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CASE REPORT

Laryngeal Actinomycosis -A Case of 36 Year Old Female at Hawassa University Comprehensive Specialized Hospital in Hawassa, Sidama, Ethiopia, Ethiopian Patient

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Background: Actinomycosis is a granulomatous infection produced by filamentous, gram-positive anaerobic bacteria. Due to its rarity, ambiguous symptoms, and resemblance to more frequent disorders, including cancer, Crohn's disease, and tuberculosis, it is a challenging disease to identify preoperatively.

Case Presentation: Our case was a case of a 36-year-old woman from the Oromia region's West Arsi zone, who presented with a 6-month history of snoring, dysphonia, and cough, as well as significant but unquantified weight loss, fatigue, and low-grade and intermittent fever, for which she had visited various health facilities with no noticeable improvements in her symptoms. She has had diabetes for the past 5 years and is on Metformin 500 mg twice a day with poor glycemic control. The physical findings at the presentation, including the throat examination, were unremarkable. The random blood sugar level was 300 mg/dl at the time of presentation (elevated). Laryngoscopy revealed an irregular tumor on the anterior one-third of the vocal cord bilaterally, involving the anterior commissure. The biopsy result revealed actinomycotic granules with abscess formation. The patient was then started on Penicillin G, and there was a resolution of her symptoms during follow-up, and then on put on Amoxicillin for the next 6 months, which was discontinued when she had fully recovered from her symptoms and the mass had been cleared on follow-up laryngoscopy.

Conclusion: Laryngeal actinomycosis closely resembles laryngeal cancer and other common inflammatory conditions like laryngeal tuberculosis. It is recommended that clinicians, particularly otolaryngologists, be aware of such rare but eerily similar disease conditions so that unnecessary interventions can be avoided on time.

Keywords: Laryngeal actinomycosis, laryngeal cancer, Penicillin G

Introduction

Actinomycosis is a rare chronic granulomatous disease caused by anaerobic filamentous gram-positive bacteria.[1,2] It has a global distribution, primarily affects middle-aged people, and is two to four times more common in men.[2–5] The main human pathogen is Actinomyces israelii.[1,2,5] Granulomatous tissue, extensive reactive fibrosis and necrosis, abscesses, draining sinuses, and fistulas form as the infection progresses[5].

Actinomyces species, which are not usually thought of as opportunistic infections, use tissue injury or mucosal breach to infiltrate surrounding structures in the head and neck. As a result, dental infections and oromaxillofacial injuries are common predisposing factors.[6,7] Direct infection is the most common mode of transmission, with hematogenous spread occurring only in rare cases. The potential for cervicofacial actinomycosis to spread despite anatomical obstacles such as fascial planes or lymphatic drainage, as well as the development of numerous sinus tracts, is a defining feature.[1,2]

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The cervicofacial area (50%) is the most common site of infection, followed by the abdominal (20%) and thoracic (15–20%) regions.[1,2] As the infection spreads continuously, regardless of tissue planes, the disease tends to stay confined. Lymphadenopathy is a rare occurrence.[1,2,5]

Symptoms include lethargy, fever, weight loss, and upper airway obstruction (UAWO), and the disease has a prolonged, indolent course. A palpable lump, visible sinus tracts, and fistulas are all possible physical findings. Two possible laboratory findings are anemia and leukocytosis.[1,2]

Histologic detection of actinomycotic sulfur granules and/or culture of Actinomyces are commonly used to make a conclusive diagnosis.[1,2] Pus is the best specimen for culture. Sulfur granules are Actinomyces colonies that are distinguished by a granulation tissue zone enclosing one or more oval eosinophilic granules. These granules emit beaded or filamentous gram-positive bacilli that are not acid-fast.[8] Sulfur granules are suggestive of actinomycosis but not pathognomonic, as species such as Nocardia or Aspergillus species might have a similar appearance.[8,9.]

