**3,4**]** ·Trans-oral approach (TOA) was recently used to remove the SMG [1,5,6**]** that allowed the SMG removal without exterior cervical incision [2,7**].** But very restricted studies investigated this method, and it is still not popular. Thus, the aim of this work was to assess the feasibility and outcome of transoral removal(TOA) of the SMG in comparison to the standard TCA.

METHODS

A prospective study took place in Otorhinolaryngology, Head and Neck Surgery Department, Zagazig University Hospitals to compare the TOA and the TCA for SMG removal from October 2018 to October 2019. Before initiating this study, the protocol, the informed consent form, and all interventional maneuvers that would be done to patients, were reviewed and approved by the Ethics Committee of the Zagazig university hospitals (IRB). The study was done according to The Code of Ethics of the World Medical Association (Declaration of Helsinki) for studies involving humans.

The study included 18 patients who diagnosed to have benign SMG swelling indicated for submandibular gland excision. Patients with malignant SMG pathology, with previous oral surgery and unfit patients for general anesthesia were excluded from the study.

Patients were divided into two groups: Group 1 included 9 patients who were operated via the standard TCA. Group included 9 patients who were operated via the recent TOA.

A full history was taken from all patients. Complete clinical and physical examination was performed including general and local ear, nose, throat and neck examination. Radiologic investigations including SMG ultrasonography **[14]** and CT was sometimes done to assess the gland. The routine preoperative laboratory tests were also assessed. Follow up of the patients was done at the second post-operative day, third and seventh postoperative day then weekly for one month then at 6 months postoperatively with registration of the postoperative data. Criteria of comparison between the two groups included mainly: operative time, post operative pain **[11,12]** (0 means no pain, 5 means maximum pain), hospital stay, feasibility of surgery, and postoperative complications.

VAS for difficulty in swallowing was taken when soft diet started, 1week and 4 weeks postoperative (0 means no difficulty, 5 means maximum difficulty). Postoperative edema was categorized subjectively into: a- Mild (just noticeable). b- Mild to moderate (more obvious edema without occlusion of sublingual angle). c- Moderate to severe (edema partially occluding sublingual space). d- Severe (edema totally occluding sublingual space). Degree of edema was registered (photos) daily till edema subside (within the first postoperative week) then maximum registered edema grade was recorded and compared between both studied groups. Any OSA or airway obstruction was also recorded **[15]**.

Patient satisfaction score: Patient satisfaction (asking patient if he is highly satisfied, satisfied, relatively satisfied, unsatisfied) **[**16] Subjective patient satisfaction with incision scar 6 months after surgery. The subjective score was evaluated by visual analog scale ranging from 0 to 10, with higher scores meaning better patient satisfaction **[17]**.

- **TOA for SMG excision:**

Surgical procedure: The patient was positioned in the supine position and intubated transnasal for general anesthesia [2]. The method might be done with oral intubation [18], but we favored nasal intubation to have an unobstructed view of the patient's mouth cavity. The mouth gag was located in an inverted manner into the mouth cavity helping complete mouth floor visualization (Fig. 1) as made by Kauffman et al [19] and a lip retractor was utilized for the mouth floor exposure like Çukurova et al [20].The mouth cavity was sterilized with saline irrigation and aqueous iodine and we retracted the tongue to the contralateral surface of the SMG to be removed as mentioned by Lee et al. In preparing the mouth floor incision, attention was paid to leave 1 to 1.5 cm of mucosa alongside the mandibular lingual surface to permit for simple closure at the end as advised by Kauffman et al **[**19**]**. Following of 1% lidocaine with epinephrine 1: 100,000 administration into the mouth floor, to reduce mucosal bleeding, the Wharton's duct opening was cannulated to ease the duct localization within the surgical field as recommended by Brown et al. An incision was done from the Wharton's duct caruncle to the retromolar trigone and the mucosal flaps were cautiously raised surrounding the sublingual gland for the paralingual space un roofing as previously described by Lee et al. A cuff of gingival mucosa was conserved to offer a surface for tension-free wound closure and to lessen the hazard of the tongue tethering as recommended by Çukurova et al. The lingual nerve (LN) that is situated on the SMG superior-posterior-lateral surface, was carefully dissected away from the SMG and preserved as described by Çukurova et al**.** The sublingual gland was subsequently dissected and removed completely, with Wharton's duct isolation and the LN protection. Due to the crossing pattern of the LN and the duct, the duct was required to be elevated off the LN anteriorly, but posteriorly, it requests to be tunneled beneath the LN as recommended by Lee et al. After that, the duct was followed to the SMG deep lobe, and the SM ganglion was freed from the SMG to release the LN from the gland.

