

Concomitant bilateral vocal cord cysts in a 12-year-old patient treated by one-stage mini-microflap CO₂ laser microsurgery: case report and review of the literature

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Abstract

Introduction: Dysphonia in the pediatric population has effects on school, family and social activity. Among the benign pathologies causing dysphonia in children, concomitant bilateral vocal cord cysts are quite uncommon. Vocal cord cysts can be congenital or acquired. The diagnosis is not always straight, but when defined the only treatment is surgery.

Objective: The authors reported a pediatric case of chronic dysphonia due to bilateral vocal cord retention cysts and discuss the etiopathogenetic hypothesis and surgical treatment with the CO₂ laser mini-microflap technique. A review of the literature was also performed.

Case report: A 12-year-old patient was admitted at the Unit of Otorhinolaryngology, Department of Surgery, Azienda Ospedaliero-Universitaria di Cagliari, University of Cagliari, Italy for chronic dysphonia lasting from one year: Grade Roughness Breathiness Asthenia Strain scale was 3 in every specific area, and Pediatric Voice Handicap Index score was 48. Laryngoscopy showed a bilateral vocal cord swelling at the middle third of both vocal cords suspect for cystic lesions. The patient underwent microlaryngoscopy under general anesthetic and CO₂ laser microsurgical excision with the mini-microflap technique. The procedure was performed with the aid of an UltraPulse® Duo (Lumenis®, Tel Aviv, Israel) CO₂ laser (10 W, CW, UP, AB 2 mm and 1 mm) with an AcuBlade™ focusing system by Lumenis® (Tel Aviv, Israel). Histology confirmed the diagnosis of bilateral vocal fold epidermoid cyst. One month after surgery, indirect

laryngoscopy showed complete healing of both vocal cords with an adequate vibratory pattern. Dysphonia recovered completely.

Conclusion: CO₂ laser microsurgery can be safely and effectively performed even in the pediatric population.

Keywords

Microflap, glottic, cyst, laser, pediatric, phonosurgery.

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Introduction

Voice disorders in children have a remarkable impact on quality of life, affecting school, family and social life. Pediatric voice disorders may be caused by benign laryngeal lesions, such as vocal fold nodules, polyps and cysts. Among the benign laryngeal pathologies causing dysphonia in the general population, vocal cord cysts are estimated to account for 6-13% of the lesions [1] and are rarely found in children.

The vocal cord cysts may develop within the lamina propria superficialis (Reinke's space), also involving the ligament, and can be classified as epidermal or mucous retention related. The epidermal cysts are generally considered congenital due to a remnant of epithelium inside the lamina propria superficialis. Two opposing hypotheses have been pointed out: according to the first hypothesis, the vocal cords cysts are congenital due to development aberrations of the larynx occurring in the fourth and sixth branchial arches [2]; the second one supports that epidermoid cysts are acquired lesions with traumatic etiology, by analogy with the etiology of cutaneous dermoid cysts. The pressure of the secretion could promote squamous metaplasia [3]; ingrowth of squamous elements from the

free edge of the membranous vocal cord is also considered a consequence of microtrauma from vocal abuse [4, 5]. Retention cysts are generally considered acquired, caused by obstruction of a minor mucous gland, probably due to vocal abuse and misuse [6].

Vocal cord cysts are usually unilateral, rarely bilateral, being more frequently associated with a contralateral traumatic lesion [7], generally observed in adulthood. However, when they are observed in pediatric patients, physicians could face the dilemma about the appropriateness of a more conservative approach like speech therapy instead of the surgical approach.

The authors report a pediatric case of chronic dysphonia due to bilateral vocal cord epidermoid cyst and discuss the etiopathogenetic hypothesis, surgical treatment with the review of the pertinent literature.

Case report

A 12-year-old female was referred to our Unit (Unit of Otorhinolaryngology, Department of Surgery, Azienda Ospedaliero-Universitaria di Cagliari, University of Cagliari, Italy) in January 2019 with a clinical history of recurrent dysphonia worsening in the last year. Previous speech therapy was only temporarily effective and, after precise counseling with her relatives, a surgical approach was planned.

Preoperative work-up included local examination with flexible scope coupled with narrow-band imaging (NBI). Perceptual voice analysis was performed using the Grade Roughness Breathiness Asthenia Strain (GRBAS) scale. Ratings of these aspects vary from 0 (normal) to 3 (severely injured). The parameters were rated by two speech pathologists. Self-assessment of voice quality was measured using the Pediatric Voice Handicap Index (PVHI) [8]. Voice evaluation pointed out a severe grade of dysphonia: GRBAS score of 3 in every specific area, and the Maximum Phonation Time (MPT) of 3.2". According to the patient's discomfort, PVHI showed a middle perception of disability (score of 48).

