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Primary Sinonasal Tuberculosis: A Diagnostic Challenge

Primer Sinonazal Tüberküloz: Tanısal Bir Güçlük

Case Report
Olgu Sunumu

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Abstract

Primary sinonasal tuberculosis remains a diagnostic challenge as it mimics sinonasal granulomatous disease or neoplasms. Its characteristic signs and symptoms may be non-specific and highly variable. Here we report a unique clinical condition of a 24-year-old man who presented with unilateral nasal obstruction and epiphora for 2 years. He was without any comorbidity, was immunocompetent, and was otherwise healthy. Subsequent investigations, including a computed tomography scan of the paranasal sinuses, and perioperative findings revealed a sinonasal mass involving the lateral nasal wall and paranasal sinuses. The histopathology was consistent with the features of tuberculosis. There was no evidence of pulmonary or any other primary tubercular foci elsewhere in the body, suggesting the diagnosis of primary sinonasal tuberculosis.

Keywords: Sinonasal tuberculosis, extrapulmonary tuberculosis, diagnosis, endoscopic debridement

Öz

Primer sinonazal tüberküloz, sinonazal granülomatöz hastalık veya neoplazileri taklit ederek tanısal güçlük oluşturur. Karakteristik semptom ve bulguları spesifik olmayıp, oldukça değişken olabilir. Burada, tek taraflı burun tıkanıklığı ve iki yıldır süren epifora yakınmaları ile başvuran 24 yaşında bir erkek hastayı, ilginç klinik tablosuyla sunduk. İmmünokompetan hastanın herhangi bir komorbiditesi ya da sağlık sorunu yoktu. Paranazal sinüslerin bilgisayarlı tomografi görüntülemesini de içeren sonraki incelemeler ve operasyon bulguları lateral nazar duvar ve paranazal sinüsleri ilgilendiren sinonazal bir kitleyi ortaya koydu. Histopatoloji tüberküloz özellikleri ile uyumluydu. Pulmoner ya da vücudun başka bölgelerinde primer odak bulunmasına yönelik kanıt olmaması, primer sinonazal tüberküloz tanısını ortaya koydu.

Anahtar kelimeler: Sinonazal tüberküloz, ekstrapulmoner tüberküloz, tanı, endoskopik debridman

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Introduction

Primary sinonasal tuberculosis is rarely encountered even in developing and endemic countries, and it is usually secondary to pre-existing foci elsewhere in the body. The mechanical protection offered by the unidirectional ciliary clearance of the mucosal conveyer belt and the effective bactericidal properties of nasal secretions can explain why this entity is not a usual diagnosis in routine clinical practice.

The first patient with nasal tuberculosis was documented by the Italian anatomist Giovanni Battista Morgagni (1) who studied the impact of tuberculosis in the human body. In 1761, he reported ulceration of the nose, soft palate, and nasopharynx during an autopsy of a young man with pulmonary tuberculosis. According to Sim and Crowther (2), Clarke (3) presented the first case of primary nasal tuberculosis to the Pathological Society of London in 1876. In 1971, Messervy (4) reported another patient with primary nasal tuberculosis, which mimicked a malignant granuloma. Reviewing English-language medical literature spanning 95 years, Butt (5) identified only 35 patients with primary nasal tuberculosis. Our PubMed/MEDLINE-indexed English-language literature review since 2000 yielded 38 results corroborating this diagnosis.

In this report, we present the case of a young patient with primary sinonasal tuberculosis and attempt to describe the clinical presentation, possible etiopathogenesis, and management aspects of this rare disease.

Case Presentation

A 24-year-old man presented with right-sided nasal obstruction, occasional mucoid nasal discharge, and watering of the right eye, which had been occurring for two years. There was no specific history of known allergy; his symptoms did not clinically corroborate with those of allergic rhinitis. He was non-diabetic, not addicted to any substances, and otherwise healthy. His serology profile was non-reactive.

