CASE REPORT

A Rare Case of Ductal Laryngeal Cyst and Its Surgical Outcome

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Abstract

Background: Benign lesions of vocal fold commonly occur in general population, causing significant hoarseness and interfering with routine vocal use. We aim to report a combination of microlaryngeal surgery and steroid injection intralesion in treating vocal fold ductal cyst.

Case description: A 46-year-old male presented with chronic and persistent dysphonia for the past 2 years. Flexible laryngoscope examination showed a pale white mass on the one-third anterior of the right true vocal fold. Significant improvement was acquired following microlaryngeal surgery and vocal fold steroid injection.

Conclusion: A vocal cord ductal cyst, a rare apparition, manifests clinically similar to other benign laryngeal lesions. Therefore, a common management algorithm can be utilized. Postoperative care is a notable matter in the management of a ductal laryngeal cyst to achieve good vocal outcome.

Keywords: Case report, Dysphonia, Laryngeal cyst, Microlaryngeal surgery, Steroid injection, Vocal cord cyst.

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Background

Benign lesions of vocal fold are abnormal masses of laryngeal tissue that grow excessively in an uncoordinated way.[1] Benign vocal fold lesions diminish sound production efficiency causing hoarse and breathy voice, forcing the patient to speak with more effort. Benign vocal fold lesions are classified into neoplastic and nonneoplastic lesions. The nonneoplastic vocal folds consist of vocal cord nodules, vocal cord polyps, and vocal cord cysts.[1,2] Laryngeal cysts may emerge at any mucosa-lined tissue of the larynx and contribute to 5–10% of all benign laryngeal lesions. DeSanto et al.'s categorization, widely employed, distinguishes cysts as either ductal or saccular. Ductal cysts, akin to mucus retention cysts, are situated in the mucous membrane. Saccular cysts, on the other hand, are typically larger, manifest within the anatomical plane of the laryngeal saccule, and are entirely submucosal.[2,3] Most benign laryngeal cysts are female dominant. The indications and manifestations of laryngeal cysts vary based on the lesion's size and anatomical placement. More massive lesions, especially those that are closer to the glottis, tend to exhibit severe symptoms. Pathological analysis is essential for a conclusive diagnosis of cystic lesions; imaging serves as a supplementary tool to refine the differential diagnosis and strategize treatment.[4] Conservative vocal management usually proves ineffective against vocal fold cysts. Opting for complete excision, despite its technical complexity, is the preferred treatment, and it is linked to a favorable prognosis.[4]

Case Description

A 46-year-old male presented to the ear, nose, and throat (ENT) Outpatient Clinic in Dr Sardjito Public Hospital with a complaint of chronic and persistent dysphonia for the past 2 years with no difficulty in respiration. He denied any recent history of upper respiratory tract infection, cough, swallowing difficulty, or history of stomach acid reflux. He was a regular smoker, consuming around two packs of cigarettes each day. We evaluated his voice using the Voice Handicapped Index (VHI) with a score of 34 and grade, roughness, breathiness, asthenia, and strain (GRBAS) scoring of G2-R2-B3-A2-S2. Direct flexible laryngoscope examination showed a single well-defined pearl-like, smooth, and semitranslucent mass over the anterior middle third portion of the right true vocal cord (Fig. 1). There was no phonation gap, with normal mobility of both vocal cords. The rest of the head and neck examination was within normal limits.

The patient then underwent endoscopic microlaryngeal surgery. During the operation, we found that the mass was cyst with clear jelly-like mucous filling, wrapped inside a capsule. The mass was excised completely along with its capsule without damaging the surrounding structure (Fig. 2). During the procedure, we invented thick anterior and posterior fibrotic bands tethering the cyst, the trimming of these bands assisted in complete excision of the cyst without tearing the capsule. A sharp sickle knife was used to decrease the risk of recurrence due to the micro-injury to the surrounding structure. We also injected the mucosal layer at the base of the excised mass and its adjacent structure with 1 mL of 10 mg triamcinolone.