According to the University of Wisconsin videostroboscopic ratings, glottic closure had hourglass pattern, and supraglottic activity, vertical level, vocal fold edge, amplitude, non-vibrating portion, phase closure, phase symmetry, regularity showed the following scores respectively: 1, 1, 5, 1, 3, 2, 4, and 1 [9].

The patient underwent examination under general anesthetic and concomitant transoral CO₂ laser phonosurgery. Larynx exposure was achieved by the Kleinsasser laser laryngoscope modified by Rudert with the Göttingen model suspension system (Storz, Tuttlingen, Germany). Image enhanced endoscopy with Image1 S™ (Storz, Tuttlingen, Germany) associated with the contact endoscopy in the form of Enhanced Contact Endoscopy (ECE) confirmed the inflammatory nature of the lesion (pattern I according to the classification of Puxeddu et al.) [10] (**Fig. 1**).

The procedure was performed with the aid of the UltraPulse® Duo (Lumenis®, Tel Aviv, Israel) CO₂ laser (10 W, CW, UP, AB 2 mm and 1 mm) with an AcuBlade™ focusing system by Lumenis® (Tel Aviv, Israel), in a safe environment and with a well-trained dedicated team according to recommendations of Ossof [11]. The laser was always used as a cutting tool and never with vaporization technique. The mucosal incision was performed approximately 2 mm lateral to the cyst to avoid being on the free edge of the vocal cord. A bilateral mucosal mini-microflap was harvested, allowing for blunt dissection of the cyst to leave intact the epithelium and avoid direct trauma of the vocal ligament (**Fig. 2**).

The cysts were enucleated en bloc with minimal removal of the Reinke's space content as confirmed by histology (**Fig. 3**).

Voice improved immediately after surgery. The patient was discharged after 24 hours from surgery.

Histology confirmed the diagnosis of bilateral epidermoid cyst: the lesions were lined by stratified epithelium, and some inflammatory reaction was found within the chorion (**Fig. 4**).

The patient underwent postoperative speech therapy showing a subjective and objective voice improvement 3 months after surgery (PVHI score was 4, and GRBAS score was 1 in every specific area).

After 3 months, postoperative laryngoscopy showed the complete healing of both vocal cords, the glottic edge appeared linear and regular, and no major scar or granulation tissue were detected (**Fig. 5**). According to the University of Wisconsin videostroboscopic ratings, the glottic closure had a complete pattern, and supraglottic activity, vertical level, vocal fold edge, amplitude, non-vibrating portion, phase closure, phase symmetry were regulars (score of 1), but the mucosal wave appeared lightly reduced (score of 2 out of 7) [9].

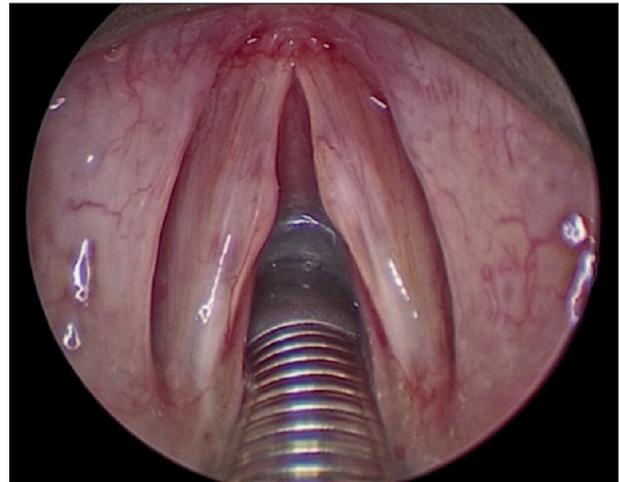


Figure 1. Intraoperative endoscopic view (Image1 S™ – Clara + Chroma) of the bilateral sub-epithelial lesion.

