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## CASE REPORT

### Extraganglionic Localization of Tuberculosis in Otorhinolaryngology: Rare Presentations About 29 Cases

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## ABSTRACT

The extra nodal tuberculosis is a rare disease in Ear, Nose and Throat. The aim of this work is to study the epidemiological, clinical, therapeutic profiles of rare locations of ENT extra nodal tuberculosis.

This is a retrospective study between June 2003 and June 2019, including 29 cases of ENT extra nodal tuberculosis.

There were 6 cases of laryngeal tuberculosis, 5 cases of nasopharyngeal tuberculosis, 5 cases of salivary gland tuberculosis, 3 cases of tonsillar tuberculosis, 1 case of tuberculous retropharyngeal abscess, 3 cases of nasal tuberculosis, 3 cases of tuberculosis of the middle ear, 1 case of thyroid tuberculosis, 1 case of tuberculosis of the bony palate, and 1 case of mandibular tuberculosis.

All ages were affected with a predominance of young adults. The clinical signs, as well as radiological or endoscopic examinations, were not specific. The diagnosis was based on histology and/or bacteriology. Antibacterial therapy was instated in all cases.

Extra nodal tuberculosis in ENT is rare. It is dominated by the laryngeal location. It must be considered as a differential diagnosis in front of any atypical symptoms, particularly in endemic countries.

**Keywords:** tuberculosis, ENT, extranodal, therapy.

## INTRODUCTION

Tuberculosis is an infectious disease caused by *Mycobacterium tuberculosis* or bacillus Koch (BK), which mainly affects the lungs; however, extrapulmonary locations can occur in one-third of cases<sup>1</sup>. Cervical lymph node involvement is the most common extrapulmonary location and it's mostly represented by the lymph node localisation<sup>2</sup>.

Extranodal ear, nose, and throat (ENT) sites are rare and are dominated by laryngeal localization<sup>3</sup>. The diagnosis is hard to suspect and, given the clinical polymorphism, it could be hard to assess by clinicians if it is not considered, leading to multiple complications and sequelae that could be prevented by an early diagnosis assessment and treatment by chemotherapy<sup>2-4</sup>.

Thus, the importance of our study where we describe the epidemiological, clinical, therapeutic, and evolutionary profile of rare extra-nodal ENT tuberculosis localizations. This will make clinicians aware of these rare localizations of tuberculosis and suspect the diagnosis.

## MATERIALS AND METHODS

This is a retrospective descriptive study about 29 cases reporting patients presenting extra-nodal tuberculosis in ENT tuberculosis, over a period of 16 years from June 2003 to June 2019.

For each location, we analyzed the epidemiological characteristics, the diagnostic aspects, and the therapeutic modalities.

- Epidemiological characteristics, we studied the following parameters: age, sex, socioeconomic level, vaccination status, and personal and/or family history of tuberculosis.
- Clinically, we studied the functional symptoms, the general condition, the existence of signs of

tuberculosis, or the existence of another tuberculosis location.

- On the paraclinical level:
  - we first performed diagnosis orientation examination: intradermal tuberculin reaction, biological assessment, radiological assessment;
  - Then paraclinical exams for diagnostic confirmation:
    - Bacteriological confirmation: by the search for acid-fast bacilli by direct examination or culture on Lowenstein cultivation environment.
    - And/or histological confirmation: by showing an epitheliogigantocellular granuloma with caseous necrosis.

The medical and surgical treatment modalities are specified, as well as the results of the treatment.

## RESULTS

Our study included 17 males and 12 females, aged between 9 and 65 years with an average age of 35,6 years. 19 cases (80%) were vaccinated against tuberculosis, 5 cases had a history of treated pulmonary tuberculosis.

The locations consisted of: 6 cases of laryngeal tuberculosis (fig 1), 5 cases of nasopharyngeal tuberculosis (fig 2), 5 cases of salivary gland tuberculosis (fig 3), 3 cases of tonsillar tuberculosis, 1 case of tuberculous retropharyngeal abscess, 3 cases of nasal tuberculosis (fig 4), 3 cases of tuberculosis of the middle ear (fig 5), 1 case of thyroid tuberculosis, 1 case of tuberculosis of the bony palate (fig 6), and 1 case of mandibular tuberculosis (Table 1).

**Table 1:** distribution of patients according to the location of tuberculosis involvement

| Tuberculosis locations | N | %     |
|------------------------|---|-------|
| Larynx                 | 6 | 20.68 |
| Nasopharynx            | 5 | 17.24 |
| Salivary glands        | 5 | 17.24 |
| Tonsils                | 3 | 10.34 |
| Nasal                  | 3 | 10.34 |
| Middle ear             | 3 | 10.34 |
| Retropharyngeal space  | 1 | 3.44  |
| Thyroid gland          | 1 | 3.44  |
| Palate bone            | 1 | 3.44  |
| Mandible               | 1 | 3.44  |



Fig 1: Laryngeal tuberculosis; **A:** Laryngeal tumor in endoscopic view  
**B:** frontal view of a cervical scanner, showing an obstructive tumor process of the larynx