After tongue and mouth floor retraction, the mylohyoid muscle could be recognized, and on lateral the mylohyoid muscle retraction, we searched for the superficial lobe of the SMG, and the SMG anterior and upper surface was shown by

blunt dissection. **[2]**Digital pressure applied under the SM triangle pushes the SMG into the surgical field, a trick that was mandatory to find and deliver the superficial lobe of the gland out of the neck as recommended by Brown et al. More blunt dissection than cautery was used to free the SMG from its attachments beginning posteriorly and after that move medially, subsequently anterior and lateral dissection. More blunt dissection than cautery must be applied while freeing the SMG lateral aspect to avoid injury to the MMN as suspected by Brown et al. The gland was then gripped cautiously with long tissue forceps, bluntly dissected and pulled up throughout the incision.The facial artery and its glandular branches are evident upon blunt dissection and can often be completely freed from the SMG. To avoid severe intra operative bleeding and postoperative haematoma, the vessels to the SMG must constantly be recognized and ligated. The LN and hypoglossal nerve (HN) could be recognized in the surgical field bed. The SM ganglion was recognized and separated away from the LN [20]Bimanual palpation should be performed, before the ending of the surgery, to identify any residual gland. The surgical field was then profusely irrigated and loosely closed with interrupted vicryl sutures with no drainage leaving a small hole posteriorly for efflux of the blood**[20]Fig. 1)**.

Lateral trans-cervical approach:

About 6 centimeter incision was located in a lateral neck crease about more than 2 centimeters under the mandibular lower edge. Subplatysmal skin flap was elevated till the facial vein was recognized and ligated at the lower border of the SMG and was superiorly reflected with the fascia over the SMG. This skill exposes the SMG to guarantee the MMN protection. The facial artery was ligated or conserved by ligating only the branches of the facial artery to the gland. Blunt dissection subsequently continues towards the superiomedial gland where the mylohyoid muscle was anteriorly retracted to complete the dissection. Posterior and inferior SMG traction made recognition and differentiating of Wharton's duct, the LN with its connection to the SM ganglion, and the HN easily. The SM duct was, after that, ligated and divided next the mouth floor. Then the gland was freed from the SM ganglion and removed protecting the LN and HN. The cervical wound was closed and a rubber drain was left in wound that was removed 2 days after if no excess leak (Fig. 2).The platysma was reapproximated using absorbable sutures. The skin is closed for best cosmetic result. We normally situate a light pressure dressing or "jaw braw" too for 24 hours if no drain is utilized**[21]**.The wound was inspected carefully for hemostasis. It is wise to examine the mouth floor mucosa before emergence from anesthesia if there was considerable fibrosis or complex dissection. If a mucosal injury is recognized this ought to be primarily repaired with absorbable sutures **[21]**.

Systemic antibiotics, metronidazole and mouth wash were used in the postoperative time. We alerted patients against forceful activity or heavy carriage for 5 days to decrease the threat of hematoma formation. **[21]** Patients classically were discharged on the same day of surgery.

STATISTICAL ANALYSIS

All data were analyzed using Microsoft Excel software and Statistical Package for the Social Sciences (SPSS version 20.0, Inc., Chicago, IL, USA).Quantitative data were expressed as the mean± SD. Paired t test was used to compare between pre and post operative results of normally distributed variables. All tests were two sided. P-value ≤ 0.05 was considered statistically significant, p-value > 0.05 was considered statistically insignificant and p-value <0.001 was considered highly significant.