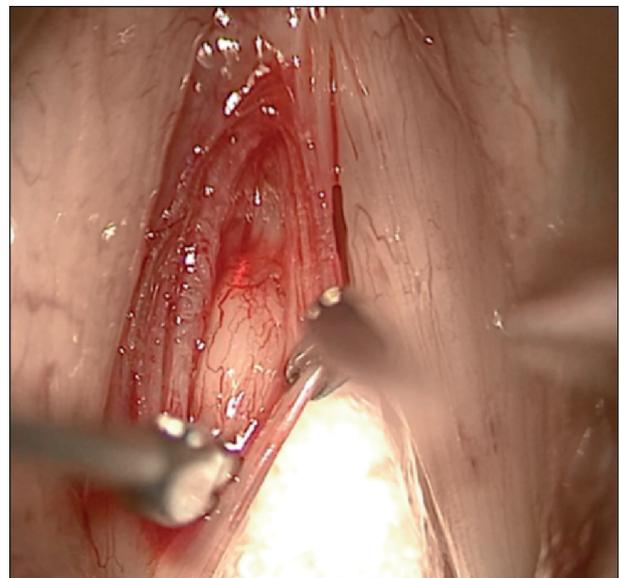


Figure 2. Intraoperative view showing the dissection of the left cyst with the aid of the laser beam.

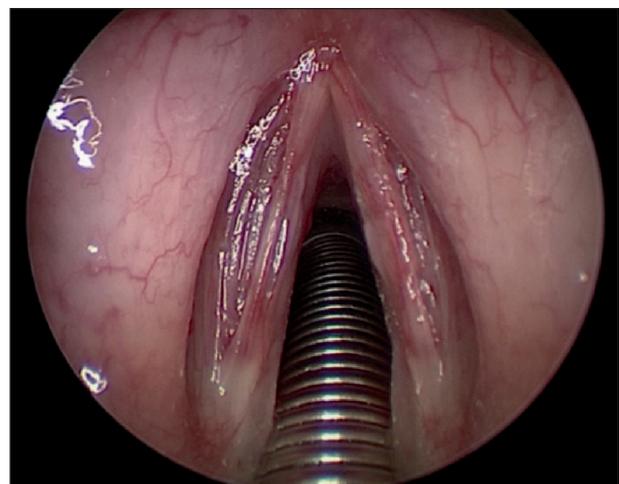


Figure 3. Endoscopic view of the glottis after the CO₂ laser mini-microflap technique.

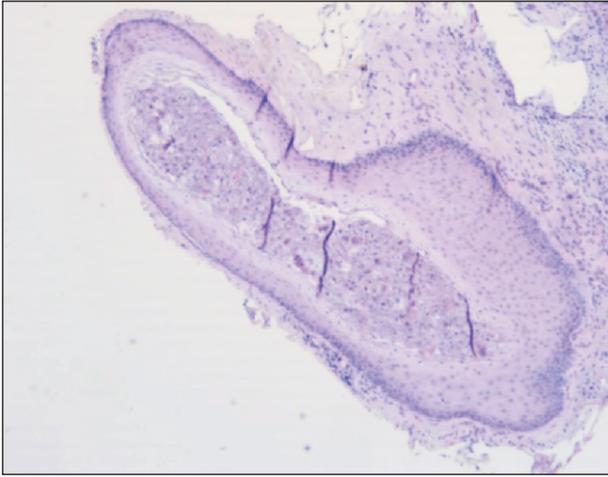


Figure 4. Histologic view (20x) of the right vocal cord cyst.

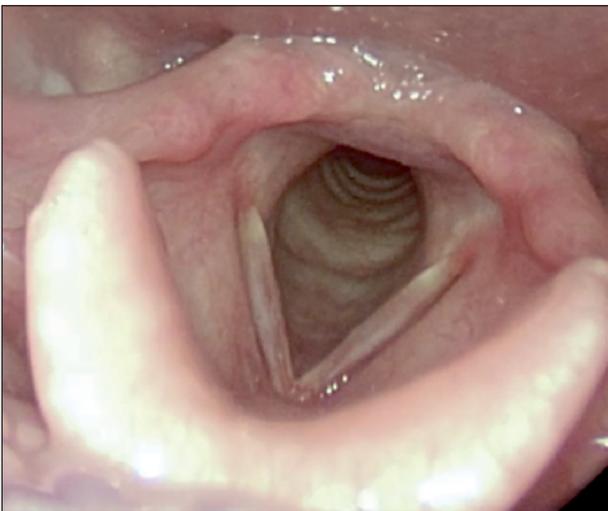


Figure 5. Outpatient endoscopic view of the glottis 3 months after surgery.

Discussion

Over 5 million children are affected by dysphonia in the United States, and school-aged children from 8 to 14 years are the most affected age group with a reported prevalence of 3.9-23.4% [12].

