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# Pleomorphic adenoma of the parotid gland concomitant with tuberculosis infection: A case report and review of the literature

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## Abstract

Tuberculosis of the salivary glands and particularly of the parotid gland is a localization that remains rare even in endemic countries. The association of intra parotid tuberculosis with a benign tumor has been found only in rare cases in literature. A 50-year-old woman with a history of normal pressure hydrocephalus treated surgically, non-smoker, presented with a right parotid swelling progressively increasing in size for 3 years. Clinical examination revealed a 4 cm long, firm, mobile, painless parotid swelling without inflammatory signs and without accessory lymphadenopathy. The oropharyngeal examination was without abnormalities. Ultrasound showed a mass of 31 × 27 mm suggesting a pleomorphic adenoma. MRI confirmed the suspicion of a pleomorphic adenoma of both lobes. The patient underwent a conservative total parotidectomy. The extemporaneous examination was in favor of a pleomorphic adenoma while the final pathology showed the coexistence of active tuberculosis lesions. The patient was put on long-term antituberculosis treatment with good clinical evolution. The clinical presentation of parotid tuberculosis is nonspecific mimicking any other tumor and the diagnosis can only be made by histological examination. Therapeutic management is based on long-term antituberculosis treatment.

## Keywords

Primary tuberculosis, pleomorphic adenoma, total parotidectomy, parotid gland

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## Introduction

Head and neck tuberculosis represents approximately 10% of all mycobacterial infections,[1] yet parotid gland tuberculosis is very rare accounting for less than 1% of those localizations.[1,2] It was first described by von Stubenrauch in 1894.[3] Since then, fewer than 200 cases of parotid tuberculosis have been reported in the literature[4,5] and only one other case of pleomorphic adenoma concomitant to the mycobacterial infection has been described in recent literature.[6]

## Case report

A 50-year-old woman with a history of normal pressure hydrocephalus treated surgically, non-smoker, presented with a right infra-auricular swelling progressively increasing in size for 3 years. The patient had no history of tuberculosis, no recent weight loss or chronic cough, and did not report any other symptoms. Clinical examination revealed a 4 cm long, firm, mobile, painless parotid swelling without inflammatory signs. No accessory lymphadenopathy was palpable. No other facial deformities or signs of facial nerve weakness were found. The oropharyngeal examination was also without abnormalities. Blood counts and chest x-ray were normal. The tuberculin test and polymerase chain reaction (PCR) analysis for mycobacterial tuberculosis were not done because parotid tuberculosis was not suspected before surgery. Ultrasound showed an oval hypoechoic heterogeneous mass of 31 × 27 × 31 mm suggesting a pleomorphic adenoma (Figure 1). MRI showed an intermediate signal

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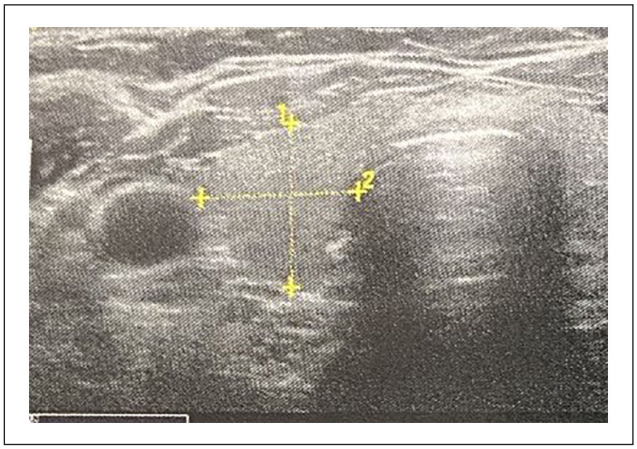

**Figure 1.** Cervical ultrasound showing a tissular heterogeneous hypoechoic mass with clear contours evoking a pleomorphic adenoma of the parotid gland.

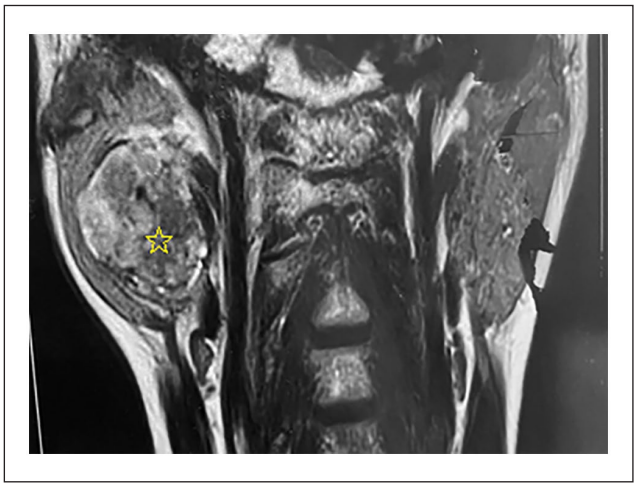

**Figure 2.** Axial T2-weighted study MRI showing a heterogeneous hypersignal mass of the right parotid gland involving both lobes.

