An Unusual Cause of Laryngeal Dyspnea

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Summary: Introduction. Medialization laryngoplasty with autologous fat (MLA) is indicated in some patients with glottic insufficiency. The approach is usually safe but long-term complications are poorly described. **Case report.** We present the history of a patient who developed progressive dyspnea and dysphonia two decades after bilateral MLA, which were due to the development of laryngea lipoma into the site of fat injection.

Discussion. The potential relationship between MLA and the development of laryngeal lipoma was discussed. The lipoma may be a long-term survival of too much fat tissue, which was reorganized into a well-limited lipoma over the long-term. Another hypothesis consisted of the injection of fat tissue, including fatty stem cells, and the development of a lipoma over the year through the neovascularization process.

Conclusion. We reported the first case of lipoma developed into the laryngeal site of fat injection. Future studies are needed to explore the long-term evolution of injected fat tissue in the context of MLA.

Key Words: Autologous fat grafting-Injection laryngoplasty-Complications-Laryngeal lipoma.

INTRODUCTION

The first medialization laryngoplasty with autologous fat (MLA) was reported by Mikaelian *et al.* in 1991.¹ Since then, MLA was well-used around the world as a treatment of some vocal fold insufficiencies. Autologous fat is a biocompatible material, easy to collect and associated with low donor site morbidity. The MLA aims to improve the phonation through a better adduction of the free edge of the vocal folds.² To date, only a few papers reported complications, which may include early fat extrusion, granuloma, immediate postoperative airway obstruction, under- and overcorrection of the disorder.^{3,4} In this paper, we report a case of severe laryngeal dyspnea related to a laryngeal lipoma developed two decades after MLA.

CASE REPORT

A 40-year-old voice professional female was referred to our Laryngology Unit for progressive respiratory dyspnea. The patient had a 3-month history of severe roughness, breathiness, voice breaks and reported an inspiratory stridor. There was no swallowing disorder. The patient history reported bilateral vocal fold nodule resection 20 years ago, and the postoperative development of vocal fold scar and related glottic insufficiency, which led to the realization of a bilateral MLA. The postoperative voice quality of patient was adequate over the follow-up period (18 years). The videolaryngostroboscopical examination reported a right laryngeal mass, partly impairing the airway and the light of the

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ipsilateral hypopharyngeal cavity (Figure 1A, 1B)). The laryngeal mobility was correct. However, the vocal fold wave was impaired, and laryngologist observed a supraglottic strain behavior during the phonation of a sustained vowel /a/. The voice handicap index (VHI) was 76. The maximum phonation time (MPT) was 4 seconds. The GRBAS (grade, roughness, breathiness, asthenia, strain) assessment consisted of: G3R3B2A2S3. The biology was unremarkable. The magnetic resonance imaging showed a well-delineated right sided glottic and paraglottic lesion displacing the vocal cord. The mass was extending to the ipsilateral supraglottic space and considerably reduced the airway (Figure 1C, 1D, 1E).

Regarding the dyspnea, the senior laryngologist performed a CO_2 laser assisted resection of the laryngeal mass. The mass was removed and consisted of an indurated fatty mass.

Two experienced pathologists analyzed the lesion reporting an encapsulated well-differentiated lipoma of the paralaryngeal space. The histopathological characteristics (x20) reported a polylobulated mass with mature adipocytes, cells without atypia, mitosis, or variation in cell and nuclei size (Figure 1G). The immunohistochemistry for HMGA2 was negative, which does not exclude the diagnosis of a lipoma knowing that in 20% of cases the marker is absent. Furthermore, the immunohistochemistry for MDM2 was negative excluding the diagnosis of a well-differentiated liposarcoma.

The patient was discharged the day after the surgery. The postoperative respiratory and voice outcomes were adequate (Figure 1F), reporting a MPT of 9 seconds and a 1-month VHI at 48.

DISCUSSION

MLA is a common surgical approach for some vocal fold insufficiencies, leading to good postoperative outcomes.⁵⁻⁸ To date, a few studies interested to long-term postoperative outcome and complications of the technique. To the best of our knowledge, we reported the first case of a potential lipoma degeneration that occurred two decades after a bilateral MLA.

