Case Report

Coexistence of an Epidermal Inclusion Cyst and Actinomyces in the Palatine Tonsil: A Case Report

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Abstract

Epidermal inclusion cysts have an incidence of less than 0.01% in locations such as the tonsillar fossa and can result in a foreign body sensation while swallowing in patients. Epidermal inclusion cysts in a palatine tonsil are an infrequent finding, let alone with their association with Actinomyces colonies. Although radiology and fine needle aspiration cytology can guide clinicians, the final diagnosis lies with histopathology. A 34-year-old female patient presented to the ear-nose-throat department with chief complaints of swelling in the right side of her throat for the last 1.5 years and foreign body sensation while swallowing for the last 3 months. Histomorphology showed multiple cystic lesions surrounded by lymphoid tissue and lined by keratinizing stratified squamous epithelium intermixed with numerous cleft spaces and keratin flakes. Alongside also seen were mixed inflammatory infiltrate and multiple colonies of Actinomyces among the hyperplastic lymphoid follicles. A final diagnosis of multiple tonsillar epidermal inclusion cysts along with Actinomyces colonization was made. Owing to the rarity of such conditions, we would like to add another case report to the English literature, so that the treating physicians and diagnosing histopathologists are well-versed with such conditions.

Keywords: Actinomyces; Epidermal inclusion cyst; Palatine tonsil; Histopathology

Introduction

Several cysts are known to arise in the tonsillar area, with tonsillar retention cysts being the most common. Other rare lesions include epidermal inclusion cysts (EC), lymphoepithelial cysts, and hydatid cysts.[1] Among them, EC are lesions with an incidence of only 1.6 to 7% in the head and neck region.[2] On an extensive English literature search, tonsillar location remains extremely rare, with an incidence of less than 0.01%.[3, 4, 5] Two hypotheses have been put forth for the genesis of EC, one being the inclusion of ectodermal tissues during embryogenesis and the other being the introduction of metaplasia in response to prolonged irritation.[6] Actinomyces acts as a commensal of the oropharynx, but some authors attribute it to a local chronic irritant resulting in the development of tonsillar EC.[7] Tonsillectomy, cyst extirpation, and histology confirmation are the treatments for these lesions. Due to its rarity, we would like to add another case of EC in the tonsillar area with Actinomycetes colonization.

Case Report

A 34-year-old female patient presented to the ear-nose-throat (ENT) department with chief complaints of swelling in the right side of her throat for the last 1.5 years and foreign body sensation while swallowing for the last 3 months. Local examination

revealed grade III right tonsillar hypertrophy with swelling 3x3 cm in size, and the uvula was seen pushed to the opposite side by the mass. Contrast-enhanced computed tomography (CECT) neck was done, showing a well-defined lobulated lesion measuring 2.2x2x1.4 cm with mild patchy enhancement arround the right palatine tonsil and right oropharyngeal wall protruding into the oropharyngeal airway, causing its narrowing. (Figure-1A, C, D) A course of antibiotics was initially administered, but there was little improvement. The patient, then, underwent a right tonsillectomy along with an excision of the mass. Intraoperative findings revealed a lobulated growth over the right superior pole of the tonsil, accompanied by right tonsillar hypertrophy. (Figure-1B)

On removing intraoperatively, cheesy material was seen extruding from the growth over the right superior pole of the palatine tonsil. The cut surface showed multiple cystic cavities with cheesy pultaceous material on gross examination. (Figure-2) Histomorphology showed multiple cystic lesions surrounded by lymphoid tissue and lined by keratinizing stratified squamous epithelium intermixed with numerous cleft spaces and keratin flakes. Alongside also seen are mixed inflammatory infiltrate and multiple colonies of Actinomyces among the hyperplastic lymphoid follicles. (Figure-3) A special stain (Grocott Methenamine Silver) was done, which showed large basophilic filamentous bacterial aggregates.

A final diagnosis of multiple tonsillar epidermal inclusion cysts along with Actinomyces colonization was made. The patient was commenced on empiric antibiotic therapy and subsequently demonstrated steady clinical improvement on follow-up. Written informed consent was obtained from the patient, and ethical approval was obtained from the institutional ethics committee.