A preliminary clinical evaluation by anterior rhinoscopy revealed greyish polypoid changes in the right middle turbinate and meatus, covered with mucoid discharge, with left-sided septal deviation. Subsequent diagnostic nasal endoscopy further revealed a proliferative mass in the right middle meatus that bled on touching (Figure 1). The left nasal cavity was unremarkable. Contrast-enhanced computed tomography (CT) revealed a heterogeneous soft tissue lesion with hyperdense areas involving the right nasal cavity and all ipsilateral sinuses (Figure 2). The lamina papyracea was pushed by the tissue bulk, apparently encroaching the medial extraconal space. The soft tissue lesion seemed to involve the left anterior ethmoid cells as well. The right maxillary sinus was hypoplastic, with a widened ostium and ostiomeatal complex. The clinicoradiologic spectrum suggested the following differential diagnoses in preferential order: fungal rhinosinusitis, granulomatous disease, and neoplasia. A subsequent histopathologic examination from the tissue obtained during diagnostic endoscopy suggested a benign inflammatory polyp, without any fungal elements, granulation, or evidence of malignancy. Lacrimal syringing showed no regurgitation from the punctae, and visual acuity was normal in refraction tests. The patient was prepared for endoscopic sinus surgery.

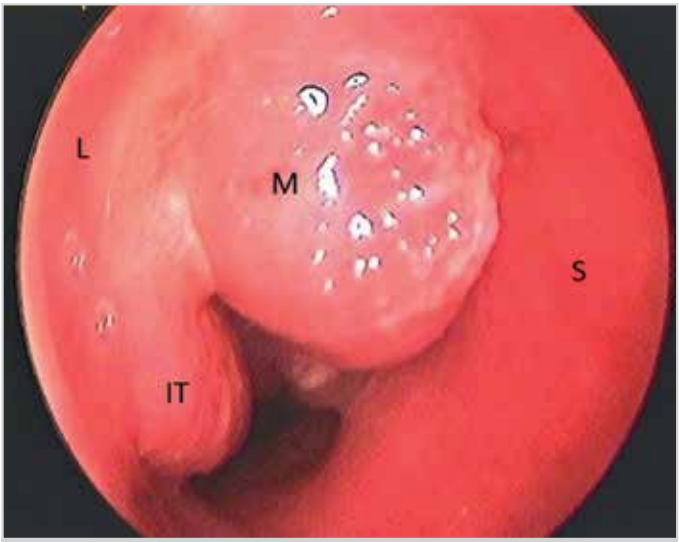


Figure 1. Diagnostic nasal endoscopy showing a proliferative mass lesion in the right middle meatus
L: lateral nasal wall; IT: inferior turbinate; S: septum; M: mass

Routine investigations as a part of the pre-operative work-up, including hematology results and chest X-ray, were unremarkable, except for the erythrocyte sedimentation rate (45 mm/1st hour). During step-by-step endoscopic surgery, the sinonasal mass in the right nasal cavity was found to involve the maxillary sinus, ethmoids, the frontal and sphenoid sinuses, with firm attachment to the lamina papyracea (Figure 3). As the lesion was debulked with a microdebrider (Stryker; Michigan, USA), large middle meatal antrostomy and fronto-ethmoid sphenoidotomy were performed. The mucopus was sent for fungal culture, although no fungal debris/allergic mucin could be delineated at surgery. The middle turbinate was bolgerized to facilitate ventilation and prevent lateralization of the turbinate and resultant recurrent sinusitis.

The specimen in the histopathological analysis revealed Langhans giant cells and epithelioid cell granulomata, which was consistent with tuberculosis (Figure 4). No features of dysplasia or malignancy were noted. The Ziehl–Neelsen stain for acid fast

