



Case Report

Primary tuberculosis of the pyriform sinus: A case report

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ABSTRACT

Introduction: Tuberculosis is a chronic bacterial infection caused by the *Mycobacterium tuberculosis*. Extrapulmonary tuberculosis (EPTB) accounts for about 20% of cases in immunocompetent patients and 50% of cases in HIV-positive individuals (7). Except for the cervical lymph nodes location, head and neck tuberculosis is rare. **Clinical presentation:** A 32-year-old man presented with chronic and progressive sore throat and dysphagia lasting for 4 months. The clinical examination revealed a lesion in the left pyriform sinus confirmed by CT. A biopsy was performed and the histological results showed a chronic and granulomatous inflammatory process composed of epithelioid and giant-cellular follicles centered by large areas of caseous necrosis, concluding tuberculosis of the pyriform sinus.

Discussion: Primary tuberculosis of the hypopharynx is very rare. In most cases, it is revealed by odynophagia, dysphagia, but it can also mimic the signs of a malignant tumor, hence, the difficulty in diagnosing this localization. Few articles have been reporting cases of hypopharyngeal tuberculosis, in these articles, the main symptom was dysphagia, as was the case with our patient. Given the rarity of hypopharyngeal tuberculosis, several differential diagnoses are to consider: squamous cell carcinoma, which has a similar clinical manifestation to tuberculosis. There is also, rarely, oropharyngeal lymphoma, minor salivary gland tumors, neurogenic tumors, and Wegener's disease. The diagnosis of EPT is established when *Mycobacterium tuberculosis* bacillus is isolated and the epithelioid and giant-cellular follicles centered by large areas of caseous necrosis is found in the histopathological examination of the endoscopic biopsy specimen. The treatment is medical, by anti-bacillary chemotherapy, however the period is longer than pulmonary tuberculosis, it extends from six to twelve months.

Through this study, the main lessons are that malignant lesion is always to rule out when a suspicious lesion of the hypopharynx is found, we also come to conclude that the endoscopic and clinical aspect between tumor and tuberculosis of the hypopharynx are very similar. The diagnosis is confirmed with biopsy of the lesion.

Conclusion: primary tuberculosis of the pyriform sinus is very rare. It's mandatory to rule out malignant pharyngeal lesions and consider hypopharyngeal tuberculosis as one of the differential diagnoses in lesions especially in the countries where tuberculosis is endemic.

1. Introduction (9)

Tuberculosis is a chronic bacterial infection caused by the *Mycobacterium tuberculosis*. Extrapulmonary tuberculosis (EPTB) accounts for about 20% of cases in immunocompetent patients and 50% of cases in HIV-positive individuals [7]. Except for the cervical lymph nodes location, head and neck tuberculosis is rare. We report a clinical case of tuberculosis of exceptional localization: the pyriform sinus, mimicking a malignant tumor in a 32-year-old man.

1.1. Case report

A 32-year-old man presented with a sore throat and dysphagia persisting for four months, the patient also reported weight loss of 10 kg over this period of time, nevertheless, no night sweating nor fever were present. The patient had no personal significant features, especially no history of smoking, alcohol consumption or respiratory tract infections, allergies or discontinued medication. No inheritable conditions or health problems were found in the family history. The physical examination with endonasal endoscopy, showed a budding lesion in the left

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piriform sinus (Fig. 1), the mucosa of the mass is inflammatory, and some infra-centimetric ulcerations were also found. No cervical adenopathies were found, and the rest of the ENT examination was normal. The blood examination revealed a hemoglobin level of 10,3g/dl, a white blood cell count of 7650 cell/mm³, and serum; C-reactive protein level of 35,6mg/l. The patient was HIV seronegative.