RESULTS

A total of eighteen cases were included in the current study; nine were operated via TCA and the other nine were scheduled and prepared for TOA. But in three patients (33.3%) of the TOA cases, (all of them had erupted last molar tooth), the superficial lobe of the gland could not be removed transorally and so we change to the TCA for complete removal of the gland.

The age ranged between 20 and 57 years with a mean of 36± 4.7 years, 10 patients (43.75%) were males and 8 patients (56.25%) were females. The age range in the transcervical group was 22- 54 years with a mean of 35 ± 4.8 years. While the age range in the transoral approach was 20- 57 years with a mean of 37 ± 4.5 years. In the transcervical group, patients were 6 (66.7%) males and 3 females (33.3%) and in the transoral group, patients were 4 males (44.4%) and 5 females (55.6%). All patients complained of intermittent pain and swelling in the submandibular area. All patients were diagnosed to have chronic inflammation of the submandibular gland that was of calcular types in 15 (83.3%) patients (8, 54.3% in the transoral cases and 7, 45.7% in the transcervical cases). While the other were non-calcular chronic inflammation of the SMG (2, in the transcervical cases and one in the transoral cases). There was no significant difference as regard type of submandibular pathology between both groups (p= 0.527) **(Table 1).**

No patients had history of submandibular abscess formation. In all cases, recovery from general anesthesia was event less. Salivary pooling was

reported in six cases (66.7%) of the TOA while was not reported in the TCA. This is mostly attributed to painful swelling of the tongue and the floor of the mouth. All discomfort and drooling resolved within a period between one and three weeks after surgery. The TOA led to more severe early symptoms than TCA, including postoperative pain, swelling sensation of the mouth floor, and difficulty in eating, during the first three postoperative days, these symptoms resolved after one week and did not leave permanent sequela. Apart from temporary tongue numbness and hypoesthesia that was reported in all cases of TOA, no other lingual nerve affection was reported. Limitation of tongue movement was reported in most cases of TOA that was resolved slowly within 3 weeks. Hypoglossal nerve affection with tongue deviation on protrusion was documented in one case of the TOA (11.1%) and was resolved within three months post operatively. While hypoglossal nerve affection was not detected in the TCA cases. No marginal mandibular nerve affection was detected in the TCA cases but it was reported temporary in the TOA in one case (11.1%) that was completely cured within three months **(Table 2)**.

No postoperative teeth affection and/or loss, temporomandibular joint affection, trismus, bleeding, hematoma, salivary fistula, hypertrophic scar or keloid was detected in any of our cases in both groups. No recurrence of symptoms of inflammation of the SMG within the follow up period in any of our cases in both groups. Non-disfiguring visible neck scar was seen in all the TCA cases without scar complication. While certainly, no neck scar is present in the TOC cases except in cases that need to convert to TCA to remove superficial lobe that showed also non disfiguring visible neck scar. The mean operative time that was calculated from the first surgical incision to the last closure suture of the incision in TOA was 147 minutes and the mean operative time for TCA cases was 51 minutes. This difference in operative duration was found highly significant ($p<0.0001$) **(Table 3)**.

Table (1): demographic characteristics between the studied groups:

		Transcervical approach	Transoral approach	P value
Age	Range	20-57 years	22- 54 years	0.3753 NS
	Mean ± SD	37± 4.5 years	35± 4.8 years	(t=0.9119)
Gender	Female	6 (66.7%)	4 (44.4%)	0.628573
	Male	3 (33.3%)	5 (55.6%)	NS (X= 0.234
Pathology	Calcular	7 (77.8%)	8 (88.9%)	0.527 NS
	Non-calcular	2 (22.2%)	1 (11.1%)	(X= 0.4)