Although childhood is a period of anatomical and physiological changes of the larynx and voice, pediatric dysphonia can be caused by congenital and acquired anomalies. Common adult laryngeal pathology, such as vocal fold polyps or cysts, are only seldom found in children [13] and, despite the long experience of our group of research in the field of phonosurgery [14], this is the first case of bilateral vocal cord cyst that we observed in childhood. Although the genesis of epidermoid cysts is still non completely clarified, the histopathological features and the early symptomatic onset observed

in the present case lean forward the congenital theory of epidermal remnants in the lamina propria superficialis (Reinke's space) rather than the acquired one.

Clinical presentation of vocal cord cysts is generally characterized by effort to speak, vocal fatigue, recurrent hoarseness and even aphonia, due to disruption of the mucosal wave of the vocal folds and incomplete closure during phonation, as observed in our case. The degree of dysphonia in patients with a glottic cyst may be of variable degree and can change: voice worsening is usually due to the increase in volume of the cyst, but a sudden improvement in voice quality can occur after the opening/rupture of the cyst followed by the recovery of an adequate vibratory pattern of the mucosa; if laryngoscopy is performed at this stage of the disease, it can lead to a negative assessment with misdiagnosis. Therefore, a flexible scope examination should always be performed during the highest degree of dysphonia.

Preoperative diagnosis is based on the pediatric evaluation to assess the overall development of the patient. The Ear Nose Throat (ENT) evaluation should be performed by flexible HD laryngoscopy coupled with stroboscopy [15], but frequently only direct microlaryngoscopy under general anesthesia allows for the definitive diagnosis. ENT office-based instrumental evaluation can be a potentially traumatic experience for the pediatric patient, given the relatively invasive procedures required. Therefore, the referring pediatrician should prepare the child and his/her parents as much as possible: a clear explanation can do a massive difference regarding cooperation. When any organic cause has been excluded, a psychological evaluation could be performed because a functional issue can be related to psychological distress.

The optimal treatment of pediatric benign lesions of the larynx is complex and is based on several factors such as good patient and relatives' compliance for speech and language therapy, the choice and timing of surgery and postoperative voice therapy. Two options are considered for the management of benign glottic lesions: the speech therapy and the surgical approach (**Tab. 1**) [16-21].

Cohen and Garrett [16] evaluated 435 patients affected by glottic polyps and cysts (26.3% were cysts): voice improved in 60% of the patients with cysts, but only one lesion disappeared after speech therapy. Schindler et al. [17] pointed out the lower effectiveness of voice therapy compared to surgery:

Table 1. Literature research on different treatments performed in patients with a glottic cyst.

Publication	Patients	Treatment	Voice outcome	Comments
Cohen and Garrett, 2007 [16]	435 adult patients affected by glottic polyps and cysts (26.3% were cysts)	Voice therapy	Improvement in 49.1% of patients regardless of diagnosis.	60% voice improvement with cysts.
Schindler et al., 2013 [17]	65 adult and pediatric patients (range from 11 to 75 years, 22 patients with cyst)	Voice therapy	63% of the patients affected by cysts underwent laser surgery after voice therapy.	All patients attended 1-2 months of speech therapy.
Matar et al., 2010 [18]	49 adult and pediatric patients (range from 13 to 77 years) with unilateral vocal cords cysts	CO ₂ laser	Mean MPT from 11.1 to 12.7 ms; mean F0 from 310 to 434 Hz.	Stroboscopic findings, MPT and F0 improved in all patients. Net reduction of voice handicap index.
Martins et al., 2009 [19]	72 adult and pediatric patients (26 children with glottic cyst)	CO ₂ laser	Satisfying results in all cases.	-
Landa et al., 2017 [20]	39 children with congenital lesions (17 epidermoid cysts)	CO ₂ laser	Out of the 17 children with epidermoid cysts, dysphonia greatly improved in 11 cases, slightly improved in 3 cases, and did not improve in 2 cases (1 patient did not attend the follow-up).	Treatment's principles were the same as those in adults.
Bouchayer and Cornut, 1992 [21]	404 adult and pediatric patients with glottic cyst (191 with bilateral lesions)	CO ₂ laser	General voice improvement.	In the entire pediatric population, F0 was significantly higher, voice was less husky and strained.

F0: fundamental frequency; MPT: maximum phonation time.