Many people have their tumors removed before a diagnosis can be made. In individuals whose diagnosis has been verified prior to surgery, initial medical treatment is reasonable. Penicillin is the antibiotic of choice. The recommended treatment is Penicillin G (10 to 20 million units per day, split every 4 to 6 hours) for 4 to 6 weeks, followed by oral penicillin (2 to 4 g/day) or amoxicillin for 6 to 12 months. Tetracycline, erythromycin, or clindamycin are good choices for penicillin-allergic patients.[5,10–13]

Surgery is typically reserved for patients who have significant lesions with severe necrosis and massive abscess formation, as well as for the excision of chronic fistulas, recurring disease, or the incapacity to rule out malignancy.[13,14] Patients who have their tumors removed should also be given high-dose antibiotics. The outcome is satisfactory in more than 90% of cases when medical and surgical treatments are combined. It is unusual for people to die as a result of Actinomycosis infection.[5,13]

Ethical Review

Written informed consent was obtained from the patient for the publication of this case report after a letter of permission was gained from the Hawassa University Institutional Review Board (IRB).

Case Presentation

This is the case of a 36-year-old woman from the Oromia region's West Arsi zone, who presented with a 6-month history of snoring, dysphonia, and cough, as well as significant but unquantified weight loss, fatigue, and low-grade and intermittent fever, for which she had visited various health facilities with no noticeable improvements in her symptoms. She has had diabetes for the past 5 years and is on Metformin 500 mg twice a day with poor glycemic control. The physical findings at the presentation, including the throat examination, were unremarkable. The random blood sugar level was 300 mg/dl at the time of presentation (elevated). Laryngoscopy revealed an irregular tumor on the anterior one-third of the vocal cord bilaterally, involving the anterior commissure. The biopsy result revealed actinomycotic granules with abscess formation. The patient was then started on Penicillin G (for 1 month), with the resolution of her symptoms during follow-up, and then on Amoxicillin for the next 6 months, which was discontinued when she had fully recovered from her symptoms and the mass had been cleared on follow-up laryngoscopy.

Up on investigation, the complete blood count, erythrocyte sedimentation rate, renal function tests, and liver function tests were all within normal ranges. Serology tests for the Human Immunodeficiency Virus (HIV), Hepatitis B surface Antigen (HbsAg), and Hepatitis C virus RNA (HCV RNA) revealed negative. Her random blood sugar level was 300 mg/dl, which is high, and her glycemic control was similarly poor on the follow-up chart.

Her otolaryngology exam was unremarkable, but a flexible laryngoscopy revealed irregular mass in the anterior one-third of the vocal cord on both sides, with involvement of the anterior commissure (Figure 1).

Actinomycotic granules were seen when the mass was examined histopathologically with hematoxylin and Eosin stain. Round-oval masses with a basophilic appearance and a finely eosinophilic border defined the granules, indicating actinomycosis at different levels of magnifications (Figure 2A–C).

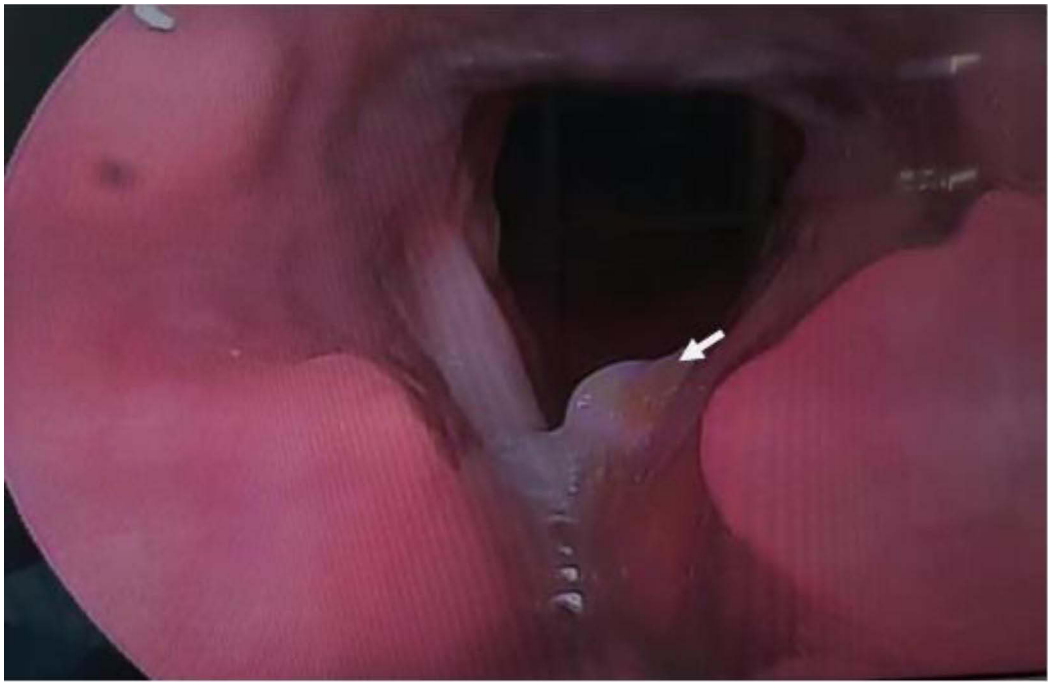

Figure 1 Shows a mass in the anterior one-third of the left vocal cord, the anterior true vocal folds bilaterally, and the anterior commissure (arrow head).