Table (2): Differences in complications between both approaches

Complication	Transcervical approach	Transoral approach	P value
Infection	0 (0%)	2 (22.2%)	P=1 NS X= 0.653
Hypoglossal nerve affection	0 (0%)	1 (11.1%)	
Tongue numbness	0 (0%)	9 (100%)	
Marginal mandibular nerve affection	0 (0%)	1 (11.1%)	

Table (3): Operative duration in both approaches

	Transoral approach	Transcervical approach	P value
Meanoperative time	147 ± 22 minutes	51± 14 minutes	<0.0001 (t = 10.3996)

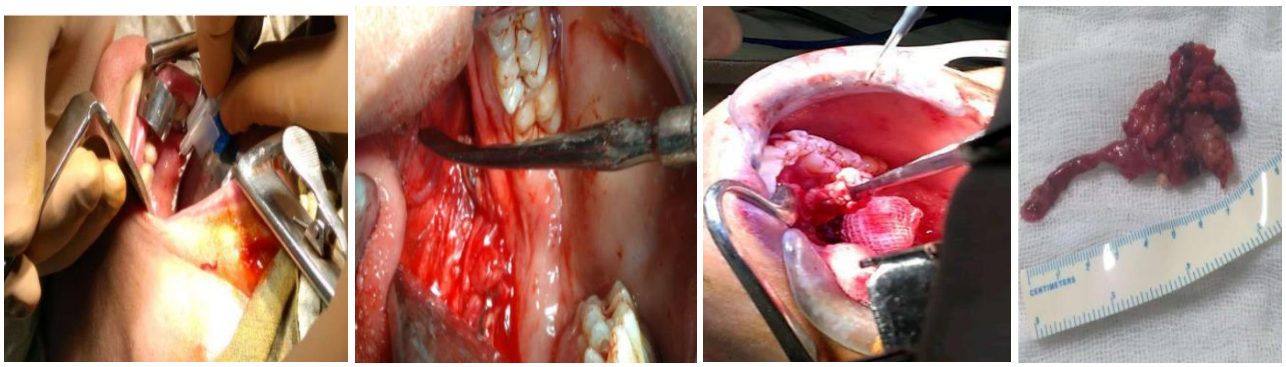

Figure (1): Wharton's duct is cannulated tofacilitate identification of the duct. lingual nerve is seen superficial to the submandibular gland. Dissection was complete for the deep lobe of the gland. The specimen including the SMG and its duct after removal.

Figure (2): One day postoperative view of the transcervical case with drain

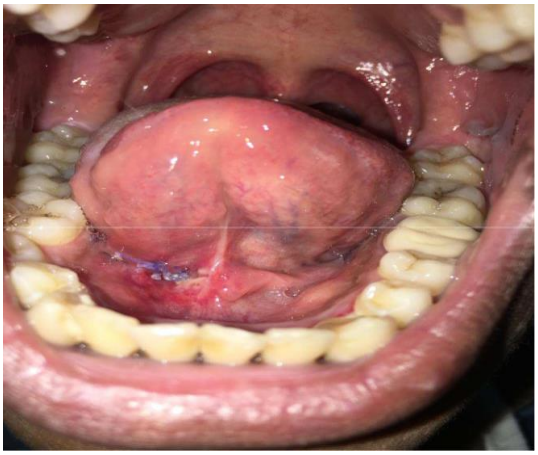

Figure (3): 12 days postoperative view of the trans-oral case**.**

DISCUSSION

The TCA for SMG removal is usually the most accepted approach **[2,23]**. Though a relatively simple method, this route causes a visible neck scar and risks the MMN injury **[**4,24**]**.In the 1960s, Downton and Qvist **[**25**]** were the first authors who recorded an TOA use to access the SMG, Downton et al.(1960) removing the SMG by shedding the mandibular periosteum and separating the mylohyoid muscle. Though, no successive reports shown in the literature till 2000, when Smith et al and Hong et al reported applying an intraoral route