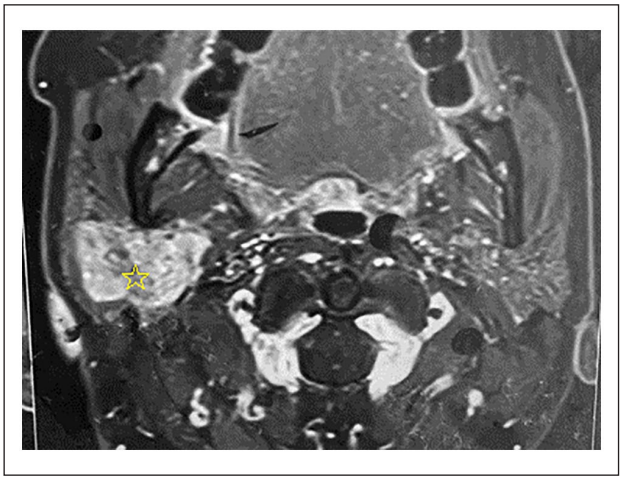

**Figure 3.** Coronal T2-weighted study MRI showing a heterogeneous hypersignal mass of the right parotid gland involving both lobes.

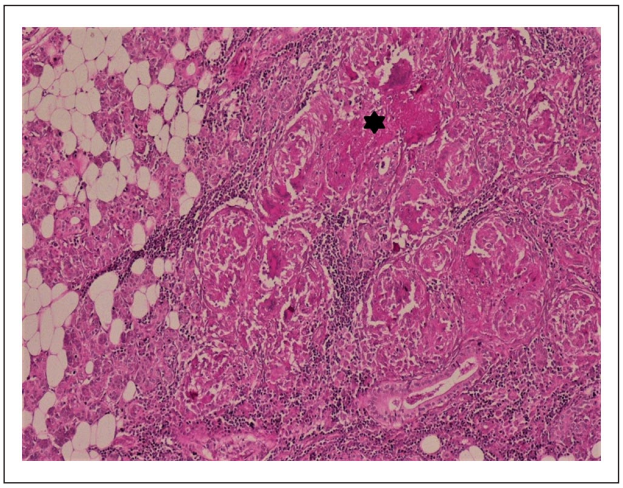

**Figure 4.** Numerous epithelioid and giganto-cellular granuloma with central caseous necrosis in the parotid parenchyma (HEx20) ★.

intensity on T1-weighted study, heterogeneous high signal intensity on T2-weighted images (Figures 2 and 3), and heterogeneous contrast uptake. Diffusion weighted study showed a hypersignal mass, but ADC was not measured. This confirmed the suspicion of a pleomorphic adenoma of both lobes, so the patient underwent a conservative total parotidectomy. Facial nerve monitoring was not available and thus was not used in this case. The intraoperative findings showed a tumor involving both the superficial and deep lobes with mass effect on the cervical branch of the facial nerve, which was laminated and congestive.

The extemporaneous examination was in favor of a pleomorphic adenoma while the final pathology showed the coexistence of active tuberculosis lesions (Figure 4). During the postoperative course in hospital, a paresis of only marginal mandibular branch was noted and recovered spontaneously. A thoracic, abdominal, and pelvic computed tomography was done and showed no other concomitant tuberculosis lesions. The patient was put on antituberculosis treatment for 8 months. It consisted of a combined quadritherapy for 2 months, according to the guidelines of the World Health Organisation[7]: isoniazid, rifampicin, pyrazinamide, and ethambutol then a 6-month bitherapy: isoniazid and rifampicin. Because tuberculosis was not initially suspected and was an accidental discovery, diagnosis was confirmed solely by pathology. Cartridge Based Nucleic Acid Amplification Test (CBNAAT), Acid Fast Bacilli stain (AFB stain), and PCR were not done in this present case. The patient was followed up for 1 year after the parotidectomy and the anti-tuberculosis chemotherapy: both clinical examination and ultrasound showed no recurrence of the lesions. Treatment was well-tolerated, and no adverse signs were noted.

## Discussion

Tuberculosis is common in developing countries but its incidence has been increasing in developed countries in recent years due to multiple factors such as the development of resistant strains and co-infection with HIV.[8] Tuberculosis often presents in the head and neck, with the cervical lymph nodes being one of the commonest sites of extrapulmonary tuberculosis,[1] but the salivary glands appear to be rarely infected.