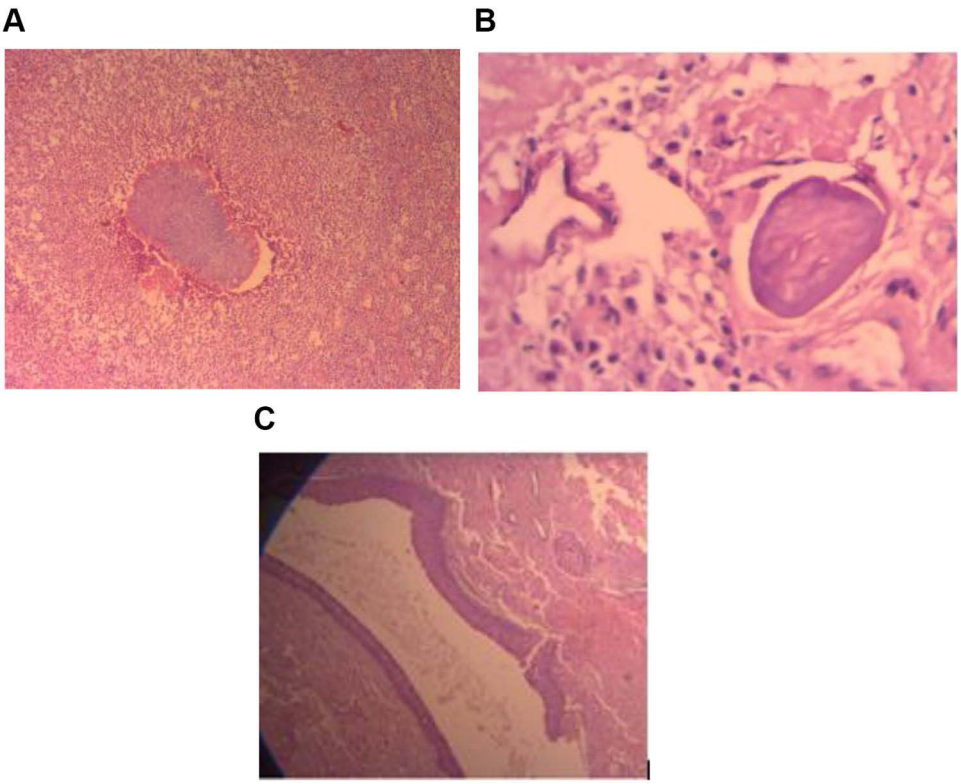

Figure 2 Shows Histopathologic images of the case at different levels of magnifications. Histopathological image at low magnification (4x) shows dense sheets of neutrophils around huge colonies of Actinomyces and a pink rim at the colony's edge, known as the splendore-Hoeppli phenomenon (**A**). Histopathological picture of the squamous epithelial lining of the larynx with subepithelial sulfur granules at medium power magnification (20x) (**B**). Sulfur granules surrounded by dense sheets of neutrophils in a high magnification (40x) histopathology picture (**C**).

Discussion

Laryngeal actinomycosis has been nicknamed "head and neck disease's great masquerader"[6,15]. This emphasizes not only its enigmatic nature, but also the fact that most doctors are unfamiliar with it. As a result, proper procedures for obtaining a sufficient specimen for histopathologic evaluation and culture are frequently overlooked.[16,17]

Because actinomycosis generally responds, at least initially, to short courses of broad-spectrum antibiotics administered in the false notion that the patient has a bacterial odontogenic infection, this patient stayed such a long time with the diseases without remarkable improvement of her disease condition. She did not responded remarkably, perhaps, due to inadequate dosing/duration of treatment. As a result of the delayed diagnosis, the illness course becomes more chronic and fibrosis becomes more prevalent. The increased fibrosis also makes histologic study of typical sulphur granules difficult. As a result, it is prudent to consider such unusual instances, particularly when the response to empirical antibiotics is prolonged.[6,10,15,18]