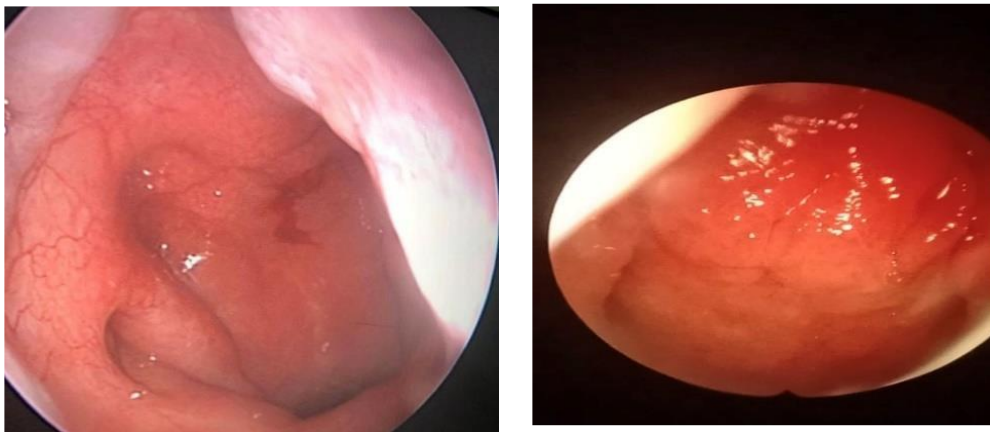


Fig 2: Nasopharyngeal tuberculosis, manifested by a nasopharyngeal bump in endoscopy



Fig 3: Parotid gland tuberculosis; **A:** Parotid bump infiltrating the local skin, **B:** Parotid bump with a fistulae



Fig 4: Nasal tuberculosis; anterior rhinoscopy showing a septal perforation



Fig 5: Middle ear Tuberculosis; **A:** Otoscopic aspect of tuberculous otitis media with multiple tympanic membrane perforations. **B:** Retroauricular lump fistulae



Fig 6: Hard palate tuberculosis; an ulcerous lesion of the hard palate

A second tuberculosis location was found in 10 cases, including three pulmonary locations concomitant with laryngeal tuberculosis and seven cervical lymph node locations in four patients with rhinopharyngeal tuberculosis and two patients with

parotid tuberculosis and one case of middle ear tuberculosis.

Clinically, general signs of tuberculosis found were presented as follow:

**Table 2:** frequency of general signs of tuberculosis in our series

| General signs | N  | %  |
|---------------|----|----|
| Weight loss   | 18 | 70 |
| Fever         | 10 | 40 |
| Anorexia      | 13 | 50 |
| Night Sweats  | 9  | 35 |

On the other hand, patients presented signs specific to the locations. The signs and the examination results are structured in table 3:



**Table 3:** Patient presentation and clinical examination according to the tuberculosis location

| Location              | Main patient complains                               | Associated Signs                     | Clinical examination results  |
|-----------------------|--|--------------------------------------|---|
| Larynx                | Chronic Dysphonia                                    | Dyspnea                              | Laryngeal Tumor (Fig 1A)<br>Epiglottis ulceration   |
| Rhinopharynx          | Cervical nods  | Epistaxis<br>Unilateral hearing loss | Rhino pharyngeal tumor (Fig 2)  |
|                       | bilateral nasal obstruction                          | -                                    | Rhinopharyngeal ulceration  |
| Salivary glands       | parotid tumefaction                                  | -                                    | Parotid mass  |
|                       | Submandibular tumefaction                            | -                                    | Submandibular Mass  |
| Nasal                 | persistent purulent rhinitis                         | -                                    | septal perforation (Fig 4)<br>vegetation of the inferior cone   |
| Tonsil                | Odynophagia  | -                                    | unilateral tonsillar hypertrophy<br>tonsillar ulcers  |
| Retropharyngeal space | Torticollis  | Dysphagia                            | - a bulge of the oropharynx<br>- exquisite pain on palpation of the transverse process of the 4th cervical vertebra                           |
| Thyroid gland         | anterior cervical pain                               | -                                    | firm and slightly tender palpable thyroid mass without inflammatory signs   |
| Bone palate           | ulceration of the hard palate resistant to treatment | -                                    | Hard palate ulceration  |
| Middle ear            | purulent otorrhea resistant to treatment             | - otomastoiditis<br>- Hypoacusia     | - Purulent otorrhea<br>- Renitent retro auricular mass with inflammatory signs ( Fig 5B)<br>- Multiple tympanic membrane perforation (Fig 5A) |
| Mandible              | Mandibular swelling                                  | -                                    | - Painful mandibular swelling<br>- Intra-Oral fistula   |

Paraclinically, the sedimentation rate was performed in only 18 patients and was positive in 15 cases.

The intradermal tuberculin reaction was positive in 18 cases, negative in two cases and not performed in nine cases; two cases of tonsillar localization, a case of submandibular localization, a case of parotid localization, a case of thyroid localization and a case of the mandibular localization and three cases of middle ear localization.

Chest radiography was performed in all cases and showed images in favor of progressive pulmonary tuberculosis in three cases while in five cases it showed images of sequelae lesions.

CT scans were performed in 2 cases of laryngeal involvement (Fig 1B), 2 cases of rhinopharyngeal involvement, 2 cases of parotid swelling, one case of retropharyngeal abscess (Fig 7), one case of otomastoiditis (Fig 8) and a case of mandibular tuberculosis (Fig9).