Computed Tomography of the neck and chest revealed a well-defined mass of 3 × 2cm in the left pyriform sinus, near the supraglottic area, (Fig. 2). Given the clinical presentation, a malignant lesion is the first and urgent diagnosis suspected, so under general anesthesia, a pan-endoscopy was performed by a third year ENT intern under the supervision of the chef of the head and neck department un the university hospital, it consisted on performing a direct laryngoscopy to examine the upper airways, vocal cords and subglottic region, the examination of the base of the tongue and the tonsils region and an esophagoscopy that confirmed, after the aspiration of salivary secretions, the presence of a solitary ulcero-budding lesion localized int the pyriform sinus, no esophageal extension was noticed. The biopsy of the left pyriform sinus was realized at the end of the procedure, to prevent any bleeding that can disturb a good exploration of the aerodigestive tract. The histological results showed a chronic and granulomatous inflammatory process composed of epithelioid and giganto-cellular follicles centered by large areas of caseous necrosis. The diagnosis of tuberculosis of the pyriform sinus was then retained. The direct examination of the patient's sputum found no mycobacterium tuberculosis and the CT scan of the Chest found no suspicious lesions.

After discussion with the regional anti-tuberculosis department, the decision was to start an exclusive medical treatment with Anti bacillar chemotherapy. The protocol consisted on d two months association of pyrazinamide 30mg/kg/day and ethambutol 20mg/kg/day, followed by 6 months of rifampicin 10mg/kg/day and isoniazid 3mg/kg/day, no surgical intervention is indicated with a monthly surveillance in ENT consultation with clinical examination including endonasal fibroscopy.

After 7 months of treatment, a total response the to the Anti bacillar treatment was retained, the patient showed a clinical Improvement, he gained 10 kg of weight, a complete regression of the pharyngeal lesion was confirmed in the fibrosopic examination (Fig. 3: normal mucosa of the left pyriform sinus, no medical allergies or resistance were noticed.

At one year follow up, the patient maintained a full recovery.

2. Discussion

Primary tuberculosis of the pyriform sinus is very rare. It occurs in less than 1% of patients with head and neck tuberculosis infection, usually the Palatine tonsils are the organs frequently concerned [2]. The age prevalence is 39–60 years old(2,3). The symptoms include odynophagia and dysphagia [3–5], but also, it can be revealed by neck mass with abscess or mimicking a malignant tumor of the neck, like it was



Fig. 1. Endoscopic view of the left pyriform sinus lesion.

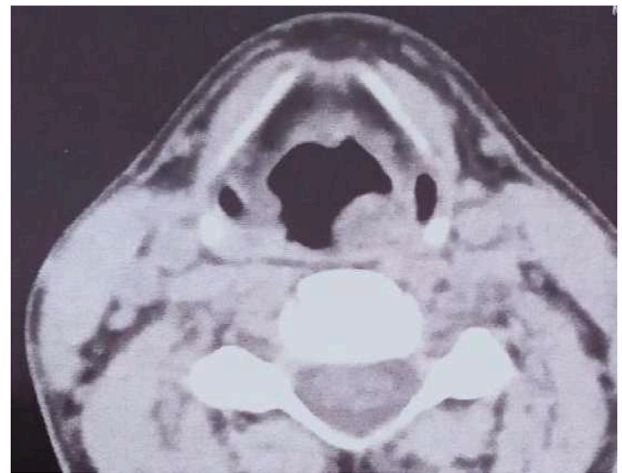


Fig. 2. Axial slice of CT can of the neck:well defined mass of the left pyriform sinus.



Fig. 3. Endoscopic view of the left pyriform sinus after 7 months of treatment.

reported in the literature [8,9], thus the difficulty in diagnosing this localization [1,2].

Hematogenous and lymphatic routes are most likely to be involved the mycobacterium tuberculosis spread, especially that no primary pulmonary involvement was found in the literature, as it was also the case with our patient [1]. A notoriously difficult dilemma is to differentiate primary tuberculosis of the pyriform sinus and carcinoma, as the clinical presentation is grossly similar. There is also, rarely, oropharyngeal lymphoma, minor salivary gland tumors, neurogenic tumors, and Wegener's disease.

The diagnosis of the hypopharyngeal tuberculosis, even though challenging, it includes an endoscopic biopsy with The typical caseating granulomas and acid-fast bacilli (AFB) stained by Ziehl-Neelsen. A positive TB culture remains the gold standard, but diagnosis may be delayed (3–8 weeks) [1,7].

The imaging appearance of extranodal Tuberculosis of the head and neck is a nonspecific soft-tissue mass. In advanced cases, an abscess can develop, the can be referred to as cold abscess because it develop slowly so that there is little surrounding inflammation [6,8]. The presence of necrosis, rim enhancement, calcifications, nodal disease, and pulmonary findings may orient toward the diagnosis of tuberculosis, that should be confirmed with biopsies and microbiologic studies.