to remove the SMG [.23,26] The surgeon can recognize the SMG deep lobe, by following Wharton's duct, and by the TOA, the duct can be excised totally and the hazard of possible residual duct inflammation avoided. Brown and Yao recommended the duct probing in the early stage of the operation [18], as in their belief, duct Cannulation eases its localization inside the surgical field. Once the sublingual gland has been excised entirely, the duct and LN can be seen obviously, and can be distinguished easily by color and site; the duct is light blue in color and passing posteriorly over the LN that is bright white in color. The matter of residual gland is a significant one that has to be mentioned when applying the TOA, particularly while treating benign tumor patients, for example a mixed tumor that might return in any residual gland[27,28] .In the current study, we establish that the TOA is a surgically difficult route with the major surgical challenge is the discovery and removal of the SMG superficial lobe due to the narrow and deep operative field. So, proper grasping and upward pulling of the SMG with tonsil forceps or long tissue forceps and cautious blunt dissection of the SMG capsule prevents the gland falling apart throughout the operation, even as external digital pressure upwards under the SM triangle allows the gland exposure and reduce the strength of gland traction. In adding, bimanual palpation should be regularly made during trial of removal of the superficial lobe of the gland to help to recognize any residual gland and decrease the residual gland risk. This is in agreeing with the suggestion of Guerrissi et al and Kauffman et al [19,29] subsequent to their studies. However, we encountered that even these maneuvers may not be enough to permit complete SMG removal transorally and this was met in about one third of the TOA cases in the recent study. Similarly, Weber et al and Hong et al recorded that the problem of residual gland (mostly the SMG superficial lob) is a significant one that must be mentioned when applying the TOA, particularly when treating benign tumor patients like a mixed tumor that can return in any residual gland [27,28,30].Weber et al was obliged to convert to the TCA in one of his seven cases (14.3%). In addition, in the study of Downton et al, they suggested that when the molar teeth were present, a small cervical incision was needed to assist to bring the gland into the surgical field. They similarly noticed also that all cases need to change to TCA to completely remove the gland in their cases of TOA who had erupted the last molar teeth. This in agree with Lee et al, that when the molar teeth were current, TOA roughly could not permit complete SMG excision. Thus in their study, 33.3% of the TOA patients, all had their last molar teeth erupted, TOA could not remove the SMG totally and they required to modify to TCA. Similarly, a cervical incision was required to help to bring the SMG into the operative field in the study where the molar teeth were present. [2] While Hong et al recorded that 4 of their cases has residual SMG after TOA and Çukurova et al reported Recurring sialoadenitis happened in a patient treated via the TOA. While in the study of Chang et al and Hughes et al [7,31] no operation was changed to a traditional external route but they made a few number of cases and chosen small glands. We agree with Lee et al that appropriate case selection is obligatory to make the TOA possible. So, we agree with Lee et al belief, that patients having trismus or any other circumstances that restrict exposure of mouth-opening or mouth floor, a short neck, or extreme subcutaneous fat on the SM triangle, a TCA is expected to be essential [2] Furthermore, in all TOA, surgical team ought to know and ready for change to TCA and they must have consent from patient for that after argue that with the patients. In the present study, post-operative infection (mouth floor infection) was found in 2 cases (22.2%) of the TOA cases with pus formation that require drainage in one case. Similar finding was noted by Chang et al; Weber et al and Hong et al. While Lee et al recorded no postoperative infection. Temporary tongue numbness and hypoesthesia that was reported in all cases of TOA in the current study; was also reported by Lee et al and Weber et al alongside the tongue due, possibly, to the LN contusion by retraction [2] Similarly, Chang et al reported that 88% of their patients complained of temporary postoperative tongue anesthesia, which was also cured within a week. Weber et al reported that the LN injury incidence in their TOA was considerably high (43%) and also in the study of Kauffman et al (25%). The authors recommended that the vital maneuver to lessen the risk is that the sum of LN Skeletonization should be restricted throughout the operation[28]Postoperative lower lip asymmetry was referred by Chang et al to be resulted from the platysma muscle division and they noticed it in 38% of their patients in the TCA but normally recovered during three months of surgery.

