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# Tuberculosis of the parotid gland

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## ÖZET

### *Parotis bezi tüberkülozu*

*Tüberkülozun izole parotis kitlesi olarak görülmesi nadirdir. Preoperatif tanı güçtür ve semptomatoloji nonspesifiktir. Hastaların çoğunda önceden bir parotis tümörü öyküsü vardır, sıklıkla pleomorfik adenomdur. Bu yazıda, sağda parotiste 6 aydır şişlik olan 35 yaşında bir kadın hastayı sunuyoruz. Kitle sert, hassasiyet olmayan, ipsilateral servikal lenfadenopatinin eşlik etmediği parotis malignitesini düşündürür özellikteydi. Bilgisayarlı tomografi pleomorfik adenoma benzeyen parotis içi tümörü gösterdi ve hastaya yüzeysel parotidektomi uygulandı. İnce iğne aspirasyon biyopsisi yapıldı, ancak tanısal değildi. Histolojik incelemede intraparotideal lenf bezinde sarkoidoz veya tüberkülozu düşündüren granülomatöz değişiklikler saptandı. Ziehl-Nielsen boyası negatifken, tüberkülin deri testi (PPD, 5 IU) pozitif. Hastaya 2 ay izoniazid, rifampin, pirazinamid ve etambutol ile başlangıç tedavisi verildi, ardından 7 ay süreyle izoniazid ve rifampin ile idame yapıldı. Postoperatif olarak gelişen hafif fasiyal sinir parezisi 1 hafta sonra düzeldi. Parotis tüberkülozu nadirdir, ancak parotis kitlelerinin ayırıcı tanısında dikkate alınmalıdır. İnce iğne aspirasyon biyopsisi tanı için önemlidir, çünkü bu durumda tedavi yaklaşımı esas olarak konservatif olacaktır. Cerrahi özellikle diğer tanısal incelemeler başarısız olduğunda hem tanısal hem de terapötik olabilir.*

**Anahtar Kelimeler:** Tüberküloz, parotis bezi

## SUMMARY

### *Tuberculosis of the parotid gland*

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*Tuberculosis (Tbc) presented as an isolated parotid mass is rare. Preoperative diagnosis is difficult and the symptomatology is nonspecific. In the majority of the cases an initial diagnosis of a parotid tumor, often a pleomorphic adenoma, is made. We present a 35-year old woman with a six months duration right parotid lump. The mass was firm and nontender without ipsilateral cervical lymphadenopathy, suggesting a parotid neoplasm. The computerized tomography scan showed an intraparotideal tumor resembling a pleomorphic adenoma and thus the patient underwent to a superficial parotidectomy. Fine needle aspiration biopsy was performed but it was not diagnostic. Histological examination revealed an intraparotideal lymph node with changes of granulomatous lymphadenopathy type, like those demonstrated in the tuberculosis and sarcoidosis. Ziehl-Nielsen staining was negative, while the tuberculin skin test (PPD, 5 IU) was positive. The patient's treatment regimen consisted of a 2-month initial phase of isoniazid, rifampin, pyrazinamide and ethambutol followed by a 7 month continuation phase of isoniazid and rifampin. Postoperatively, there was only a mild paresis of the facial nerve resolved a week after. Parotid Tbc is very rare but should be considered as a differential diagnosis of parotid lumps. Fine needle aspiration biopsy (FNAB) is of utmost importance for diagnosis, since the treatment of this entity is primarily conservative. However, surgery could be both therapeutic and diagnostic, especially when other diagnostic examinations fail.*

**Key Words:** Tuberculosis, parotid gland.

*Mycobacterium tuberculosis* infects one third of the world's population and causes 8 million new cases of tuberculosis (Tbc) and approximately 2 million deaths each year (1). TB can affect virtually every organ, importantly the lungs. Extrapulmonary Tbc comprises approximately 20% of the overall active Tbc (1). Mycobacterial disease involves many tissues within head and neck area, especially cervical lymph nodes and larynx. Tbc of the parotid gland is quite unusual, firstly described by von Stubenrauch in 1894 and De Paoli in 1896 (2,3). Tbc comprises of 2.5 to 10% of parotid pathology and may directly involve the parenchyma of the gland or the intraparotid and periglandular nodes (2,3). Clinically, it can be presented as a common parotid inflammation or more frequently as a slow growing parotid lump resembling a neoplasm. As a result, diagnosis often can be missed or delayed and usually made after surgical excision. We present a single case and discuss our experience with this rare clinical entity.