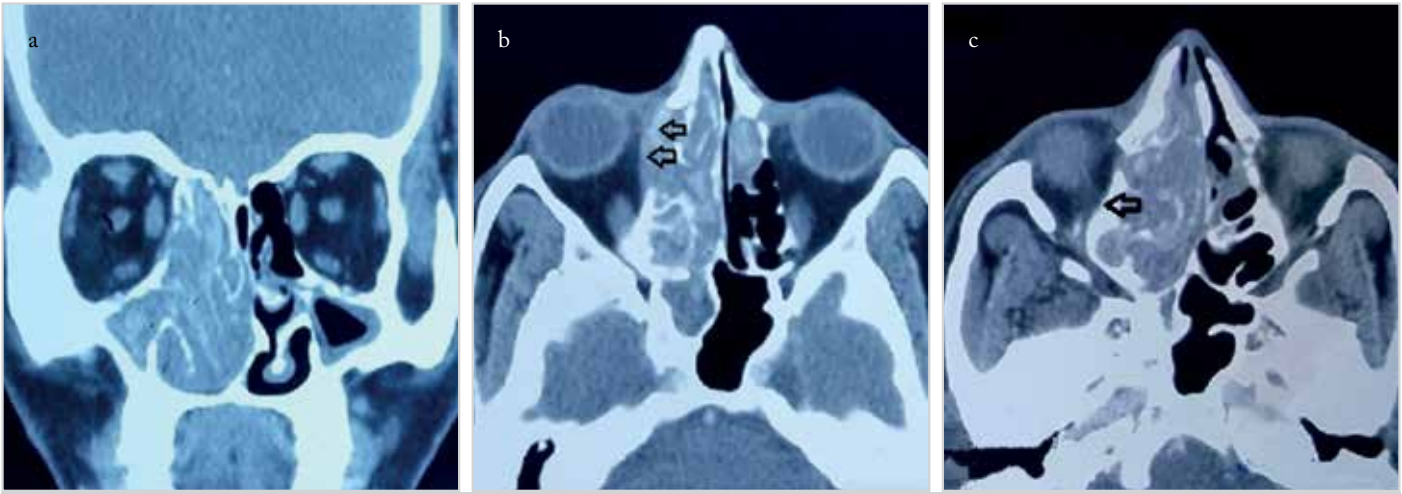
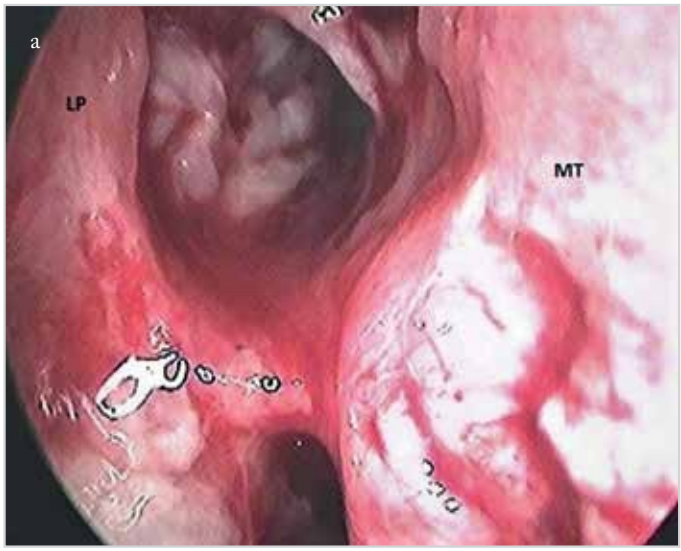


Figure 2. a-c. Coronal (a) and axial (b, c) contrast-enhanced CT scan revealing a heterogeneous soft tissue lesion with hyperdense areas involving the right nasal cavity and all ipsilateral sinuses; b, c. The axial cuts showing the lamina papyracea pushed by the tissue bulk that appeared to encroach the medial extraconal space (arrows)



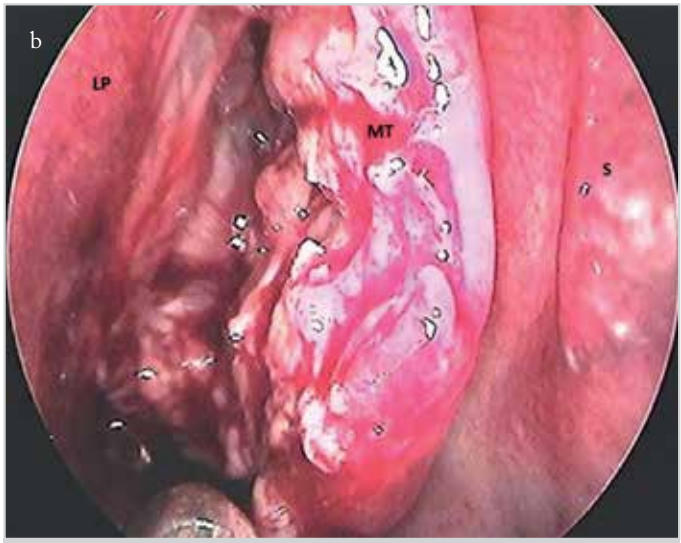


Figure 3. a, b. During surgery, the sinonasal mass in the right nasal cavity, that involved multiple paranasal sinuses, was found to be in proximity with the lamina papyracea
LP: lamina papyracea; MT: middle turbinate; S: septum

bacilli was negative. Fungal culture revealed no growth after two weeks of incubation in Saboraud's agar medium.

