Open Access

Journal of Biomedical Sciences

CASE REPORT

DOI: https://doi.org/10.3126/jbs.v11i1.69239

Tubercular Sialadenitis: A rare case report

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Information about the article:

Received: April 30, 2024
Accepted: July 25, 2024
Published online: Aug. 24, 2024

Cite this article:

Jabo A, Afodun AM, Mukangendo M, Kayitesi I, Eze DE, Uwimana L, Ganza J. Quadri KK. Tubercular Sialadenitis: A rare case report. Journal of Biomedical Sciences. 2024;11(1):12-16.

Publisher

Nepal Health Research Society, Bahundhara -6, Gokarnesowor Municipality, Kathmandu, Nepal
eISSN 2382-5545, ISSN 2676-1343 (Print)

ABSTRACT

Background

Tubercular sialadenitis is an uncommon condition of extrapulmonary tuberculosis. It is responsible for fatal infections and ailments caused by Mycobacterium Tuberculosis. Due to diverse non-specific symptoms, it is often misdiagnosed as a form of parotid gland neoplasm. This diagnostic dilemma poses a challenge to medics, especially in a low-income setting of a developing country.

Case presentation

We discuss a case of parotid salivary tuberculosis in a 25-year-old man who was scanned via a high-frequency 7.0 MHz transduced ultrasound scan and later verified histologically. On examination, bilateral submandibular and parotid bulges along the lateral neck region with marked lymphadenopathy were found. Sonography diagnosed him with a case of submandibular gland tuberculosis and confirmed bacteriologically by mycobacterial culture.

Conclusion

Staining for AFB, biopsy and culture studies are necessary and recommended. Establishing a cure requires high awareness, suspicion and early diagnosis.

Keywords

Parotid gland, sialadenitis, tuberculosis, salivary gland, ultrasound

Background

Tubercular sialadenitis is a rare manifestation of tuberculosis, with few reported cases globally. It is classified as extrapulmonary tuberculosis, which mainly affects the salivary glands. The disease often presents with nonspecific symptoms and is challenging to diagnose without appropriate imaging and laboratory support [1]. Ultrasound imaging has shown promise in diagnosing tubercular sialadenitis by identifying characteristic features such as bilateral nodal/reticular changes in the glands [2]. This case report describes tubercular sialadenitis's clinical presentation, diagnostic approach, and ultrasound features.

Even though the pathogenesis of extrapulmonary tuberculosis is still unclear, two mechanism theories explain how tuberculous infection might be present in extrapulmonary organs. It can either be through the hematogenous or lymphatic system [3]. Often, extrapulmonary tuberculosis is a secondary infection with a primary source, most likely in the lungs. From the primary source, it spreads through connective systems like the lymphatic system or blood to the glands, forming an infection resulting in gland inflammation.

A report from the World Health Organization (WHO) on tuberculosis indicates that about 25% of the reported tuberculosis cases are from India [4]. From this percentage, roughly about 30% of tuberculosis cases in India are extrapulmonary tuberculosis. A large portion of these cases are associated with enlarged cervical lymph nodes. The submandibular glands are the most affected by a long-standing infection of the glands by 48%. On the contrary, in about 25% of the cases, the parotid and submandibular glands are affected simultaneously [5].

Clinically, tuberculous sialadenitis presents as swelling in the neck region, which can be unilateral or bilateral. Multiple imaging modalities have been used to assess sialadenitis; however, ultrasound and Computed Tomography are the leading modalities, with 65% and 98% sensitivity and specificity, respectively [6]. Ultrasound shows enlarged glands with reticular or nodular changes and increased vascularity [2]. In this presented case, ultrasound shows multiple nodular changes in the salivary glands of a 25-year-old male patient.

Case presentation

Case Report

A 25-year-old male patient presented to our hospital with neck swelling suggestive of cervical lymphadenopathy. The patient had no history of cough, chest pain, or pain with breathing or coughing. He complained about the swelling on the right lower mandible lasting about a month, with pain radiating to the midline. Initial physical examination indicated bilaterally palpable, multiple, and enlarged (lobulated) lymph nodes in the submandibular, submental, and cervical regions.

Laboratory investigations were carried out. The Auramine Rhodamine Stain (ARS) and Acid-Fast Bacilli test (AFB) were negative, and a gene Xpert MTB/RIF test was later carried out. Mycobacterium tuberculosis DNA was present (MTB was positive); however, there was no resistance to rifampicin (RIF was negative).

Due to persistent symptoms, ultrasound imaging of the neck revealed multiple matted hypoechoic ovoid nodules with several central cystic areas in both the submandibular and submental glands. Based on these findings, an ultrasound (7.0 MHz linear probed/ Vinno X1 Medical Ultrasound 3D and 4D Color Doppler Diagnostic System, model 3KJV-34, made in China) scan confirmed a diagnosis of tubercular sialadenitis, and we recommended biopsy fine needle aspiration (FNA). A sonographic examination was done at the University Teaching Hospital in Kigali, Rwanda. The biopsy concluded a necrotizing granulomatous inflammation likely due to a mycobacterium infection.