In the current study, limitation of tongue movement was reported in most cases of TOA that was resolved slowly within 3 weeks. Similar findings were reported by Hong et al and they referred the restricted tongue movement to the lateral tongue and the mouth floor swelling and were completely recovered during a few weeks[27]. As found before by Hong et al, though the TOA led to more early symptoms than TCA, comprising postoperative pain, the mouth floor swelling sensation, and dysphagia, throughout the

first three post-operative days, these symptoms recovered after one week and did not leave lasting sequela. Another possible drawback of the TOA is the more difficult recognition and ligating the SMG vessels to as the working field is comparatively narrow and the facial artery and vein are situated at the SMG posterior portion. **[**19,32**].**However, we found that bleeding was not a problem during the TOA and also removal of the deep lobe was not difficult. Similarly, Lee et al; Song et al; Kauffman et al and Hughes et al noticed no haemostatic complications in the TOA of their series. The key trick is taking care to remain the dissection plane directly on the capsule while accurately gripping and pulling the gland via long tissue forceps or tonsil forceps and using external digital pressure underneath the SM triangle aid to the gland exposure. Careful and slow dissection by this way avoids bleeding from the vessels and prevents the other structures injury **[** 2].The postoperative scars were undetectable and concealed because they were situated on the floor of the mouth in all TOA, while the scar were evident even while done in the natural neck skin crease in most TCA. Similar findings were recorded by Chang et al. In adding; no drain was required to be located in the TOA since postoperative drainage spontaneously occurred via the oral incision avoiding much care to the patient **[**19**].** No gustatory sweating symptom (Frey's syndrome) was noticed in our cases as almost all preceding studies except one case that was reported by Hong et al. No taste smell affection was reported in the current study, while Çukurova et al reported the taste of smell affection. In the current study, it was obvious, that TOA had notably longer time of operation and consequently more cost however this generally might be due to the less awareness of the anatomy from top to bottom and less experience with this novel method beside it obstacles from the small, deep and limited route. Similar long duration was reported in the previous studies **[**2,30] .We agree with Lee et al that trismus or other circumstances that restrict the mouth opening range or exposure of the mouth floor, obesity, a short neck or a big tumor size may be measured as relative contraindications of the IOA [2] .Further, in SMG malignant patients or expected malignant tumors, the TCA is essential. Sialolithiasis, small sized benign SM tumors with range less than 2 cm, ranula, and mildly SMG chronic inflammation are thought to be appropriate for the TOA. Though, the gland, which has subjected to chronic sialadenitis with scarring presents a hard surgical case as the gland is frequently stuck to the nearby tissues, and thus, gland mobilization is difficult, particularly inferolaterally as of an intraoral route [30.] .So transoral route is not favored in the moderate and severe SMG inflammation or in preceding oral surgery. But, it was established that the transoral route for the SMG removal appears to be not an easy work and it is performed through a restricted surgical field and need a long operative duration and might carry higher risk of injury to the related nerves even though it could be temporary.

So we do not recommend the TOA as a routine approach for the SMG excision but we recommend keeping this approach for patient with skin abnormalities such as known patient keloid tendency and symptomatic residual deep lobe of the gland or duct. Attempts with larger number of patients and in benign neoplastic lesions, which may have less adhesion characteristics are still required. In addition endoscopic guided transoral approach need to be evaluated.

CONCLUSION

A deep recognition of complex anatomical associations of the SMG to nearby structures is the key for efficient and safe SMG surgery. The SMG can be removed by TOA with the avoidance of an external scar but we think that it is helpful in cases with residual deep lobe or with keloid tendency and avoidance of possibility of remnant duct disease.

However, the TOA is not easy and takes a long duration and most patients temporarily complained of neurologic troubles of the lingual nerve, but these were totally resolved within 2 months postoperative.

Conflict of Interest: None.

Financial disclosures: none

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