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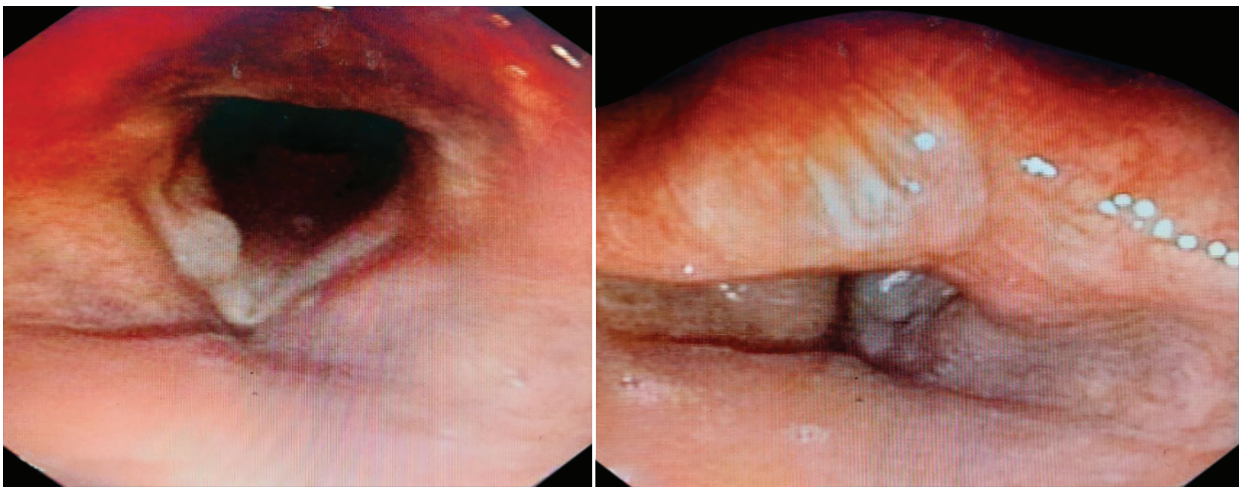
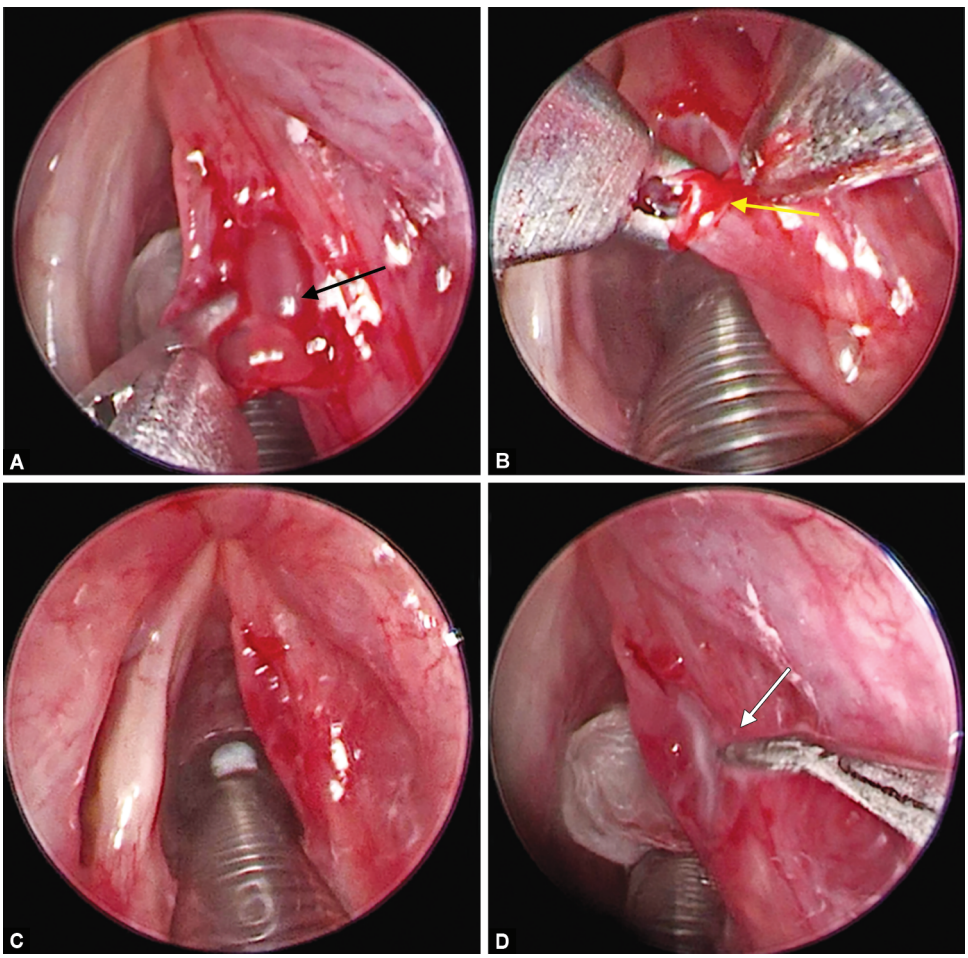