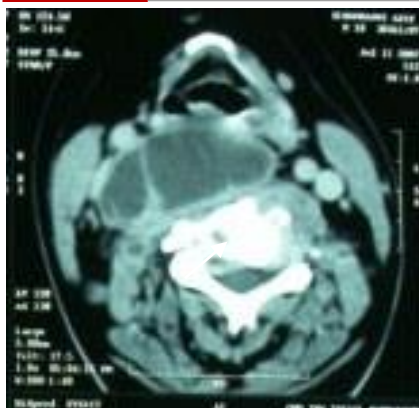


Fig 7: Retropharyngeal tuberculosis abscess; Cervical CT, The arrow showing an important retropharyngeal collection, reaching the cervical spine with spondylodiscitis

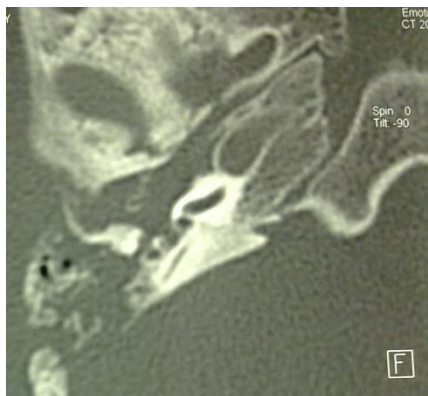


Fig 8: Otomastoiditis secondary to tuberculosis; Mastoidian CT Scan showing a filling of the tympanic cavity with osteolysis of the mastoid

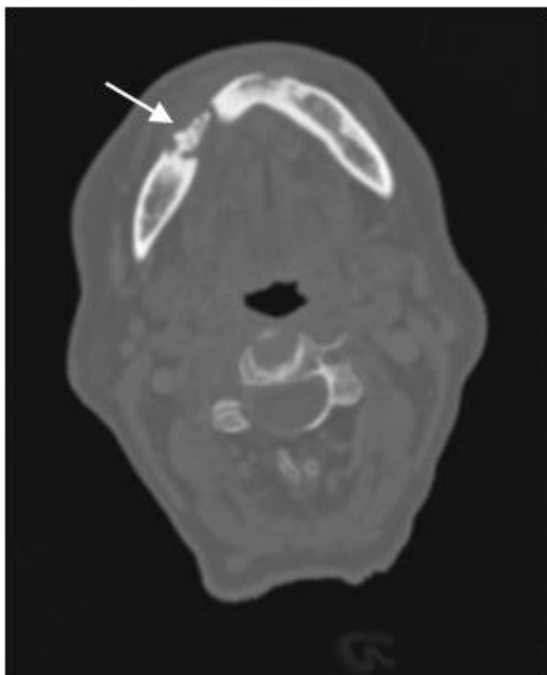


Fig 9: mandibular tuberculosis; facial CT scan in axial section, the arrows showing a right parasymphseal osteolytic process type Ib of Lodwick with bone sequestration

The confirmation of the diagnosis was based on histological examination in all cases; established on biopsies or on excisional parts; that showed an epitheliogiantocellular granuloma with caseous necrosis.

All our patients received a 6-month regimen of antituberculosis drugs; a 2-month loading phase based on rifampicin, isoniazid and pyrazinamide and a 4-month maintenance phase based on rifampicin and isoniazid; Except for the patient diagnosed with the retropharyngeal abscess associated to pott's sickness in whom the maintenance phase of treatment was maintained for 7 months.

Seven patients benefited from a surgical treatment including: one case of tracheotomy for a severe dyspnea caused by laryngeal tuberculosis, a case of submandibulectomy, a case of parotidectomy, two nasal septum perforation reconstruction, a left loboisthmectomy.

The evolution was favorable for all patients. A patient with nasopharyngeal location relapsed at the end of the treatment; and two cases of parotid localizations kept retractile scars in the parotid region.

The following table is a recap of the findings of our study:

**Table 4:** Localization, epidemiology, diagnosis and treatment of our series' cases.

| Case | Localization          | Age | Sex | History of tuberculosis | Second tuberculosis localization | Vaccine | IDR | Confirmation                 | Treatment                       |
|------|-----------------------|-----|-----|-------------------------|----------------------------------|---------|-----|------------------------------|---------------------------------|
| 1    | Larynx                | 65  | M   | Lung                    | -                                | Unk     | +   | Pathology                    | + 2RHZE/4RH<br>2SRHZ/4RH        |
| 2    |                       | 32  | M   | -                       | Lung                             | +       | +   | Bacteriology                 |                                 |
| 3    |                       | 49  | M   | -                       | Lung                             | +       | +   | Pathology                    |                                 |
| 4    |                       | 27  | M   | -                       | -                                | +       | +   | Bacteriology                 |                                 |
| 5    |                       | 34  | M   | -                       | Lung                             | +       | +   | Pathology                    |                                 |
| 6    |                       | 48  | M   | Lung                    | -                                | +       | +   | Pathology                    |                                 |
| 7    | Nasopharynx           | 40  | M   | Lung                    | -                                | +       | +   | Pathology                    | 2RHZE/4RH                       |
| 8    |                       | 60  | M   | Lung                    | Cervical nods                    | Unk     | +   | Pathology                    | 2RHZE/4RH                       |
| 9    |                       | 28  | M   | -                       | Cervical nods                    | +       | +   | Pathology                    | 2RHZE/4RH                       |
| 10   |                       | 30  | M   | -                       | Cervical nods                    | +       | +   | Pathology                    | 2RHZE/4RH                       |
| 11   |                       | 21  | F   | -                       | Cervical nods                    | +       | +   | Pathology                    | 2RHZE/4RH                       |
| 12   | Parotid gland         | 20  | F   | -                       | Cervical nods                    | +       | +   | Pathology                    | 2RHZE/4RH                       |
| 13   |                       | 13  | F   | -                       | -                                | +       | ND  | Pathology                    | Parotidectomy +2RHZE/4RH        |
| 14   |                       | 38  | F   | -                       | Cervical nods                    | Unk     | +   | Pathology                    | 2RHZE/4RH                       |
| 15   |                       | 60  | M   | -                       | -                                | Unk     | +   | Pathology                    | 2RHZE/4RH                       |
| 16   | Sub mandibular gland  | 55  | M   | -                       | -                                | Unk     | ND  | Pathology                    | sub-maxilectomy+2RHZE/4RH       |
| 17   | Tonsil                | 18  | M   | -                       | -                                | +       | ND  | Pathology                    | tonsillectomy+2RHZE/4RH         |
| 18   |                       | 23  | M   | -                       | -                                | +       | ND  | Pathology                    | tonsillectomy+2RHZE/4RH         |
| 19   |                       | 39  | F   | -                       | -                                | +       | +   | Pathology                    | 2RHZE/4RH                       |
| 20   | Retropharyngeal space | 38  | M   | -                       | -                                | Unk     | +   | Pathology<br>Bacteriology    | + surgical drainage+2RHZE/7RH   |
| 21   | Nasal cavity          | 17  | F   | -                       | -                                | -       | +   | -                            | septal reconstruction+2RHZE/4RH |
| 22   |                       | 43  | F   | -                       | -                                | -       | -   | Pathology                    | septal reconstruction+2RHZE/4RH |
| 23   |                       | 45  | F   | -                       | -                                | -       | -   | -                            | 2RHZE/4RH                       |
| 24   | Hard palate           | 32  | F   | -                       | -                                | +       | +   | Pathology                    | 2RHZE/4RH                       |
| 25   | Thyroid gland         | 50  | F   | Lung                    | -                                | +       | ND  | Pathology                    | Loboisthmectomy+2RHZE/4RH       |
| 26   | Middle ear            | 9   | M   | -                       | Cervical Nods                    | +       | +   | Pathology                    | and                             |
| 27   |                       | 12  | F   | -                       | -                                | +       | ND  | bacteriology                 | 2RHZE/4RH                       |
| 28   |                       | 18  | M   | -                       | -                                | +       | ND  | Bacteriology<br>Bacteriology |                                 |
| 29   | Mandible              | 70  | F   | -                       | -                                | -       | -   | Pathology<br>bacteriology    | and<br>2RHZE/7RH                |