63% of a cohort of 65 patients (range from 11 to 75 years, 22 patients with cyst) underwent surgery after completion of the speech therapy; in this study, authors observed that an improvement was seen in some perceptual parameters, but GRBAS never improved for the full group, and none of the lesions disappeared after voice therapy. Ogawa et al. [22] recently reviewed the effectiveness of voice therapy in dysphonic patients with benign mass lesions in the vocal folds; according to his study, at present there is not enough evidence to support the use of voice therapy alone for the treatment of benign vocal cords lesions, while it is strongly indicated for the treatment of functional dysphonia without any organic glottic abnormalities. In our experience, the concept of “sandwich therapy” (voice therapy – surgery – voice therapy) is applied either for adults and children.

Several papers advocate the efficacy of the microsurgical approach for the treatment of benign vocal cord lesions, either with cold surgery or CO₂ laser.

Matar et al. [18] submitted to laryngeal cold microsurgery 49 patients (range from 13 to 77 years) with unilateral vocal cords cysts: stroboscopic findings, fundamental frequency (F0), MPT and a voice handicap index improved in all patients within 1 month after surgery.

The effective role of microsurgery with cold instruments for the treatment of glottic cysts was also confirmed by Martins et al. [19] (26 children with glottic cyst) and by Landa et al. [20] (39 children with congenital lesions, 17 epidermoid

cysts); in the latter study postoperative voice outcomes were classified in 3 categories (highly improved, slightly improved, not improved), and 17 patients (65%) highly improved, 7 (17%) slightly improved, while only 4 patients (11%) did not experience any improvement.

Bouchayer and Cornut in 1992 evaluated the voice improvement in 404 patients collected in 21 years period, who underwent cold microsurgery for glottic cyst, and all patients with bilateral lesions (n = 191) underwent two different surgical procedures; in this study, the few children experienced a significant improvement of voice parameters (F0 improved of 12.8% between the first and the second assessment), and the voice timbre was significantly better, less husky and strained [21].

Surgical excision of vocal cord cysts in adults has been widely advocated in the literature; on the opposite in the pediatric population, according to Tibbets and co-workers [23], surgery can be burdened by excessive scarring of the vocal cord with a consequent likely poor vocal outcome if carried out aggressively. In our opinion, vocal cord cysts are organic disorders that cannot recover by vocal hygiene, and surgical excision is the only effective treatment even in pediatric patients.

CO₂ laser is a well-established tool for the endoscopic treatment of benign and malignant lesions of the larynx [24-27]. CO₂ laser-assisted microsurgery allows for a bloodless dissection associated with minimal thermal damage of the surrounding tissue [27], and an optimal visualization and respect of the vocal cord

anatomy such as the epithelium, the lamina propria superficialis and the vocal ligament. Maintenance of the epithelium, and in particular of the content of the Reinke's space (lamina propria superficialis), allows the recovery of the vibratory pattern of the affected vocal cord whatever surgical technique is used (laser versus cold instruments). A gentle dissection can be easily obtained by using new technologies like ultrapulsed lasers and digital AcuBlade™, like in the present case, providing minimal thermal damage since the ultrapulse mode is optimal for precise incision and ablation. The degree of thermal build-up can be minimized according to the pulse energy setting (milli-joule) for a relative amount of precision and hemostasis. The surgeon indeed can set the average power and the pulse energy, and then the laser determines the repetition rate accordingly.

The one-step or two-step microsurgery for bilateral lesions could be a controversial issue while planning surgery. In our experience, bilateral benign lesions of adults are generally managed with a one-stage microflaps or mini-microflaps technique, without any further general anesthetics [25]; in such cases, the surgeon must preserve the mucosa bilaterally as to avoid the iatrogenic webbing with subsequent disastrous outcomes. Preservation of enough content of lamina propria superficialis and all the epithelium warrants a good vibratory pattern in the long-term postoperative course.

After surgery, speech therapy could be useful to improve functional recovery [22], supporting the importance of the multidisciplinary approach in all phases of the management of pediatric patients with glottic disorders.

Conclusions

Dysphonia in children must be carefully evaluated by ENT surgeons and speech pathologists to obtain a precise diagnosis and assess the degree of the impact of the pathology on the quality of life. The present case supports the effectiveness of the one-step CO₂ laser mini-microflap technique even in the pediatric population with bilateral benign glottic cysts when performed with new generation lasers and micromanipulators.

Declaration of interest

The Authors declare that they do not have any financial interests to disclose, and they confirm no conflict of interest concerning this manuscript.

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