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RESEARCH ARTICLE

ISOLATED TUBERCULOSIS OF PAROTID GLAND:A COMMON DISEASE AT AN UNCOMMON LOCATION WITH REVIEW OF LITERATURE

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Abstract

Tuberculosis infection is a common health problem in developing countries like India which is caused by Mycobacterium tuberculosis (MTB). It commonly affects the pulmonary system. Although it can affect any organ of the body but salivary glands especially the parotid gland arethe rarest entity that is involved in tuberculosis. Less than 200 cases have been reported in literature till now and mostly diagnosed in post-parotidectomy specimens. Generally, it presents with a painless growing mass and hence, its clinical diagnosis is challenging and may be misdiagnosed as it can mimic parotid gland neoplasm on physical examination. Radiological investigations such as ultrasonography (USG) and computed tomography (CT) are not much beneficial. In such cases, fine needle aspiration cytology (FNAC) is the preliminary step for the workup of the patients. Histologically diagnosis is confirmed by the presence of well-formed epithelioid cell granuloma and acid-fast bacilli (AFB) stain is positive for tubercular bacilli. Here we report a case of tubercular parotitis in a 50-year-old female diagnosed on histopathological examination and patient treated with anti-tubercular therapy (ATT) successfully.

Introduction:-

Tuberculosis (TB) is a common disease in developing countries like India. In the literature, less than 200 cases of parotid TB were reported worldwide.[1,2]Lungs are most commonly affected and present with pulmonary manifestations.TB can also affect the gastrointestinal tract, musculoskeletal system, lymph nodes, central nervous system, and reproductive system. Apart from lymph nodes, tuberculosis rarely affects the head and neck system. Salivary gland involvement by tuberculosis is the rarest condition because the continued flow of the salivary gland stops the tubercular bacilli from accumulating at that site.[3]TB in the parotid gland is a rare entity, itusually presents as a slow-growing mass, which can be mistaken for parotid gland neoplasm.[1,4,5.]Herein, we report a case with left parotid swelling which was difficult to diagnose as TB on Fine needle aspiration cytology (FNAC).

Case report:-

A 50-year-old female presented in the Otorhinolaryngology outpatient with complaints of pain and swelling in the left parotid region in July 2022, clinically considered acute parotitis (Figure 1).The patient was treated at various centreswith antibioticsand anti-inflammatory drugs. However, no improvement in symptoms was noticed. Ultrasonography of the parotid region was performed and it was suggested suppurative lesion. Fine needle aspiration cytology (FNAC) ofleft parotid swelling showed features of granulomatous sialadenitis. However, the Ziehl Neelsen

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(ZN) stain for Acid-fast bacilli (AFB) was negative. Simultaneously, an X-ray chest was performed to look for findings of primary pulmonaryTB (PTB)but it was normal (Figure 2).
Further, MTB PCR was performed and no MTB complex was detected.Excision biopsy was doneunder local anaesthesia and sent for histopathological examination which showed numerous necrotizing epithelioid cell granulomas. However, AFB stain for tubercular bacilli and Per-iodic stain for fungal elements were negative Since MTB PCR was also negative, no antitubercular drugs were started. The patient was also doing well after the excision biopsy but after two months of postoperative periods, the patient developed a draining sinus. The patient was managed conservatively withantibiotics and anti-inflammatory drugs. However,no improvement was noticed.Finally, surgical exploration along with parotidectomy was performed under general anaesthesia. Thehistopathological examination of the specimen showed a similar picture of necrotizing granulomatous sialadenitis. Post-surgery, the patient developed a draining sinus in infra parotid region which was not responding to antibiotics. Thereafter,the histopathological slides were reviewed. At this time her erythrocyte sedimentation rate (ESR) was 62mm/hr and C-reactive protein (CRP) was 11.8 mg/L. On microscopic examination, H &E-stained sections showedepidermis and dermis with many necrotizing epithelioid cell granulomas with multinucleated giant cells and Langhans-type giant cells. The granulomas were extending into the underlying muscle and subcutaneous tissue. Also, the parotid gland showed dense lymphoid infiltration, and acinar atrophy (Figure 3a and 3b).Ziehl Neelsen stain for AFBwas performed which displaysa few pink-coloured curved, beaded and non-refractile acid-fast bacilli indicating tuberculosis (Figure 3c). The final impression was rendered as Tubercular parotitis. Then, the patient took anti-tubercular therapy for 6 months resulting in the complete resolution of the parotid pain and swelling.The patient came for follow-up after 6 months and is completely fine.