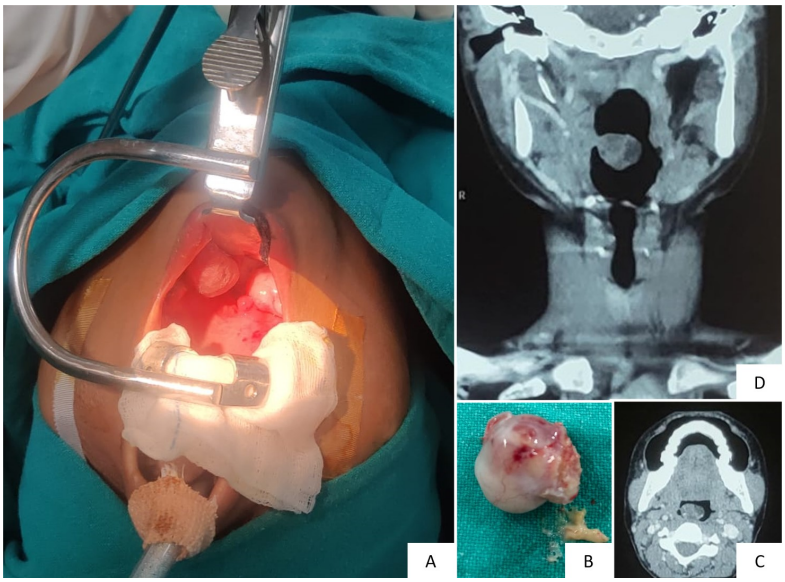

Figure 1: (A, B) Intraoperative photographs of a lobulated mass in the palatine fossa, (C, D) CECT showing a well-defined lobulated lesion measuring 2.2x2x1.4 cm with mild patchy enhancement arround the right palatine tonsil and right oropharyngeal wall protruding into the oropharyngeal airway, causing its narrowing.

Discussion

The palatine tonsil is a mass of lymphoid tissue situated in the tonsillar fossa, where it lies in the lateral wall of the oropharynx between the anterior and posterior faucial pillars. Many intraoral cysts, including epidermal, dermoid, and teratoid, are identified, which are considered cystic malformations lined by squamous epithelium and show overlapping features.[8] Out of them, EC is a common cystic lesion lined by stratified squamous epithelium, and the cyst wall does not contain any sebaceous glands, hair follicles, or eccrine glands.[3] EC can occur in any part of the body, with about 7% of them occurring in the head and neck region. In the neck region, ECs need to be differentiated from a ranula, thyroglossal duct cyst, branchial cleft cyst, cystic hygroma, cervical thymic cyst, and cervical bronchogenic cyst. The palatine tonsil becomes an extremely rare location for them to occur (less than 0.01%), which makes our case worth mentioning.[3, 4, 5]

The majority of patients lie in the age group of 15 to 35 years, with male preponderance. Pathogenesis underlying the development of EC includes various theories, such as ectodermal tissue during embryogenesis and metaplastic changes due to chronic irritation.[7] Actinomyces can colonize keratinous spaces, as seen in our case.[9] These are gram-positive, non-acid-fast, anaerobic, or microaerophilic filamentous, branched bacteria that are difficult to grow in culture and are mainly seen in the cervicofacial region.[9]

A few hereditary conditions, like Gardner syndrome and Lowe syndrome, are known to be associated with EC.[6] Gardner syndrome is a variant of familial adenomatous polyposis (FAP) associated with extra-colonic features. There are 50-65%