Fig. 1: Flexible laryngoscope of well-defined pearl-like, smooth, and semitranslucent mass over the middle third portion anteriorly of the right true vocal cord



Figs 2A to D: (A) A laryngeal cyst with clear jelly-like mucous filling, wrapped inside a capsule (black arrow); (B) Removal of the cyst capsule (yellow arrow); (C) Post-cyst removal of the true vocal cord; (D) Mucosal layer was injected with 10 mg of triamcinolone (white arrow)

The excised mass was sent to the pathology anatomy department for its histopathologic feature. The result showed a laryngeal vocal cord ductal cyst with typical features of a capsular structure with its wall covered with complex squamous epithelial lining and a few parts with stratified columnar epithelial lining, the surrounding mucous and fibrous layer with was edematous, with small vessel dilatation and inflammatory cell infiltration which was lymphocyte dominant, some neutrophil, and eosinophil. No sign of malignancy was observed in the specimen, suggesting true vocal cord ductal cyst (Fig. 3).

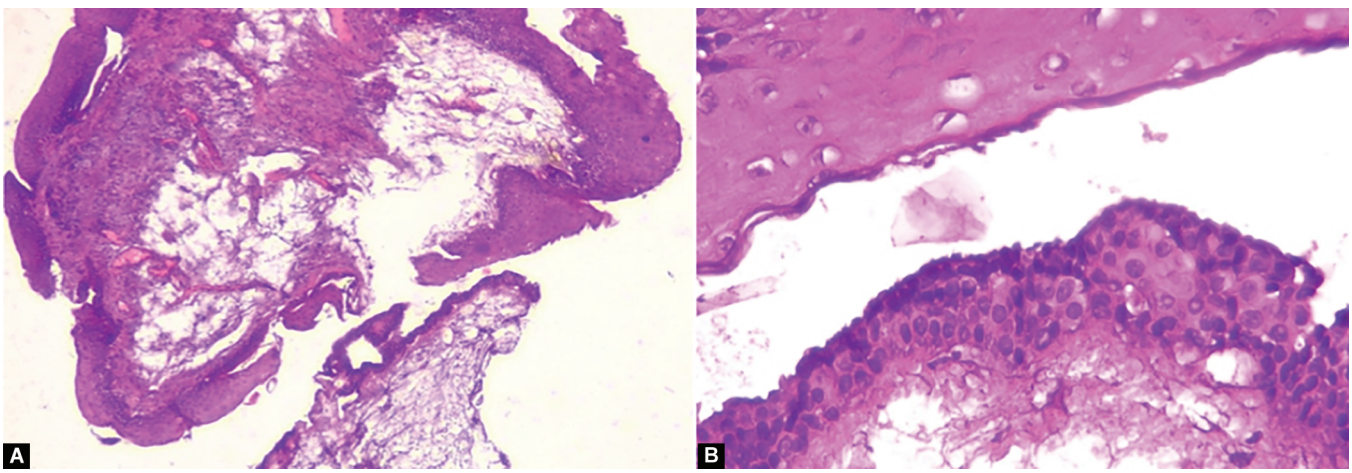
After being discharged from the hospital one day after surgery, the patient was managed with complete voice rest, antibiotic, anti-inflammation, anti-reflux, and analgesic therapy, also advised to come to the ENT Outpatient Clinic for post-surgical follow-up.