Discussion:-

Salivary glands are the exocrine glands. They secrete saliva into the oral cavity and help with swallowing and chewing. These glands are divided into major and minor salivary glands. The largest salivary gland is the parotid gland followed by the submandibular gland which is the second largest. [6,7]Tuberculosis is a necrotizing granulomatous condition caused by Mycobacterium Tuberculosis Bacilli (MTB) with varied clinical manifestations. Worldwide, according to the Global TB Report 2022, the estimated incidence of tuberculosis was reported 10.6 million cases in 2021.[8,9] In recent times,the incidence has increased in developed countries due to factors like co-infection with HIV and the development of drug-resistant strains. PTB is the commonest presentation while 15-20% of cases show extrapulmonary tuberculosis such as cervical lymphadenitis. TB in the parotid gland is extremely rare even in endemic countries.[10,11]The Salivary glands are usually spared of TB because of the bactericidal action of saliva due to the presence of thiocyanate ions and proteolytic enzymes. The parotid gland most commonly gets infected by direct extension of mycobacterium from the oral cavity via its duct.[1]

The pathogenesis of parotid gland TB remains unclear and there are different theories. One of the earliest reports given by Van Stubenrauchmentioned that the main mode of salivary gland TB is the extension of infection along the Stenson's duct from the oropharynx. Bockhorn proposed a hematogenous spread from any primary focus in the body. Another report given by Berman and Fein proposed that the infection can reach to parotid gland via lymphatics, usually from infected tonsils.[11,12]

Clinically, parotid TB presents varyingly but the most common manifestation noted is the painless slowly growing mass in the parotid region. Sometimes it presents as sialadenitis with diffuse enlargement of the parotid gland as seen in the present scenario.

Parotid TB is a true diagnostic challenge in the absence of clinical symptoms and without involvement of the respiratory disease.Radiologyand FNAC are important tools for early diagnosis. As reported, FNAC has 81-100% sensitivity and 94-100% specificity for diagnosing cases of parotid TB.[13]However, sometimes it is difficult to make a diagnosis on cytology due to the presence of necrosis in both tubercular aetiology, fungal infection and malignancy and especially when AFB stain is negative.[12]Hence, post-parotidectomy surgery histopathological examination is needed. After confirming the diagnosis of TB, ATT is required for treatment. Early diagnosis is required with high clinical suspicion to avoid unnecessary surgical intervention.

According to the World Health Organisation (WHO), a six-month regimen is recommended for drug-sensitive TB except for CNS, joint or bone cases. The six-month regime involves two months of intensive phase therapy of rifampicin, isoniazid, ethambutol, and pyrazinamide followed by a fourmonth continuation phase regimen of rifampicin and isoniazid.[14]

A review article published in 2022 documented a total of 160 articles on parotid TB. Among these, 84 articles were in a different language or did not report cases of tuberculosis on parotid. 25 articles from the remaining 76 articles were published before the year 2000, 27 articles were published between 2000 to 2010 years and only 24 articles were published after 2010. From India,the majority of articles published approximately 26 in number and from Turkey (15), the UK (8), the US (5), China (4), Greece (3), Morocco (3), South Africa (3), and Taiwan (2) in number.[15]The research highlights that cases of parotid TB are rarely diagnosed because it is mainly seen in the tubercular prevalent areas andwhere the immigration rate is high. Another reason for the missing findings in slides is the good AFB stain and vigilant screening of the AFB slides, especially if the bacilliary load is low and trained eyes and it is generally seen in under-developed healthcare sectors [15]as we have seenin the present case.

Conclusion:-

This case report highlights the rarity and diagnostic challenges associated with parotid TB.Due to its similarity with the other conditions of parotid neoplasm both clinically and radiologically, a high index of suspicion for parotid TB is necessary especially if there are non-resolving draining sinuses or recurrence of the lesion in tuberculosis prevalent regions. Early recognition can prevent unnecessary surgical interventions and facilitate prompt initiation of appropriate medical treatment. Additionally, raising awareness about this unusual presentation can aid in timely diagnosis and management.

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Figure.1: 50-year-old female with parotid swelling on left side.

Figure.2 Chest X-ray (PA view) of the patient reveals no significant abnormality

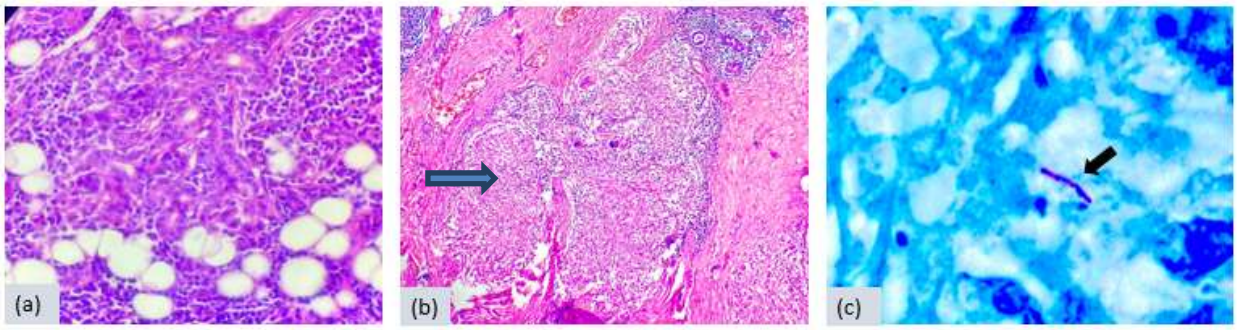

Figure3(a) Sectionshowing normal parotid gland morphology (Hematoxylin& Eosin stain, 40X) (b) Section showing well-formed epithelioid cells granuloma.(arrow)(Hematoxylin& Eosin stain, 20X) (c) Ziehl Neelsen staining under oil immersion (100x) showing acid-fast bacilli (arrow).