M:Male; F: Female; Unk: Unknown;ND: Not Done; R:Rifampicin, H: isoniazid; Z: pyrazinamide ; E: Ethambutol; S: Spiramicin

## DISCUSSION

Extra pulmonary tuberculosis can be seen in one third of cases<sup>1-2</sup>. Cervical lymph node localization is the most common extra pulmonary localization. Moreover, other ENT sites are even rarer<sup>1-4</sup>

### *Laryngeal tuberculosis*

Laryngeal localization of tuberculosis is rare and represents less than 1% of the localizations of this disease<sup>5</sup>. Nevertheless, in the ENT sphere, this localization remains the most frequent and can reach 46%<sup>6,7</sup>. It is often associated with a known pulmonary location. In our study, 50% of the cases of laryngeal tuberculosis had concomitant pulmonary tuberculosis.

Koch's bacillus can infect the larynx by two different routes: either directly from the external environment in primary laryngeal tuberculosis or indirectly from a distant site in secondary laryngeal tuberculosis<sup>8,9</sup>. The signs of primary and secondary tuberculosis of the larynx are different. Laryngeal tuberculosis is easy to be misdiagnosed as laryngitis and laryngeal tumors, thus the importance of the pathological exam<sup>10</sup>.

The age of onset is variable but the age group most affected seems to be between 40 and 50 years with a predominance of male, this is probably related to high alcohol and tobacco consumption. The latter acts as a factor of local irritation<sup>6</sup>. There are no specific clinical symptoms. Cervical adenopathy is present in less than 10% of cases<sup>11</sup>. The laryngeal lesions affect with predilection the glottis, the laryngeal vestibule and the subglottic floor<sup>5</sup>. Macroscopically, laryngeal tuberculosis can present in several forms: ulceration, polyp or diffuse inflammation<sup>12</sup>. The diagnosis of certainty is based on endoscopy with multiple biopsies, at different sites, in order not to miss a maligned lesion. The pathological study confirms the presence of epitheliogigantocellular granulomas with caseous necrosis and the bacteriological study shows the presence of alcohol-acid-resistant bacilli on direct examination or after culture of laryngeal samples. The diagnosis of laryngeal tuberculosis should lead to a systematic search for associated pulmonary involvement by requesting a chest X-ray and by looking for BK in bronchial secretions<sup>8,13</sup>.

### *Nasal and rhinopharyngeal tuberculosis*

Nasal localization is exceptional and mostly affects middle-aged women<sup>14</sup>. This localization remains rare thanks to the specificities of the nasal mucosa<sup>15</sup>:

- mechanical protection provided by ciliary movements,

- bactericidal properties of nasal secretions
- And the lymphatic density of the pituitary mucosa resist the development of BK.

However, certain local factors (trauma, chronic atrophic rhinitis) or general factors (poor hygiene, immunosuppression) could facilitate the infection by BK<sup>1</sup>. Endoscopy finds the aspect of a granuloma or a superficial mucosal ulceration most often located in the anterior part of the nasal septum or in the inferior turbinate and rarely on the floor of the nasal cavity<sup>14-16</sup>. The lesion is often unilateral but bilateral lesions may occur in 30% of cases<sup>11</sup>. Untreated, it may be complicated by septal perforation, atrophic rhinitis or nasal synechiae<sup>1,3</sup>. Nasopharyngeal localization is very rare. It mainly affects young adults between 20 and 40 years<sup>1,2</sup>. Cervical nodes are almost constant, usually bilateral, voluminous and mobile. They are sometimes a revelation mode of the disease showing the lymphatic richness of the cavum. Rhinoscopy can show different macroscopic aspects; these may include one or more ulcerations, regular mucosal enlargement, a polypoid tumor or irregular swelling<sup>16,17</sup>. Biopsies allow anatomopathological and bacteriological confirmation and rule out cavum cancer.