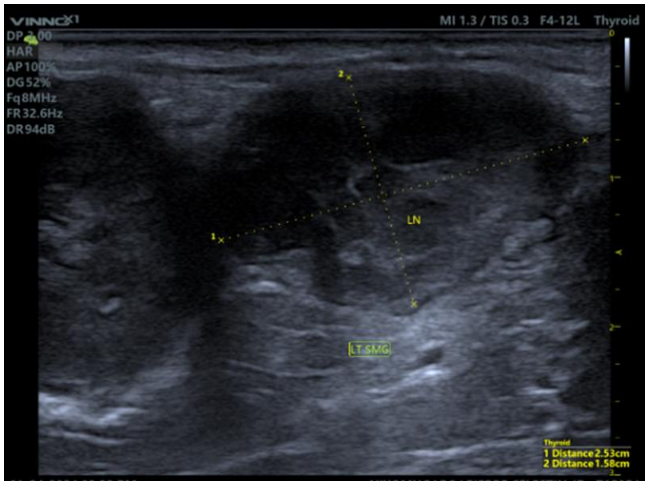

Figure 1: A sonogram of the left submandibular gland using a linear probe shows the dominant enlarged hypoechoic ovoid nodule with central anechoic areas, measuring about 2.53x1.58 cm in AP diameter.

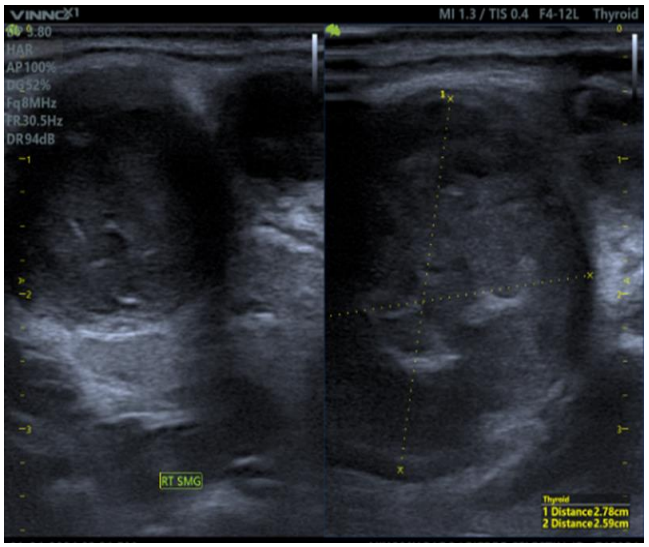

Figure 2: A sonogram of the right submandibular gland in longitudinal and transverse planes using a linear probe shows a well-defined heterogenous ovoid hypoechoic nodule in the right submandibular gland. It measures 2.78x2.59 cm in AP diameter.

These sonograms show bilateral nodular changes in the salivary glands, manifesting tuberculosis. Additionally, the internal anechoic areas represent the necrotic changes of nodules, as later confirmed by biopsy.

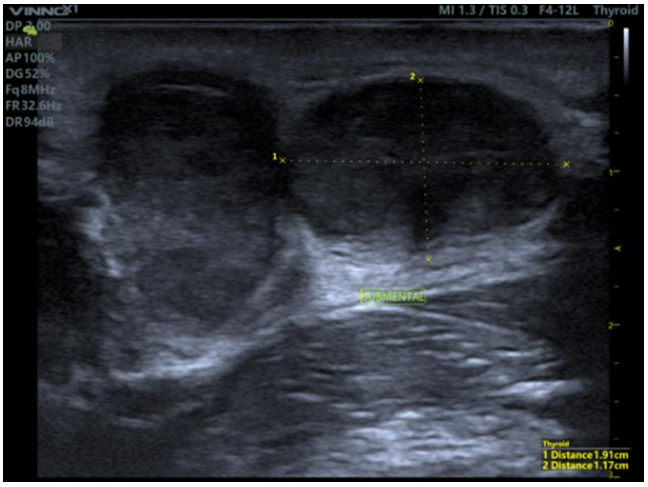

Figure 3: A sonogram of the submental gland taken using a linear probe shows multiple well-defined ovoid hypoechoic nodules in the submental gland with internal necrotic cystic areas - the largest measures 1.91x1.17 cm in AP diameter.

Discussion

Management with surgical operation seems limited from the literature; the high possibility of submandibular tuberculosis gland relapse is common. Removal of the affected glands may be inevitable in some cases; however, the role of surgery is limited if benign and non-metastatic. Information later reached us that he was relapse-free after treatment. Tubercular sialadenitis is a rare capsule of different clinical syndromes. Sonographer intra-observer suspicion is needed in Rwanda; we suggest advanced histopathologic biopsies, micro-bacterial culture, and AFB staining techniques. The recently developed PCR is the most sensitive methodology for demonstrating mycobacterium aspirate.