### CASE REPORT

A 35-year old woman from Albania was presented at the ENT department of the University Hospital of Patras, Greece, because of a painless lump in the right parotid gland which had

been presented 3 months ago. No other symptoms or complaints such as, fever chills or night sweats were reported by the patient. There was no history of exposure to cats, farm animals or risk factors for human immunodeficiency virus (HIV) infection. She had no medical history and was normal on physical examination. All laboratory examinations were within the normal limits. The level of angiotensin-converting enzyme was normal. Tests for antinuclear antibodies, rheumatoid factor and anti-HIV antibodies were negative.

Computerized tomography scan of the parotid region revealed a circumscribed solid mass with maximum diameter 1.8 cm, strongly suggesting a benign parotid tumor like pleomorphic adenoma (Figure 1). Fine-needle aspiration biopsy (FNAB) of this mass failed to make a definitive diagnosis because of the small number of aspirated cells. However, there were not any malignant cells in the examined specimen. Consequently, the patient underwent a right superficial parotidectomy without intraoperative complications. The mass didn't display any specific macroscopic characteristics to outdraw a possible diagnosis. Pathologic examination revealed the presence of an intraparotideal lymph node showing changes of granulomatous

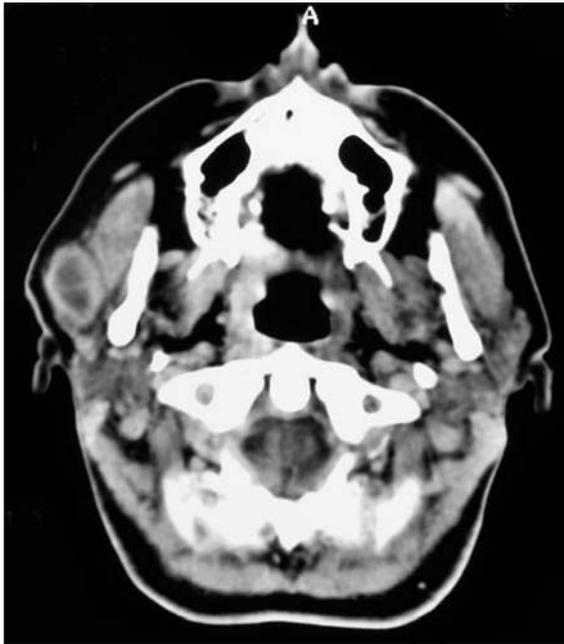


Figure 1. CT-scanning demonstrating a right parotid mass.

lymphadenopathy. More specifically, sections from the lymph node, showed the presence of multiple epithelioid granulomas without associated caseous necrosis (Figure 2). A Ziehl-Nielsen stain was also performed but did not demonstrate any acid fast bacilli. Thus, pathologic differential diagnosis included tuberculosis and sarcoidosis. A PPD skin testing was performed which was positive (20 mm). The chest X-ray

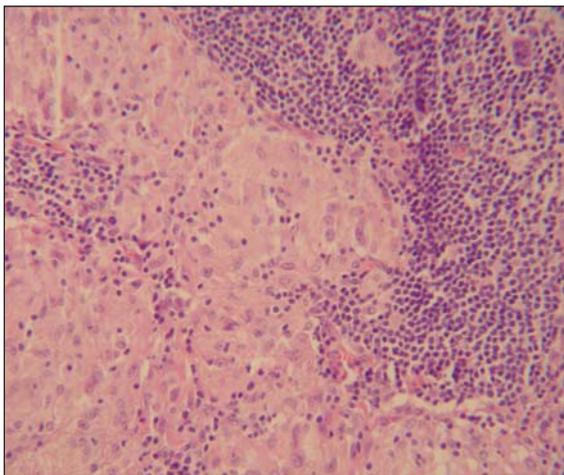


Figure 2. Photomicrograph showing the presence of multiple epithelioid granulomas within the lymph node (HE, x400). Note the absence of caseous necrosis.

was negative for pulmonary lesions of Tbc. We decided to treat the patient with the classical anti-Tbc therapeutic regimen consisting of isoniazid 300 mg/day, rifampin 600 mg/day, ethambutol 1200 mg/day and pyrazinamide 1500 mg/day. All drugs were given for 2 months, followed by a 7 month continuation phase of isoniazid and rifampin.

The patient had an uneventful postoperative recovery except from a mild paresis of the right marginal mandibular branch of the facial nerve, but the patient recovered a week later without any sequelae. She was discharged eight days after surgery and since then she followed up regularly in outpatient clinics of our hospital.