Treatment includes isoniazid combined with of two or three anti-bacillars, rifampin, ethambutol, and pyrazinamide. However the period

is longer the pulmonary tuberculosis, it extends from of six to twelve months. Pyrazinamide and ethambutol are recommended. for the first two months treatment.

The clinical improvement, starts in the first two weeks of treatment, as well as decreasing the contagious risk of the micro-organism [1]. However, sinus pyriform tuberculosis as it's the case of the rest of the Extrapulmonary tuberculosis requires a longer period of therapy. At least, twelve months to control the infection and to prevent its recurrence.

3. Conclusion

Primary tuberculosis of the pyriform sinus is very rare. It's mandatory to rule out malignant pharyngeal lesions and consider hypo-pharyngeal tuberculosis as one of the differential diagnoses in lesions especially in the countries where tuberculosis is still endemic.

Ethical approval

Hereby, I Safaa Touihmi consciously assure that for the manuscript Primary tuberculosis of the pyriform sinus: case report, the following is fulfilled:

- 1) This material is the authors' own original work, which has not been previously published elsewhere.
- 2) The paper is not currently being considered for publication elsewhere.
- 3) The paper reflects the authors' own research and analysis in a truthful and complete manner.
- 4) The paper properly credits the meaningful contributions of co-authors and co-researchers.
- 5) The results are appropriately placed in the context of prior and existing research.
- 6) All sources used are properly disclosed (correct citation). Literally copying of text must be indicated as such by using quotation marks and giving proper reference.
- 7) All authors have been personally and actively involved in substantial work leading to the paper, and will take public responsibility for its content.

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Safaa Touihmi: Writing, study concept of the paper

Ilham rkain:supervising.

Trial registry number

1. Name of the registry:
2. Unique Identifying number or registration ID:
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Guarantor

Safaa Touihmi.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Provenance and peer review

Not commissioned, externally peer reviewed.

Declaration of competing interest

The authors declare no conflicts of interest.

References

- [1] A.M. Al-Serhani, K. Al-Mazrou, Pharyngeal Tuberculosis, *Am J of Otolaryngol*, 2001 [Revue].
- [2] A. Goyal, N.M. Nogarkar, Tuberculosis of the pyriform fossa, *J. Laryngol. Otol.* 112 (8) (1998) [Revue].
- [3] K. Iravani, S.B. Jahromi, M.J. Ashraf, Primary tuberculosis of the vallecula and pyriform sinus: a case report and review of literature, *Iran. J. Otorhinolaryngol.* 8 (2015) 834. [Revue].
- [4] Kamyar Iravani1 Shahindokht Bassiri Jahromi2, Mohammad Javad Ashraf primary tuberculosis of the vallecula and pyriform sinus: [Revue]. - [s.l.], Iran. *J. Otorhinolaryngol.* 27 (2) (Mar 2015). Serial No.79.
- [5] Y.S. sai, L.W. Chen, C.Y. Ou, C.Y. Chu, C.C. Liu, S.M. Huang, Tuberculosis pyriform sinus Fistula infection mimics subacute thyroiditis, *Clin. Thyroidol.* 26 (1) (2014 Jan 1) 21–24 [Revue].
- [6] Touiheme N* Hmidi M, Nakkabi I, Belatik H, Attifi H, Boukhari A, Nadour K Primary Hypopharynx Tuberculosis [Revue]. - [s.l.] : Touiheme N*, Hmidi M, Nakkabi I, Belatik H, Attifi H, Boukhari A, Nadour K.
- [7] Steven DiLauro, F. Nancy, Crum-cianfone. "Ileitis: when it is not Crohn's disease, *Curr. Gastroenterol. Rep.* 4 (2010) 58–249.
- [8] Case 154: Pharyngeal and Retropharyngeal Tuberculosis with Nodal Disease Aditya Bharatha, Eric S. Bartlett, Eugene Yu.
- [9] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, for the SCARE Group, The SCARE 2020 guideline: updating Consensus surgical CAsE REport (SCARE) guidelines, *Int. J. Surg.* 84 (2020) 226–230.