Tuberculosis Sialadentis can develop into a local infection of the buccal cavity, as shown in Figures 1 and 2, and spread to salivary cell parenchyma by bacteria. This may be facilitated by the anatomy of the gland ductal system. Tubercular sialadenitis has numerous clinical signs that contribute to its diagnostic difficulty. Weight loss, high temperature, nausea, and loss of appetite are usually suppressed in (non-secondary) tubercular sialadenitis. In agreement with our findings (Figures 2 and 3), swelling and abscess formation involving the parenchyma of the pivotal gland related to lymphatic mode infections (lymphadenopathy) were noted. Differentials from our case report may differ from others, such as the situation in Sjogren's syndrome, autoimmune parotitis, facial palsy, and/or sarcoidosis [7].

A drug combination of ethambutol, pyrazinamide, and rifampicin may be recommended as a treatment option. The anatomy of the salivary gland makes it a rare location for tuberculosis, perhaps due to the presence of enzymatic proteolytes and ions of thiocyanate. A logical science to support the above is the lysozyme – enzyme content in salivary juice preventing bacterial build-up. Occasionally, it is impossible to diagnose tubercular sialadenitis before surgical gland excision and histopathological confirmation. A key microscopic feature is the centralized necrosis surrounded by epithelioid cells and giant Langhans cells.

Ultrasound is the radiologic modality of choice when assessing palpable growth and abnormalities of the parotid gland. In line with an observation by Wu et al. [8], the prevalence of tuberculosis is increasing daily; the involvement of extra-pulmonary structures, e.g., the main salivary gland, revives a medical (diagnostic) dilemma. Good clinical observation in line with patient history, medical records, and a high level of clinical suspicion must be highlighted before treatment starts. What makes our case outstanding is because of its isolated nature and negative history of tuberculosis in his family.

Tuberculosis is endemic in some regions of India. Parotid tuberculosis accounts for about 3.5 – 10% of salivary gland tuberculosis. In agreement with our hypothesis (Figure 3), it is assumed there is a spread of mycobacterium from a proximal infected and adjacent source, from the dentition, Waldayer's tonsils, or the uvula. Direct inoculation by sputum will spread bacilli through the 'sensory' lymphatic duct. Similar to the assertions of Thakur et al. [9], the regional spread of mycobacteria is more to the submandibular gland, the opening of Wharton's tube at the inferior (lateral) part of the buccal cavity (floor of the mouth). With ethambutol and isoniazid therapy, partial and salivary gland tuberculosis will have an improved prognosis. Clinical features change from acute to chronic sialadenitis; in line with Figure 2, the latter shows a semblance of a tumorous outgrowth. In some cases, it could be painless, asymptomatic, slow-growing, and devoid of neck lymphadenopathy. Roentgenology of salivary mass – findings can be non-specific or unilateral in both genders [10].

Conclusion

Our case analysis shows that tuberculous parotitis could hold diagnostic accuracy when lung disease manifestation or cervical lymphadenitis is absent. Two types of parotid pathology identified by ultrasound heterogeneity are the peri-parotid and parenchymal types. The superior part of the parotid gland exhibits diffuses (non-uniform) echogenicity in the parenchymal type. In contrast, the 'intra' and 'peri' glandular forms of the peri-parotid gland have embedded lymph. Salivary glands may sometimes be immune to ailments like tuberculosis due to thiocyanate and proteolytic

enzymes (lysosomes). Further studies on enzymatic assays and biopsies on salivary gland apparatus: submandibular, parotid, and lingual glands are recommended when mass swellings are identified by sonography.

Abbreviations

Acid Fast Bacillus (AFB), Auramine Rhodamine Stain (ARS), Fine Needle Aspiration (FNA), Mycobacterium Tuberculosis Complex (MTBc), Rifampin (RIF), University Teaching Hospital Kigali (UTHK), World Health Organization (WHO)

Acknowledgments

We are thankful to the entire staff of the Medical Imaging Sciences Department, School of Health Sciences, University of Rwanda

Authors' contribution

a. Study planning: JA, AAM
b. Case report: JA, AAM, MM, KI, EDE, UL, GJ, QKK
c. Follow up: JA, AAM
d. Interpretation: AAM, MM, JA
e. Manuscript writing: JA, AAM, MM, KI, EDE
f. Manuscript revision: JA, AAM, UL, GJ, QKK
g. Final approval: JA, AAM, MM, KI, EDE, UL, GJ, QKK
h. Agreement to be accountable for all aspects of the work: JA, AAM, MM, KI, EDE, UL, GJ, QKK

Funding

No funding was received.

Availability of data and materials

Figures of this case report are available as part of the article, and no additional image files are required.

Consent for publication

In accordance with the 1975 Helsinki Declaration on patient rights, consent was sought and obtained from the patient, and ethical approval was granted. No patient name tag or any form of confidential personal information is compromised in this manuscript; a copy of the approval letter is available on demand by the editor.

Competing interests

None declared.

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