Subsequent investigations directed at establishing the systemic presence of tuberculosis or existence of a primary focus, including induced sputum for acid fast bacilli, bronchoalveolar lavage analysis, and blood for the *Mycobacteria* deoxyribonucleic acid polymerase chain reaction (DNA PCR), were negative. Considering the available evidences at hand, the diagnosis of primary sinonasal tuberculosis was made.

The patient was started on anti-tuberculosis drugs for six months (category 1). During the course of treatment, he was put on regular follow-up with scheduled endoscopy sessions. At four-months, endoscopy along with a CT scan of the nose and paranasal sinuses showed no residual mass lesion, with only few areas of the polypoid tissue replacing the healthy sinus mucosa. The patient continued to be disease-free at the latest follow-up, approximately six months after completion of the anti-tuberculosis regimen.

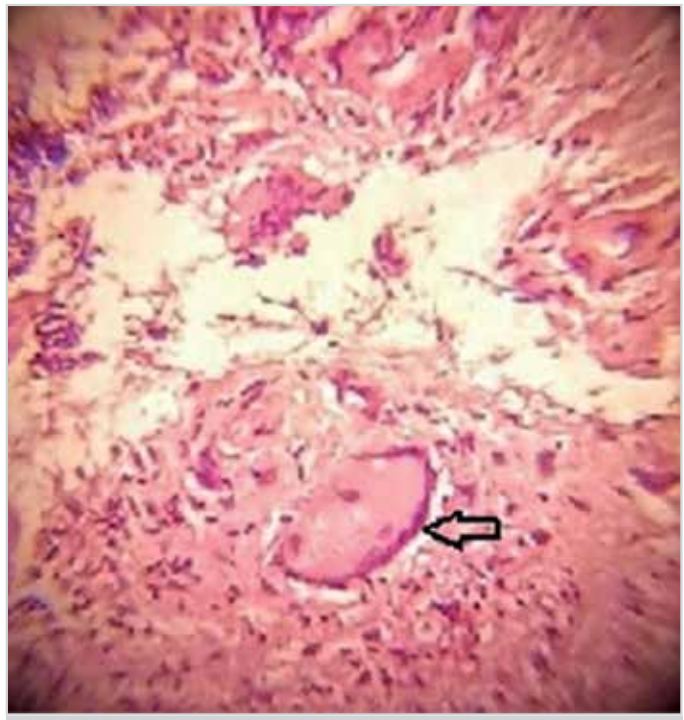


Figure 4. Histopathologic examination revealing Langhans giant cells and epithelioid cell granulomata (arrow). Hematoxylin-eosin X100

Prior to writing up this report, informed consent in written was duly obtained from the patient.

Discussion

It has been observed that although the extrapulmonary form occurs secondarily in only 15% of tuberculosis patients (6), primary tuberculosis of the nose and paranasal sinuses is indeed rare. In the upper respiratory tract, the nose and paranasal sinuses is the most resistant anatomic region to tuberculous invasion, owing to its unique epithelial architecture and bactericidal secretions. However, it can still get affected by infected particle inhalation, the immunodeficiency status, traumatic inoculation, or a combination of these.

Primary nasal tuberculosis is twice as common in women as in men, the median age being 40 years (6), and it occurs more commonly in those in the lower socio-economic strata. It usually presents unilaterally, but in one-third of the patients, it can be bilateral. The lesion can be proliferative, infiltrative, or ulcerative. It usually affects the nasal septum first, followed by the anterior segment of inferior turbinates, paranasal sinuses, choana, nasopharynx, orbit, and cranial cavity, not necessarily in that order (7). Most patients present with nasal obstruction, blood-stained nasal discharge, epistaxis, crusting, pain, dryness of the nose and throat, epiphora, post-nasal discharge, and blurred vision (if there is orbital involvement) (1, 8, 9, 10).