### *Pharyngeal location (oropharynx, hypopharynx)*

Pharyngeal tuberculosis is mainly observed between 20 and 74 years of age<sup>19</sup> with a female predominance with a sex ratio of 2/3<sup>20</sup>. Tonsillar tuberculosis is most often secondary to a pulmonary localization and is favored by alcoholism and acquired immunodeficiency syndrome. Three modes of contamination are possible: hematological, airborne and lymphatic<sup>20</sup>.

General signs are often present, and several clinical forms have been described<sup>21</sup>: the ulcerous form, the tumoral form and the so-called "tuberculous lymphoma" form with a large, hard, unilateral tonsil. In our series, two cases presented with the hypertrophic form indicating tonsillectomy, which was incidentally diagnosed by pathological study. Other isolated cases reported in the literature discuss the value of immediate tonsillectomy when the diagnosis is confirmed on tonsil biopsy in a context suggestive of tuberculosis or tonsillar tumor<sup>20</sup>.

Retropharyngeal abscess remains a very rare location. The clinical diagnosis can be difficult; the clinical symptomatology is variable and non-specific, and consists mainly of signs of oropharyngeal compression: dysphagia, odynophagia, change in voice timbre or dyspnea<sup>22</sup>. CT and MRI scans are very useful in the



diagnosis, showing a retropharyngeal abscess, its contacts and especially the extension to the cervical spine (Pott's disease) as in the case of our patient. The immunodepressive background must always be sought. Treatment with anti-tuberculosis drugs alone seems to be risky even in the absence of myelopathy<sup>23</sup>. The therapeutic management of tuberculous retropharyngeal abscesses, secondary to cervical Pott's disease, is based on surgical drainage when the abscess is large, completed by the prescription of long-term antibacillary drugs<sup>24</sup>.

### **Oral cavity**

Involvement of the oral cavity is found in 0.2 to 1.5% of cases of tuberculosis, mostly in endemic areas<sup>25</sup>. Lingual localization is the most frequently observed<sup>12</sup>. Lingual trauma, carious teeth and smoking are known to favor tuberculosis infection. On the other hand, the environment of the oral cavity is hostile for the multiplication of BK: unfavorable local pH, the salivary flow and the paucity of lymphatic tissue<sup>1,2</sup>.

Clinically, oral tuberculosis can present in several ways. Tuberculous chancre, which is unique and rare. The typical tuberculous ulcer, often unique, occurs in a known tuberculosis patient. A tuberculoma, a chronic glossitis or an indurated lesion under the mucosa are possible<sup>1-3</sup>. In our series, we observed a case of non-hemorrhagic ulceration of the hard palate whose biopsy confirmed the diagnosis.

### **Salivary glands**

Parotid tuberculosis is rare, about 200 cases have been described in the literature<sup>1,4</sup>. This remains the most frequent location of the salivary glands [26]. The mode of contamination is discussed by the authors. For the majority of them, salivary gland tuberculosis is considered as a secondary location and the infection can occur by different routes<sup>27,28</sup>:

- The hematogenous route and the lymphatic route remain the most likely.
- The direct retrograde ductal route via Stenon's canal for the parotid gland or Wharton's canal for the submandibular gland.
- The retrograde lymphatic route from a locoregional focus.
- Parenchymal invasion from an intraglandular adenopathy.

However, if the primary location is not found, salivary gland tuberculosis is described as primary. In our series, 2 cases of parotid localizations were associated with homolateral cervical nodes, allowing the diagnosis to be concluded and medical

treatment to be initiated on the basis of pathological study of the lymph nodes.

The disease is predominantly found in young female adults<sup>26</sup>. The average age of the patients is 47 years<sup>27</sup>. Nevertheless, cases have been described in children<sup>24</sup>.

The diagnosis of tuberculosis is based on the anatomopathological study of a lymph node biopsy, of a biopsy of the edges in case of fistula or on a surgical specimen after conservative parotidectomy<sup>27</sup>.

Tuberculosis of the submandibular gland may be latent for a long time and may present as a trivial sub maxillitis. General signs are often absent. Bacteriological examination after culture of the pus taken from the Wharton's canal can sometimes reveal BK. Only anatomopathological examination of the surgical specimen can confirm the diagnosis<sup>28</sup>.

### **Thyroid gland**

Thyroidal tuberculosis is most often secondary to a general dissemination of the disease during a miliary tuberculosis<sup>29</sup>. In other cases, the disease spreads directly from a laryngeal focus or a nearby lymph node or by hematogenous route<sup>29-31</sup>. The primary location is exceptional. The high resistance of the thyroid gland is explained by a very good oxygenation of its parenchyma, by a rich lymphatic drainage system and by a thick fibrous capsule in addition to thyroid hormones and iodine which play a bacteriostatic role<sup>1,4,30-33</sup>. In the majority of cases, patients are clinically and biologically euthyroid<sup>31-33</sup>, as it was the case for our patient. The diagnosis is based on the presence of an epithelial-gigantocellular granuloma with caseous necrosis in the histological examination of the surgical specimen<sup>30</sup> and can mimic a parathyroid adenoma in some cases<sup>34</sup>.

### **Middle Ear**

Tuberculosis of the middle ear is exceptional and the diagnosis is often assessed lately<sup>35,36</sup>. It is responsible for 0.05 to 0.9% of chronic otitis media<sup>37</sup>. The classic triad of painless otorrhea, facial nerve palsy and multiple tympanic perforations is rarely observed<sup>38</sup>.