During follow-up period, the voice continued to improve. VHI score was 24 at the 1st week after surgery and 4 on the 3rd week after surgery with G2-R2-B2-A2-S2 and G1-R1-B0-A1-S1, respectively. We did flexible laryngoscope three weeks after surgery and found no remaining mass nor laceration at the true vocal cord and its surrounding structure (Fig. 4), no residual dysphonia was found in this patient. Long-term evaluation was conducted 6 months and 1 year after surgery. The patient showed great vocal outcome with VHI of 2 and G1-R1-B0-A1-S1 on both follow-ups. Flexible laryngoscope one year after surgery found no recurrent mass nor scarring tissue at the true vocal cord nor at its surrounding structure.

Discussion

Benign vocal lesions refer to noncancerous abnormal tissue growth on the vocal cords. Singer's nodules, polyps, papillomas, and cysts are typical examples of these benign vocal cord lesions.[5] Laryngeal cysts are laryngeal benign lesions that can be asymptomatic until the lesion becomes significantly big to cause voice disturbance in the form of dysphonia. Laryngeal cysts contribute to approximately 5% of all benign laryngeal lesions. They commonly occur on the epiglottis and vocal cords, presenting symptoms such as dysphonia, dysphagia, a foreign body perception, and difficulty breathing.[5,6] De Santo categorized these cysts into two types—ductal cysts and saccular cysts, categorizing them according to their locations and underlying pathophysiology. Ductal cysts result from the blockade of mucous gland ducts, and these cysts, fulfilled with mucus, have a double-layered epithelium consisting of columnar and cuboid epithelium. They typically originate from true vocal cords but are rarely found in the free margin due to the limited presence of these glands.[5,6]

Disorders affecting the voice can result in communication challenges, leading to psychosocial issues and a diminished quality of life. VHI was developed to assess the severity of the symptoms. Voice-specific outcome measures based on patient experiences have the potential to offer more insight than biological and physiological variables linked to voice production, functional capabilities, and quality of life.[6] Quality of voice assessments are frequently built based on the perception of five properties—grade, roughness, breathiness, asthenia, and strain which are mentioned



Figs 3A and B: Histological feature of the specimen taken from microlarynx surgery showing features of vocal cord ductal cyst. (A) Low magnification; (B) High magnification