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FIGURE 1. Clinical, imaging, and histopathological features. The preoperative clinical features of the lipoma are reported in figures 1A and 1B. Postoperative features of vocal folds did not reveal airway obstruction (Figure C). The histopathological characteristics (HE x20) reported mature adipocyte cells without atypia, mitosis, or variation in cell and nuclei size (D). The imaging study showed in axial T1-weighted sequences (E) an homogenous hyperintense right glottic and para-glottic lesion (arrows) displacing the right vocal fold (arrowhead). The post-contrast axial T1-weighted sequence (F) displays the drop of signal intensity of the right glottic and para-glottic lesion (arrows) after fat saturation.

In practice, the injection of autologous fat may be associated with local inflammatory reaction and graft absorption.⁸ We know that less than 50% of injected tissue may be absorbed but the histological evolution of the fatty tissue architecture and organization over the long-term has never been studied. Interestingly, Anticaglia *et al.* reported a case of long-term survival of too much fat after injection into the vocal folds.⁴ Authors reported a histopathological analysis revealing a viable adipose tissue 1.5 years after the injection. As for our patient, the removal of the exceeded fat tissue of the vocal fold was associated with an improvement of the voice quality.

The occurrence of lipoma into the laryngeal site of fat injection may be explained by three hypotheses. First, the lipoma may be a long-term survival of too much fat tissue, which was reorganized into a well-limited lipoma over the long-term.² Second, the injection of fat tissue, including adipose tissue stem cells was associated with a self-renewal process and the development of a lipoma over the years, making the fat grafting procedure more than a simple soft tissue filler and rather a cell-directed therapy.^{9,10} Third, the patient had an initial undetectable laryngeal lipoma, which developed over the years, making this case a laryngeal 'incidentaloma'. Future studies are needed to explore the long-term evolution of injected fat tissue in the context of MLA.

CONCLUSION

In this paper, we reported the occurrence of a very rare cause of laryngeal dyspnea two decades after the realization of MLA. The origin of the laryngeal lipoma developed after MLA remains unclear and future studies are needed to confirm the potential transformation of injected fat tissue into lipoma.

CONFLICT OF INTEREST

Authors have no conflict of interest.

REFERENCES

- Mikaelian DO, Lowry LD, Sataloff RT. Lipoinjection for unilateral vocal cord paresis. *Laryngoscope*. 1991;101:465–468.
- 2. Shaw GY, Szewczyk MA, Searle J, et al. Autologous fat injection into the vocal folds: technical considerations and long-term follow-up.

Laryngoscope. 1997;107:177–186. https://doi.org/10.1097/00005537-199702000-00008.

- Laccourreye O, Papon JF, Kania R, et al. Intracordal injection of autologous fat in patients with unilateral laryngeal nerve paralysis: long-term results from the patient's perspective. *Laryngoscope*. 2003;113:541–545. https://doi.org/10.1097/00005537-200303000-00027.
- Anticaglia JR, Hawkshaw M, Sataloff RT. Too much fat, a rare complication of injection medialization laryngoplasty: a case report. J Voice. 2005;19:296–299. https://doi.org/10.1016/j.jvoice.2004.02.004.
- Brandenburg JH, Kirkham W, Koschkee D. Vocal cord augmentation with autogenous fat. *Laryngoscope*. 1992;102:495–500. https://doi.org/ 10.1288/00005537-199205000-00005.
- Shindo ML, Zaretsky LS, Rice DH. Autologous fat injection for unilateral vocal fold paralysis. *Ann Otol Rhinol Laryngol.* 1996;105:602– 606. https://doi.org/10.1177/000348949610500803.
- McCulloch TM, Andrews BT, Hoffman HT, et al. Long-term followup of fat injection laryngoplasty for unilateral vocal cord paralysis. *Laryngoscope*. 2002;112(7 Pt 1):1235–1238. https://doi.org/10.1097/ 00005537-200207000-00017.
- Pagano R, Morsomme D, Camby S, et al. Long-term results of 18 fat injections in unilateral vocal fold paralysis. J Voice. 2017;31. https:// doi.org/10.1016/j.jvoice.2016.10.020.9. 505.e1-505.e9.
- **9.** Eto H, Kato H, Suga H, et al. The fate of adipocytes after nonvascularized fat grafting: evidence of early death and replacement of adipocytes. *Plast Reconstr Surg.* 2012;129:1081–1092.
- Kamat P, Frueh FS, McLuckie M, et al. Adipose tissue and the vascularization of biomaterials: stem cells, microvascular fragments and nanofat—a review. *Cytotherapy*. 2020.