Currently, the most frequent reason for consultation in children is otorrhea refractory to conventional antibiotic therapy<sup>39</sup>.

In our series, the diagnosis was suspected when an otomastoiditis did not improve in a child undergoing usual antibiotic therapy. The diagnosis was confirmed by direct examination for *Mycobacterium tuberculosis* and by histological

study of the associated cervical lymph node. For the three other cases, the diagnosis was suspected before painless otorrhea and multiple tympanic membrane perforations. The diagnosis was confirmed by bacteriological examination of the pus.

Contamination can occur through dissemination from the airways to the Eustachian tube<sup>35-37</sup>, through the lymphatic route from the cervicothoracic lymph node involvement and, exceptionally, through the hematogenous route. If not diagnosed in time or not treated, the tubercular lesions may extend to the mastoid, the temporal bone, the central nervous system or the occipital condyle and the atlas, leading to instability of the craniovertebral junction<sup>35,40-41</sup>

### **Mandible tuberculosis**

Mandibular tuberculosis is extremely rare, and represents less than 2% of all forms of bone tuberculosis<sup>42</sup>. Only 0,05 – 5 % of total tuberculosis localizations have oral manifestation<sup>43</sup>

It could be presented as osteomyelitis of the mandible, arthritis of the temporomandibular joint or even a pathologic fracture<sup>44</sup>. Radiological aspects reported in the literature were condylar resorption, resorption of inferior border of the mandible owing to tuberculous lymphadenitis and osteomyelitis of the mandible<sup>42</sup>.

In our case, the patient presented a mandibular swelling and the CT scan showed a right para symphyseal osteolytic process type Ib of Lodwick with bone sequestration. The diagnosis is mainly confirmed by a bacteriological and pathological study showing the typical aspect of this disease.

The clinical symptoms of the different ENT locations are not specific. Same for radiological examinations such as ultrasound, CT scan or even MRI.

Five patients in our series did not have a intradermal tuberculin reaction because it was an insidious diagnosis; even when performed; it is not always positive and can lead the clinician wrong<sup>3,5</sup>. Contrarily to bacteriological culture, that may take several months to positivize, pathology, by revealing an epithelial-giganto-cellular granuloma with caseous necrosis, can provide formal proof within a reasonable period of time<sup>1,3-4</sup>.

Gene amplification techniques by PCR have made it possible to shorten the time required for diagnosis, but at a high cost, making this technique inaccessible in certain situations. It remains useful in cases of nasal or nasopharyngeal involvement, when histological study does not find caseous necrosis, thus suspecting a differential diagnosis

such as sarcoidosis, Wegener's granulomatosis, syphilis or leprosy<sup>17-19</sup>.

Blood tests for gamma interferon production (Quantiferon) are intended to support the diagnosis of latent tuberculosis infection and not to detect active tuberculosis.

In some cases, the diagnosis may be based only on the efficacy of a test anti-tuberculosis treatment in the context of strong suspicion, especially in cases of parotid and middle ear localization<sup>32,45,46</sup>.

In endemic countries, tuberculosis occurs in immunocompetent individuals. HIV serology is only requested if there are other signs of the disease<sup>1,3,5</sup>.

Treatment is most often medical with antituberculosis drugs for a minimum of six months<sup>1,3-5</sup>. The treatment consists of a 2-month loading phase of rifampicin, isoniazid and pyrazinamide and a 4-month maintenance phase of rifampicin and isoniazid. The role of surgery in the therapeutic strategy for extranodal ENT disease remains limited. It is limited to performing a tracheotomy in cases of obstructive laryngeal tuberculosis with inspiratory dyspnea or to treating sequels such as laryngeal stenosis or nasal septal perforations. In our series, the diagnosis was made incidentally by surgery, especially in glandular locations (submandibular, parotid and thyroid) or after tonsillectomy.

The evolution is most often favorable whatever the location. The rare cases of failure can be explained by the emergence of resistant strains<sup>47</sup>

### **CONCLUSION**

Extranodal ENT tuberculosis is rare. It is dominated by laryngeal involvement. It must be considered in the presence of any slow and atypical symptomatology in endemic countries as well as in developed countries of low endemicity, because of the recrudescence of tuberculosis due to HIV infection. Imaging is of little interest because it is non-specific. The diagnosis of certainty is essentially based on the histological study by the demonstration of an epithelio-giganto-cellular granuloma with caseous necrosis and possibly on the bacteriological study by the demonstration of BAARs on direct examination or culture on Loweinstein culture. Its evolution is most often favorable under medical treatment.