The fibrotic and "woody" induration typically resembles a malignant process when the disease advances with repeated courses of antibiotic therapy. The granulomatous appearance of a biopsy specimen is sometimes misinterpreted for tuberculosis. Even more frequently, granules are misinterpreted as a sign of nocardiosis by staining or biopsy. These mistakes can be avoided by remembering that malignant lesions should not react to antimicrobial therapy at all, and that whereas Nocardia and mycobacterial species are acid fast, Actinomyces is not.[8,19–21]

Antibiotic treatments for laryngeal actinomycosis have not been studied in randomized controlled trials. In vitro antibiotic susceptibility testing and short case series are used to develop treatment regimens.[12,22,23] Antibiotic treatments for cervicofacial actinomycosis are frequently required. In more complex cases, surgical intervention may be required. Our patient responded significantly to Penicillin G she was taking for 1 month and Amoxicillin for the next 6 months. There is almost similar reports.[12,18,24]

Antibiotic regimens effective against Actinomyces alone are frequently curative; therefore therapy does not need to be focused against other commensal flora that might be isolated alongside Actinomyces species. Because of this reason, we used a monotherapy.[25]

We treated the patient for a total of 7 months, as the length of treatment should be determined by the severity of the infection and the presence of clinical or pathologic remission.[12,25] We used the clinical and laryngoscopic remission to stop the treatment (Figure 3).

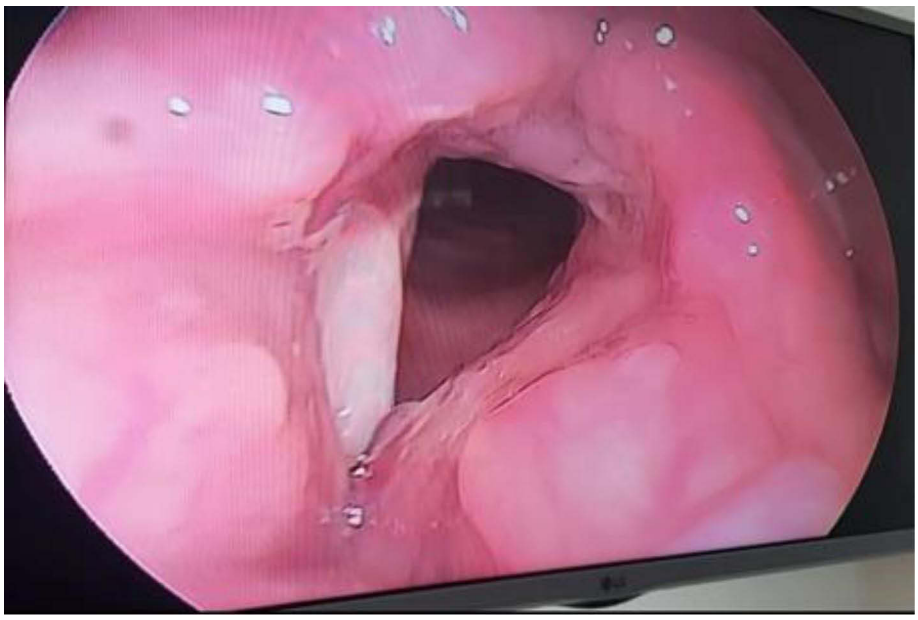

Figure 3 Shows Post treatment laryngoscopy examination showed significant improvement with no focal lesion.

A favourable clinical response is usually achieved with a combined medical-surgical strategy.[24] However, it is suggested that patients be observed for a long time after therapy to quickly detect individuals who have recurrences. Our patient is now on her 12 months since the initial diagnosis and on 5th month since we stopped the medication. She is doing fine, and hence there is no place of surgery in a so far condition.

Conclusion

Laryngeal actinomycosis is a great masquerader of laryngeal cancer and other common inflammatory conditions like laryngeal tuberculosis. It is advisable for the clinicians, particularly otolaryngologists, to be aware of such a rare, but mimickery disease conditions so that unnecessary interventions would be halted timely.

Data Sharing Statement

The data used to support the findings of this study will be available from the corresponding author upon reasonable request.

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Author Contributions

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising, or critically reviewing the article; gave the final approval of the version to be published; agreed on the journal to which the article has been submitted; and agreed to be accountable for all aspects of the work.

Disclosure

The authors declare no conflicts of interest for this work.

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