The diagnosis of primary sinonasal tuberculosis is challenging because it is seldom encountered in routine otolaryngology practice; therefore, it demands a high index of suspicion. Furthermore, as we have encountered in our patient, it might

often mimic, from the clinicoradiologic perspective, fungal rhinosinusitis, sinonasal neoplasms (including inverted papilloma, natural killer T cell lymphoma), other granulomatous diseases (such as Wegner's granulomatosis, syphilis, leprosy, sarcoidosis, rhinoscleroma, rhinitis sicca), and retained foreign bodies (8). Beltran et al. (11) proposed that the diagnosis of sinonasal tuberculosis should be based on the following criteria: a) absence of clinical response to empirical antibiotics, b) presence of caseous granulomatous inflammatory lesions in the histopathological analysis, and c) identification of *Mycobacterium tuberculosis* in the surgical specimen. It is true that patients with sinonasal tuberculosis often present with considerable clinical variations. Its most common and remarkable presentation is lupus vulgaris, while it may even present as an apparently innocuous non-ulcerated form mimicking septal swelling (12, 13).

As we have seen in our patient, sinonasal tuberculosis can be potentially misdiagnosed as relatively innocuous lesions such as polyposis and fungal rhinosinusitis; thus, it is understandable to administer empirical antibiotics inadvertently as the initial step of treatment, which can indirectly establish sinonasal tuberculosis as the diagnosis of exclusion. However, to save precious time for the diagnosis and, most importantly, to discourage an indiscriminate use of antibiotics, an initial biopsy in a patient with a strong clinical suspicion might be considered worthwhile, particularly in endemic regions. Furthermore, from our present and previous experiences, we believe that for establishing the diagnosis of primary sinonasal tuberculosis, emphasis must be placed on investigations aimed at the systemic search for any other primary tuberculous focus as the possible source of dissemination; if it turns out negative, that perhaps should strongly vouch for the most valuable diagnostic criterion.

Imaging helps in the assessment of the extent of the disease and planning surgery. In our patient, the findings of the contrast-enhanced CT scan of the nose and paranasal sinuses led us to presume the lesion to be of fungal or neoplastic origin. However, subsequent tissue biopsies revealed Langhans giant cells and epithelioid cell granulomata, which were consistent with the histopathologic evidence of tuberculosis, although acid fast bacilli were absent when Ziehl–Neelsen staining of the tissues was performed. On the other hand, we failed to demonstrate a possible primary source and resultant systemic dissemination when bronchoalveolar lavage analysis results and blood for *Mycobacteria* DNA PCR were unremarkable.

The cornerstone for the treatment of primary sinonasal tuberculosis, like that in any of its forms, is multi-drug anti-tuberculosis therapy for a minimum of six months. Ancillary procedures performed before the initiation of an anti-tuberculosis regimen might include surgical debulking of the mass lesion. However, the decision to receive anti-tuberculosis drug therapy or undergo prior surgical debulking in case of evident mass lesions remains highly individualized and is often chiefly determined by the lesion's severity and extent, any associated emergencies, the need to establish the tissue diagnosis due to a high clinical suspicion, and when the clinical presentations are misleading enough with several possible differential diagnoses, as in our patient. It should be noted that for the extrapulmonary form, apart from few patients who need an anti-tuberculosis regimen to be administered solely on the basis of a definitive and strong clinical suspicion, histologic or at least cytologic evidence is required to establish a diagnosis, which might necessitate minor surgical intervention prior to prescribing anti-tuberculosis drugs. In our patient, the clinical presentation was deceptive, as the final diagnosis was not something we expected. Here the diagnostic biopsy might not have been representative as it failed to show the characteristic histologic evidence of tuberculosis, and in the process, it made us think that the disorder had another etiology. The patient was thus subjected to thorough debridement of the sinonasal mass, which, although could have been avoided, was ultimately proved to be helpful on two aspects—it clinched the histologic evidence for tuberculosis, and it addressed the immediate pressure effects of the lesion. Therefore, the importance of tissue diagnosis for tuberculosis was re-instated, which might include relatively more invasive surgical interventions, particularly in patients presenting with clinically misleading features.

Conclusion

Sinonasal tuberculosis is a seldom-encountered clinical disorder. Owing to its rarity and often atypical presentation, a high index of suspicion is required to diagnose and treat it. This report describes how an immunocompetent patient, with no primary focus of tuberculosis and presenting with a sinonasal mass lesion and with clinical features suggestive of fungal rhinosinusitis or a neoplasm, was ultimately found to be suffering from tuberculosis following an extensive debridement procedure. The many forms of extrapulmonary primary tuberculosis mimicking clinical entities that are relatively more common in a given anatomic region must be kept in mind by clinicians with a low threshold of suspicion, particularly in endemic regions and susceptible populations.

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