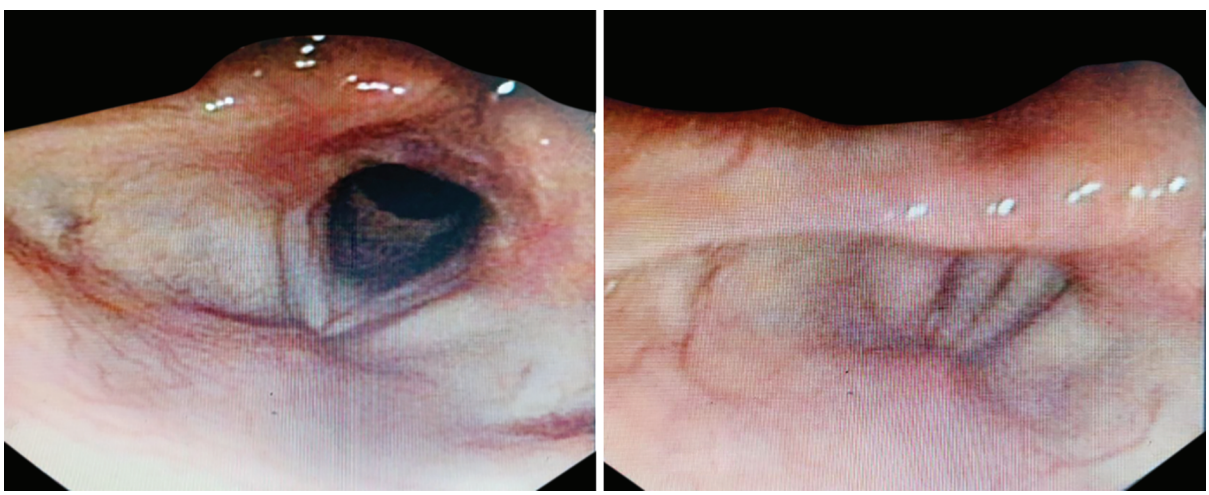


Fig. 4: Flexible laryngoscope 3 weeks after surgery

as "GRBAS." Grade (G) assesses the holistic perception of voice quality. Roughness (R) evaluates the effects of uncontrolled irregular diversities in the basic frequency and amplitude of vowel segments. Breathiness (B) gauges the volume of sound from turbulent airflow through vocal cords when partially patent. Asthenia (A) quantifies the perception of weakness or absence of energy in vocal ability. Strain (S) measures the undue effort required for speech production. Each of GRBAS properties is assessed by assigning 0, 1, 2, or 3 (0—no deviance of quality, 1—mild deviance of quality, 2—moderate deviance, and 3—severe deviance).[7]

Most individuals suffering from such cysts are smokers, but a definite causal relationship has not been confirmed due to the limited sample size. No studies point to voice abuse as the primary cause of this lesion.[6] Dysphonia serves as the primary symptom in cases of true vocal cord cysts, likely attributed to the disrupted vocal fold vibration caused by the cyst's mass effect. The intricate vibratory surface of the human vocal cord is affected by repeated injury from voice abuse or chronic inflammation resulting from factors such as smoking or gastric reflux, leading to basement membrane disruption and thickening. Vascular changes and defects lead to benign lesion growth originating within the lamina propria of the vocal cords. The mucosal wave above vocal cysts is reduced or completely absent. This occurs because the cyst anchors the mucosa, changing the biomechanical characteristics of the lamina propria and hindering the movement of the mucosal wave.[6,7]

Effective treatment involves a comprehensive approach addressing factors contributing to voice abnormalities. Medical therapy focuses on abolishing irritants and inflammatory circumstances such as allergies, reflux, and smoking while maintaining good vocal hygiene. Speech therapy aims to reduce abusive behaviors, enhance vocal efficiency, and modify vocal habits. Unlike some benign lesions such as nodules or polyps, vocal cysts typically do not respond well to conservative therapy.[7] Whereas benign vocal lesions treatment methods can be individualized based on clinical manifestations, this study contends that laryngeal microsurgery is the optimal approach for most benign laryngeal lesions.[8]

Our case used sharp dissection to excise the cyst, there was a slight rupture of the capsule during the procedure due to a thin cyst wall. According to previous study, the percentage of recurrence after blunt dissection was 30.7% and 26.08% for sharp dissection.[7,8] Laryngeal cysts are generally treated by surgical excision *via* endoscopic microlarynx surgery with or without intralaryngeal injection. Recurrence is highly rare and commonly found in cases of incomplete removal due to tissue remnant from the cyst wall which is left.[6,7] Performing surgical dissection is highly intricate, as even slight alterations in vocal cord structure may result in suboptimal voice function. Achieving complete removal with maximal preservation of the superficial lamina propria can improve vocal outcomes.[7] Most surgeons use sharp as well as blunt dissections while excising the laryngeal cysts but we prefer sharp dissection with clear identification of anterior and posterior fibrotic bands that anchor the cyst, the use of sharp instrument could avoid the rupture of the cyst and, therefore, minimizing the risk of recurrence.[7,8]