The pathogenesis of parotid tuberculosis remains unclear. Two pathways have been described: the first one, via the oral cavity, as a mycobacterial infection ascends into the salivary gland via its duct or pass to its associated lymph nodes via lymphatic drainage. The second pathway involves hematogenous or lymphatic spread from a distant primary lung lesion.[4]

Clinical presentations of parotid gland tuberculosis are variable and nonspecific. It most commonly presents as a localized mass, resulting from infection of intracapsular or pericapsular lymph nodes. Nonetheless, it can mimic an acute sialadenitis with diffuse glandular enlargement. It may also present as a periauricular fistula or as an abscess.[4,9] But the typical clinical presentation of parotid gland tuberculosis is of a slow-growing, asymptomatic localized lesion within the gland. Constitutional symptoms are usually absent, therefore the mass is often mistaken for a neoplasm.[10] Radiological examination data (ultrasound, CT scan, and MRI) are generally inconclusive and a chest X-ray is requested in order to look for a possible primary focus.[9] There is no clinical, radiological, or biological evidence to support a diagnosis of parotid tuberculosis and pre-operative diagnosis remains challenging. In fact, in the majority of reported cases, it was based on histopathological examination of the surgically removed parotid mass, and these excised tissue specimens commonly were not sent for culture for mycobacterial tuberculosis.[4] So, the gold standard is still the direct microscopic examination and culture of bacteria. Fine needle aspiration with culture of the fluid may be useful. It is indicated for all parotid masses and should be performed to determine its nature, even though in the case of tuberculosis, it is not always contributory due to the presence of necrosis.[11] Core needle biopsy is still debatable as some authors suggest that it may result in cutaneous fistula formation.[12,13]

Currently, most authors recommend PCR gene amplification techniques after glandular cell culture to increase the positive cytopuncture results.[9] In literature, CBNAAT, AFB stain were mainly helpful in the diagnosis of bone and joint tuberculosis or lymph node tuberculosis.[14]

In our case, the pathological report made the positive diagnosis showing the presence of both lesions in the excised tumor.

The most common complications reported are: facial nerve damage, hemorrhage or hematoma formation, tumor track seeding, capsule rupture, and soft tissue infection, while there are no reports of poor wound healing.[6]

Tuberculosis infection of the parotid gland along with the existence of a benign parotid tumor is very rare, Warthin's tumor is the most frequent lesion found with 13 cases reported in the English literature[5] while only one other case of pleomorphic adenoma was described in recent literature.[4] As parotid tuberculosis can usually mimic malignant or benign tumors, it is important to differentiate both entities as the treatment differs. The diagnosis in most reported cases was based on histopathological examination of the surgically removed parotid mass, which showed co-existence of both lesions. Watanabe et al.,[15] and Ozcan et al.[16] confirmed the presence of mycobacterium tuberculosis within the tumor by PCR.

Anti-tuberculous treatment should be initiated early for an optimal recovery. The standard treatment consists of a quadritherapy: isoniazid (5 mg/kg/day), rifampicin (10 mg/kg/day), ethambutol (20 mg/kg/day), and pyrazinamide (25 mg/kg/day) for 2–4 months then isoniazid and rifampicin for 6–12 months.[11] Recently, other treatment protocols have been suggested: a regimen consisting of only three drugs (rifampicin, isoniazid, and pyrazinamide) followed by two drugs (rifampicin and isoniazid) as it is a pauci-bacillary extra pulmonary form of tuberculosis or a continuation phase of three drugs (rifampicin, isoniazid, and ethambutol).[17]

## Conclusion

Tuberculosis of the parotid gland is a rare disease. The clinical presentation is nonspecific, mimicking in most cases any other tumor pathology. Pre-operative diagnosis is still challenging, thus the importance of Fine needle aspirations and biopsies. Therapeutic management is based on a long-term antituberculosis treatment.

Therefore, in endemic countries and immunodeficient cases, this diagnosis should always be kept in mind as a differential diagnosis.

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### Author contributions

D.C.: conceptualization, validation, methodology, writing – original draft preparation; N.R.: conceptualization, validation, writing – original draft preparation; E.J.: conceptualization, investigation, visualization, methodology, writing – original draft preparation; E.R.: conceptualization, methodology; F.K.: conceptualization, resources, validation, visualization; R.J.: conceptualization, resources, validation, visualization; I.Z.: conceptualization, investigation, methodology; S.N.: conceptualization, writing – original draft preparation; I.H.: conceptualization, writing – review & editing; C.M.: validation, writing – original draft preparation.

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## Ethics approval

Our institution does not require ethical approval for reporting individual cases or case reports.

## Informed consent

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