### DISCUSSION

During the 20<sup>th</sup> century it was noted a significant worldwide decrease in the number of Tbc patients. However, the last two decades the incidence of the disease has been raised especially in the countries of the third world and in a lesser degree in the western societies. The latter could be attributed to a large number of immigrants from poorer counties, but also to an increased number of HIV immunocompromised patients. The incidence of extrapulmonary Tbc has also increased in the same rate as the pulmonary disease. Tbc in the head and neck region represents approximately 15% of all mycobacterial infections, representing the commonest extrapulmonary manifestation of the disease. However, Tbc of parotid gland is extremely rare entity even in countries where the disease still remains endemic.

Even if the pathogenesis of Tbc of the parotid remains unclear, two different modes of development have been suggested regarding this issue. According to the first hypothesis, mucobacteria can reach parotid parenchyma and/or lymph nodes by autoinoculation with infected sputum or from an infection of the teeth or tonsils, via duct or afferent lymphatics. The second hypothesis suggests an infection from the lungs via a haematogenous or lymphatic route (4).

*M. tuberculosis* is the commonest pathogen in adults, while atypical mycobacteria are detected

usually in child (5). The distinction between them can be done by the use of PPD skin testing, acid alcohol fast stain of the saliva and appropriate cultures (6). However, stains and cultures are sometimes negative, even in those patients with positive PPD test and histologic confirmation (2). When the disease is presented as a diffuse parotid inflammation PPD test is weakly positive and the cause is usually atypical mycobacteria. On the contrary, when parotid lymph nodes are affected, tuberculin test is strongly positive and this type of presentation suggests Tbc from *M. tuberculosis* possibly emerging from another primary site (7). Cantrell et al, claimed that, due to a hypersensitivity reaction, the causative bacilli remain trapped in scar tissue for a long time before reactivated to cause an extrapulmonary form of Tbc, even if the primary focus cannot be seen (8).

The majority of the reported cases presented mainly as hard and nodular unilateral masses with variable degree of fixation and slow growth over a 3 to 6 months period, but there are also cases of gradual increase of up to 10 years (9). These lumps when firstly seen often considered to be parotid tumors and the correct diagnosis of Tbc is usually made only after surgical excision and histologic examination. Other conditions that have to be differentiated are malignant lymphoma, chronic lymphadenopathy, sialosis, Sjogren's syndrome and acute or chronic suppurative parotitis (10). Imaging studies and FNAB are often inconclusive and there is no any single definitive diagnostic examination. Thus, surgical intervention becomes necessary when other investigations are non-contributory. However, even if diagnosis is still based mainly on tissue specimens after resection of the affected gland, one should be very careful because of the possible risks of such an operation, like injuring the branches of the facial nerve or causing a local fistula. Besides, with the exception of large necrotic masses, abscess formation or non-responsive cases which could be considered indicative for surgery, Tbc of the parotid is mainly a medically curable disease with the appropriate anti-tuberculous medication (6). Recently, a combination of FNAB and

PCR (polymerase chain reaction) using fine-needle aspirates gives promising results and would be useful diagnostically, in cases with high index of suspicion, avoiding a surgical intervention (11).

Typical histological findings of Tbc consisted of epithelioid granulomas with central caseous necrosis. Sometimes, changes of acute inflammation may also be seen, especially in early stages of the disease, making differential diagnosis from other causes of inflammation quite difficult (6). Furthermore, acid-fast bacilli (AFB) may not be demonstrated in biopsy specimens, probably because of low bacillus concentrations in extrapulmonary lesions of Tbc. In such cases, with negative diagnostic tests but high index of clinical suspicion, one should start medical treatment to evaluate the response of the patient to the anti-tuberculous regimen.

In our patient, the pathology report was suggestive of Tbc but not diagnostic. There was absence of the typical caseous necrosis in the epithelioid granulomas and the diagnosis based mainly upon the strongly positive PPD test for mycobacterium and a high index of clinical suspicion. Based on that, we administered the classic anti-Tbc therapy to the patient.

Early administration of the appropriate regimen is a prerequisite for a successful recovery without complications. Isoniazid, rifampin, ethambutol and pyrazinamide are still used as first-line treatment, providing satisfactory results. Usually, a totally 9-months period of administration of isoniazid and rifampin is sufficient for extrapulmonary Tbc treatment (12,13).

This case depicts that a high index of suspicion is needed for the diagnosis of Tbc in a recent parotid lump, even if the chest X-ray is negative for pulmonary Tbc, especially nowadays because of the large number of immigrants from endemic countries and the increased number of HIV immunocompromised patients. Thus, one could avoid an operation and possible complications in a medically treated condition.

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