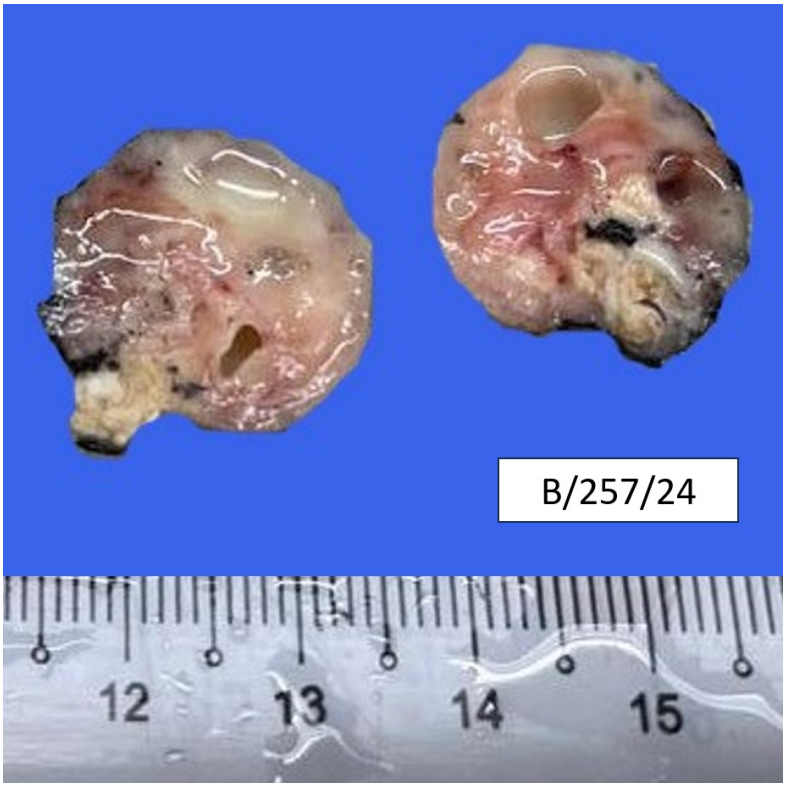

Figure 2: Cut surface of the gross specimen shows multiple cystic cavities with cheesy pultaceous material.

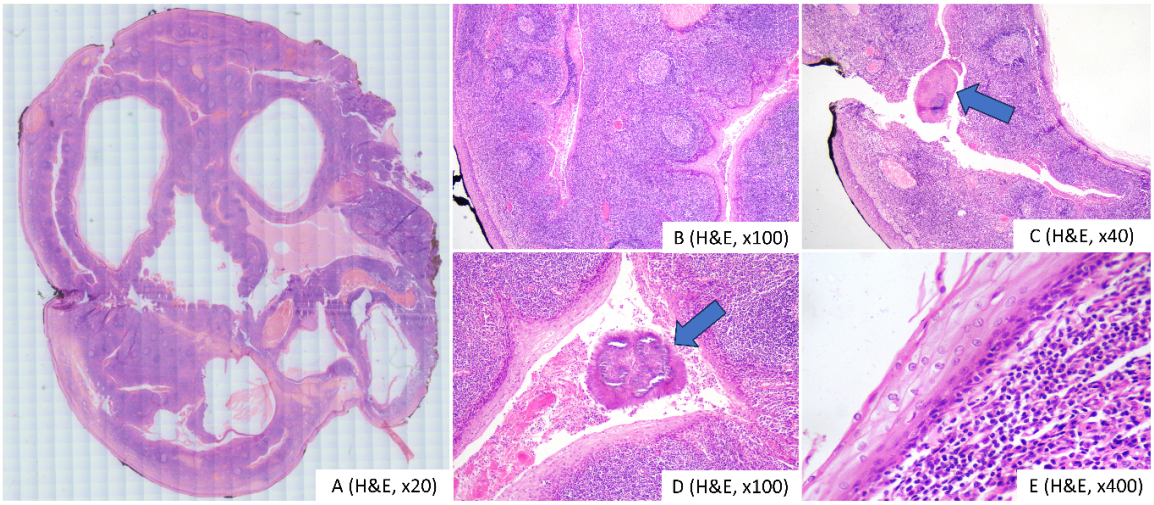

Figure 3: (A) Histopathological examination shows cystic lesions within tonsillar parenchyma (H&E, x20), (B) histopathological examination shows reactive lymphoid tissue with germinal centers (H&E, x100), (C, D) histopathological examination shows Actinomyces colonies (arrow) (H&E, x40 & x100), and (E) histopathological examination shows cystic cavity within the tonsillar tissue lined with keratinized epithelium (H&E, x400).

chances of the formation of multiple ECs in a patient with Gardner syndrome, and they occur at an early age. Lowe syndrome is also a rare genetic disease affecting the eyes, nervous system, and kidneys. Various cystic lesions have been reported to occur in the scalp, perianal region, and upper and lower extremities in Lowe syndrome.

Definitive diagnosis is made with the help of histopathological examination; however, it can be aided by fine needle aspiration cytology, in which abundant keratin flakes are evident, along with some nucleated squamous cells, multinucleated giant cells, and cholesterol crystals. Radiological investigations help in making a provisional diagnosis of cystic swellings as a low-density, unilocular mass in the background of tonsillar parenchyma, as seen in our case.[10] These investigations are aided by fine needle aspiration cytology and excisional biopsies, helping in landing a definitive diagnosis. Surgical excision remains the mainstay of treatment in such cases, with a good prognostic outcome. Actinomyces commonly reside within tonsillar crypts without causing invasive disease. True actinomycosis requires a prolonged, high-dose penicillin; however,

incidental histologic presence does not warrant treatment, as it does not correlate with symptoms or recurrence.

Conclusion

EC of the palatine tonsil, particularly when accompanied by Actinomyces colonies, can pose diagnostic and therapeutic challenges. By presenting this case, we highlight key learning points for clinicians and histopathologists—specifically, the importance of recognizing this benign entity, its rarity, accurately interpreting the presence of Actinomyces, and guiding appropriate management without overtreatment.

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