### **CONFLICT OF INTREST**

Authors declare no conflict of interest

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None

## REFERENCES

1. El Ayoubi, A., Benhammou, A., El Ayoubi, F., El Fahssi, A., Nitassi, S., Kohen, A., Nazih, N., Benbouzid, A. and Kzadri, M. (2009) La tuberculose primitive ORL extraganglionnaire. *Annales d'oto-Laryngologie et Chirurgie Cervico-Faciale*, 126, 208-215
2. Mocanu AI, Mocanu H, Moldovan C, Soare I, Niculet E, Tatu AL, Vasile CI, Diculencu D, Postolache PA, Nechifor A. Some Manifestations of Tuberculosis in Otorhinolaryngology – Case Series and a Short Review of Related Data from South-Eastern Europe. *Infect Drug Resist*. 2022;15:2753-2762 <https://doi.org/10.2147/IDR.S367885>
3. Tuang GJ, Muhammad A, Zahedi FD. 'Master of many faces: extrapulmonary tuberculosis in the eyes of otolaryngologists'. *Postgrad Med J*. 2022;98(1158):311-312. doi:10.1136/postgradmedj-2020-139414
4. Subramani S, Saravanam PK, Rajendran R. Extrapulmonary tuberculosis: atypical presentation in otorhinolaryngology. *BMJ Case Rep*. 2018;2018:bcr2018224802. Published 2018 Aug 29. doi:10.1136/bcr-2018-224802.
5. Sahtout S, Kharrat S, Jaaari F, Nouira K, Kchouk I, Besbes G et al. La tuberculose laryngée. A propos de 7 cas. *J Tun. ORL* 2004 ;13:65-7.
6. Kossowski M, Conessa C, Clement P, Roguet E, Verdalle P, Poncet JL. Aspects actuels de la tuberculose laryngée: à propos de 4 cas. *Ann Otolaryngol Chir Cervicofac* 2002;119:281-6.
7. Kandiloros DC, Nikolopoulos TP, Ferekidis EA, et al. Laryngeal tuberculosis at the end of the 20th century. *J Laryngol Otol*. 1997;111(7):619-621. doi:10.1017/s0022215100138137
8. Ben Gamra O, M'barek CH, Khammassi K, Ben Hamouda R, Hariga I, Ben Amor M et al. La tuberculose des voies aerodigestives supérieures. *Rev Tun Infectiol* 2008, 2, 3: 21-25.
9. Luckhaupt H. Zur Geschichte der Kehlkopftuberkulose [The history of tuberculosis of the larynx]. *Laryngorhinootologie*. 2021;100(9):726-730. doi:10.1055/a-1550-2802
10. Ai Y, Liu H, Xu H, Liu Y, Li L. *Lin Chung Er Bi Yan Hou Tou Jing Wai Ke Za Zhi*. 2021;35(1):38-41. doi:10.13201/j.issn.2096-7993.2021.01.009
11. Saroul N, Vellin J-F, Baud O, Nohra O, Kemeny J-L, Gilain L. Tuberculose épilaryngée: considérations épidémiologiques, cliniques et sanitaires. *Ann Otolaryngol Chir Cervicofac* 2008;125:155-159.
12. Nalini B, Vinayak S. Tuberculosis in ear, nose, and throat practice: its presentation and diagnosis. *Am J Otolaryngol*. 2006;27(1):39-45. doi:10.1016/j.amjoto.2005.07.005
13. Bass B, Shah V, Menon A, Tilley A. A 51-Year-Old Man With Hoarseness of Voice. *Chest*. 2020;157(4):e115-e119. doi:10.1016/j.chest.2019.10.003
14. Kim YM, Kim AY, Park YH, Kim DH, Rha KS. Eight cases of nasal tuberculosis. *Otolaryngol Head Neck Surg*. 2007;137(3):500-504. doi:10.1016/j.otohns.2007.04.009
15. Ahmed S, Mustahsan SM, Jamil B, Ilyas F. Nasopharyngeal Tuberculosis. *J Coll Physicians Surg Pak*. 2022 Apr;32(4):S39-S40. doi: 10.29271/jcpsp.2022.Supp1.S39. PMID: 35633007.
16. Hadid, F., Oulghoul, O., Benjelloun, H., Benhommad, O., Raji, A., & Rochdi, Y. (2021). Nasopharyngeal Tuberculosis Simulating Cancer: About 2 Cases. *European Journal of Medical and Health Sciences*, 3(3), 46–48. <https://doi.org/10.24018/ejmed.2021.3.3.852>
17. Butt AA. Nasal tuberculosis in the 20th century. *Am J Med Sci* 1997;313:332-5.
18. Kharoubi S, Vallicioni JM. La tuberculose du cavum: à propos d'une série de 6 cas. *Les cahiers d'ORL et de chirurgie cervico-faciale et d'audiophonologie* 1997;32:446-451.
19. Srirompotong S, Yimtae K, Srirompotong S. Clinical aspects of tonsillar tuberculosis. *Southeast Asian J Trop Med Public Health*. 2002;33(1):147-150.
20. Rakotoarisoa AHN, Ramarozatovo NP, Razafindrakoto RMJ, Andriatsoavinarivomanana J, Tsihoery F, Razafindrabe JAB. Tuberculose amygdalienne et ganglionnaire cervicale: à propos d'un cas. *Revue Tropicale de Chirurgie* 2010;4:6-7.
21. Gulati A, Kaushik R, Sharma J. Primary tuberculosis of the tonsil masquerading as carcinoma. *Indian J Pathol Microbiol* 2006;49:312-3.
22. Elqatni M, Sekkach Y, Hammi S, Elomri N, Fatih J, Mekouar F et Al. Un abcès rétropharyngé. *La Revue de Médecine Interne* 2011;32(5):319-320.
23. Büyükbekbeci O, Karakurum G, Daglar B, Maralcan G, Güner S, Güleç A. Tuberculous