Laryngeal microsurgery stands out as a frequently employed method for excising benign structural lesions on the vocal folds.[8] Surgical methods encompass both straightforward excision and the microflap approach. Post-procedure, certain individuals might encounter prolonged dysphonia, often attributed to imperfections in the wound healing process leading to fibrosis formation. Additional factors contributing to this condition may involve excessive tissue removal, ongoing smoking, and insufficient vocal hygiene.[9] Steroid injections have been a longstanding method for addressing hypertrophic and keloid scarring on the skin. They hinder the proliferation and biosynthesis of fibroblasts while promoting fibroblast apoptosis. This is crucial since vocal fold fibrosis, often a primary contributor to persistent dysphonia following surgery can emerge from disorganized collagen fibers.[10]

Steroids have the capacity to diminish the expression of inflammatory genes and impede fibroblast migration and collagen synthesis. Consequently, this reduces the degree of fibrosis and serves as a preventive measure against scar formation. Two prevalent drugs utilized in vocal fold steroid injections, triamcinolone, and dexamethasone, possess distinct advantages and drawbacks. Triamcinolone, for instance, tends to persist longer at the injection site but carries a heightened risk of deposition. In a study conducted by Hsu et al. in 2019, it was found that the occurrence of triamcinolone precipitation was approximately 4%, then it recovered gradually over a period of 1–2 months.[11–13] Dexamethasone initially exhibits strong effects but is quickly absorbed. The research also indicated enhancements in mucosal wave, voice quality, and subjective perception of dysphonia with the use of supplementary steroid injections. This supplementary injection could be contemplated as a rescue technique, correlating with a decrease in the postoperative fibrosis rate from 11 to 3%.[11–13]

Postoperatively, it is mandatory for patients to be on strict voice rest and anti-reflux medication should be started to reduce acidic contact with the surgical site in order to promote healing. Patients should also receive speech therapy after surgery.[14,15] Optimal recovery of vocal fold mucosa and enhanced postoperative voice quality can be achieved through voice rest. Abstaining from phonation is thought to safeguard against mucosal injury, thereby preventing the formation of scars on the vocal folds. Additionally, the extent of vocal resection during surgery plays a crucial role in the risk of developing vocal fold scars.[16] No global standard protocols are established for voice rest as well as its type and duration. The recommended duration of vocal rest ranges from 3 days to 2 weeks but most surgeons recommended 7 days. Patients are also advised to not cough, avoid heavy lifting, and throat clearing.[16] Previous studies rarely mention the period of voice recovery after surgery, but mostly the evaluation was done 1–4 months after surgery but seldom showed near back to normal vocal outcome with the decrease of VHI score around 6 points in the period of 4 months postoperatively.[16,17]

In our case, the patient showed quick vocal recovery where the initial VHI was 34 before surgery, decreased to 4 only 3 weeks after surgery with G1-R1-B0-A1-S1 which was nearly returned back to its normal state. However, the patient was still advised to limit voice usage, voice therapy, anti-reflux medication, diet and lifestyle modification, also smoking cessation in order to prevent recurrence. In our case, the patient had no recurrence seen after 6 months and 1 year of follow-up.

Conclusion

We reported a ductal vocal cord cyst in patient presenting with dysphonia which was treated successfully with microlaryngeal surgery and intralaryngeal steroid injection with the return of

normal voice within 3 weeks postoperatively. This may be a simple case, but we must not ignore the histological feature of a benign lesion and its extension, also we must consider that not all benign lesion is caused by vocal abuse, other factors may also contribute to the development of this disease so education and postoperative care is a mandatory matter in the management of a ductal laryngeal cyst to achieve good vocal outcome.

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Author Contributions

Hesti D Palupi and Anisa H Khoiria contributed to the diagnosis and management of this case. Anisa H Khoiria and Ardhila R Rasyada wrote the first paper draft. Hesti D Palupi and Anisa H Khoiria made revisions to build the final draft of the paper. All authors were responsible for the authenticity of this manuscript.

Availability of Data and Materials

All images have been submitted with the manuscript.

Consent for Publication

A signed consent by the patient was provided for the patient's images and case history used for this publication.

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