- spondylitis: abscess drainage after failure of anti-tuberculous therapy. *Acta Orthop Belg.* 2006;72(3):337-341.
24. Benhammou A, Bencheikh R, Benbouzid MA, Boulaich M, Essakali L, Kzadri M. Abcès rétropharyngés révélant un mal de Pott cervical [Cervical Pott's disease revealed by retropharyngeal abscesses]. *Rev Stomatol Chir Maxillofac.* 2007;108(6):543-546. doi:10.1016/j.stomax.2007.03.001
  25. Vidal M, Delevaux I, André M, Marroun I, Gavet F, Voinchet H, et al. Tuberculose disséminée révélée par une localisation linguale. *La Rev Med Interne* 2007;28:124-6.
  26. Raji A, Mahtar M, Essaadi M, Chekkoury IA, Benchakroun Y. La tuberculose parotidienne. À propos de six cas. *Méd Mal Infect* 2002 ;32:196-201.
  27. Benhamou A, Elayoubi A, Essakali L, Kzadri M. La tuberculose primaire de la glande parotide. *Archive de pédiatrie* 2007;14:1206-9.
  28. Manipoud P, Rerolle S, Kemeny JL, Lafaye M. Tuberculose de la glande sousmaxillaire. *Ann.Oto-laryng* 1993;110(8)482-484.
  29. Kabiri H, Atoini F, Zidane A. La tuberculose thyroïdienne. *Ann d'endocrinologie* 2007;68:196-8.
  30. Bansal LK, Gupta S, Gupta AK, Chaudhary P. Thyroid tuberculosis. *Indian J Tuberc.* 2021 Apr;68(2):272-278. doi: 10.1016/j.ijtb.2020.09.006. Epub 2020 Sep 12. PMID: 33845964.
  31. Rojo-Abecia M, Valdazo-Gómez AM, Román CF, et al. Primary thyroid tuberculosis: a case report. Tuberculosis tiroidea primaria: reporte de un caso. *Cir Cir.* 2021;89(S2):1-3. doi:10.24875/CIRU.21000027
  32. Majumder A, Sen D. Uncommon affliction of a common disease - primary tuberculosis of thyroid gland. *Indian J Pathol Microbiol.* 2020;63(1):83-85. doi:10.4103/IJPM.IJPM\_719\_18
  33. Darre T, Amana B, Pegbessou E, Winga F, Warou D, Napo-Koura G. Tuberculosis of the thyroid in Togo: a clinicopathologic study of 11 presumed cases. *Trop Doct.* 2018;48(4):313-316. doi:10.1177/0049475518789555
  34. Goyal A, Agarwal S, Goswami R, Bal C. Hyperplastic thyroid nodule masquerading as parathyroid adenoma in a patient with tubercular lymphadenitis induced hypercalcaemia. *BMJ Case Rep.* 2020;13(11):e237261. Published 2020 Nov 9. doi:10.1136/bcr-2020-237261
  35. Doan HTH, Hoang PT, Tran TPC. Tuberculous otitis media in Vietnam: Clinical features and diagnostic difficulties. *Eur Ann Otorhinolaryngol Head Neck Dis.* 2021;138(6):467-469. doi:10.1016/j.anorl.2021.05.009
  36. Hand JM, Pankey GA. Tuberculous Otomastoiditis. *Microbiol Spectr.* 2016;4(6):10.1128/microbiolspec.TNMI7-0020-2016. doi:10.1128/microbiolspec.TNMI7-0020-2016
  37. Touati MM, Darouassi Y, Chihani M, et al. La tuberculose extra-ganglionnaire primitive de la sphère ORL: à propos de 15 cas [ENT primitive extranodal tuberculosis: about 15 cases]. *Pan Afr Med J.* 2014;19:179. Published 2014 Oct 21. doi:10.11604/pamj.2014.19.179.4597
  38. Vaamonde P, Castro C, García-Soto N, Labella T, Lozano A. Tuberculous otitis media: a significant diagnostic challenge. *Otolaryngol Head Neck Surg.* 2004;130(6):759-766. doi:10.1016/j.otohns.2003.12.021
  39. Bakshi SS, Kaipuzha RR. Tuberculous otitis media. *Med Clin (Barc).* 2019 May 3;152(9):e53. English, Spanish. doi: 10.1016/j.medcli.2018.06.017. Epub 2018 Aug 27. PMID: 30166127.
  40. Nicolau Y, Northrop C, Eavey R. Tuberculous otitis in infants: temporal bone histopathology and clinical extrapolation. *Otol Neurotol.* 2006;27(5):667-671. doi:10.1097/01.mao.0000224085.08344.50
  41. Mongkolrattanothai K, Oram R, Redleaf M, Bova J, Englund JA. Tuberculous otitis media with mastoiditis and central nervous system involvement. *Pediatr Infect Dis J.* 2003;22(5):453-456. doi:10.1097/01.inf.0000066245.88741.bf
  42. Sansare K, Gupta A, Khanna V, Karjodkar F. Oral tuberculosis: unusual radiographic findings. *Dentomaxillofac Radiol.* 2011;40(4):251-256. doi:10.1259/dmfr/75047143
  43. Garber HT, Harrigan W. Tuberculous osteomyelitis of the mandible with pathologic fracture. *J Oral Surg.* 1978;36(2):144-146.
  44. Towdur GN, Upasi AP, Veerabhadrapa UK, Rai K. A Rare, Unusual Presentation of Primary Tuberculosis in the Temporomandibular Joint. *J Oral Maxillofac Surg.* 2018;76(4):806-811. doi:10.1016/j.joms.2017.09.010
  45. Sethi A, Sareen D, Sabherwal A, Malhotra V. Primary parotid tuberculosis: varied clinical

- presentations. *Oral Dis.* 2006;12(2):213-215. doi:10.1111/j.1601-0825.2005.01182.x
45. Bouayed S, Kos MI, Guyot JP. La tuberculose de l'oreille : à propos de deux cas. *Rev Med Suisse Romande* 2005;1:2396, 2399-400, 2402-3.
46. El Malki HO, El Absi M, Mohsine R, Aït Taleb K, Chefchaoui MC, Oulbacha S, et al. La tuberculose de la thyroïde, Diagnostic et traitement. *Ann Chir* 2002;127:385-7.