CASE REPORTS

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Multifocal Tuberculosis Presenting as Mandibular Swelling in a 3-Year-Old Boy: A Case Report

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ABSTRACT

Objective: To report a case of multifocal pediatric tuberculosis presenting with mandibular swelling and discuss its etiology, clinical findings, diagnosis, management, and outcome after treatment.

Methods:

Design: Case Report
Setting: Tertiary Government Training Hospital
Patient: One

Results: A 3-year-old boy presented with progressive non-tender, right mandibular swelling for 11 months. Panoramic X-ray exhibited extensive multiple loculations with lytic changes on the mandible. CT Scans revealed a peripherally enhancing hypodense mass with lytic expansion of the right mandibular angle extending across the left mandibular body with an incidental finding of right lung mass. Other extrapulmonary lesions were also detected involving the scapula, pleura with lysis of the adjacent ribs at the level of T7 and T8. Biopsy of the mandibular and lung mass confirmed the presence of caseating and non-caseating granulomas consistent with Koch's infection. The patient showed significant improvement by the 7th month of a 12-month course of anti-tuberculous therapy.

Conclusion: Multifocal TB can present as simple mandibular swelling, and a thorough workup should look for other involved sites. Early diagnosis in children may prevent debilitating sequelae and improve long-term treatment outcomes.

Keywords: *tuberculosis (TB); mandible; child; multifocal; disseminated; extrapulmonary*

Tuberculosis (TB) a granulomatous disease caused by *Mycobacterium tuberculosis*, *M. bovis* and atypical mycobacteria[1] that is highly curable but still ranks as the number one killer among all infectious diseases.[2] The World Health Organization (WHO) estimates 1.8 million people in the Western Pacific Region developed active TB in 2016 with 573,000 (32%) in the Philippines.[3]

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The majority of cases (98.9%) had pulmonary TB while 1.1% had extrapulmonary (EPTB).[4] Tuberculous osteomyelitis of the mandible is rare and is only observed in <2% of skeletal tuberculosis.[1] To our knowledge, there are two locally-reported cases of EPTB involving the mandible in a 10-year-old boy[5] and the temporomandibular region in a 33-year-old man.[6] We report what we believe is the second locally reported case of TB of the mandible in a child, a case of multifocal pediatric tuberculosis, and discuss its clinical course, diagnosis, management and outcome after treatment.

CASE REPORT

A 3-year-old fully immunized boy presented with right mandibular swelling of 11 months. Two months prior to consultation, episodes of low-grade fever were noted followed by gum bleeding admixed with purulent discharge. He was brought to a dentist and a panoramic x-ray revealed lytic changes encompassing the right mandibular angle extending across the left mandibular body. *(Figure 1)* Oral Clindamycin was taken for 7 days with no improvement of symptoms.

On the day of consult, physical examination showed an afebrile, underweight and stunted child with no developmental delay (Z-score < 2, based on WHO weight-for-age and height-for-age, 2-5 years old) with diffuse swelling of the right mandible and non-hyperemic, non-tender overlying skin. There was purulent, bloody discharge from the crown of the right second mandibular molar with diffuse hyperemia of surrounding gingival mucosa. *(Figure 2)* There were dental caries and multiple non-tender lymph nodes were palpated in both submandibular and submental areas.

There was a family history of multiple abscesses in his maternal grandfather and three uncles that lasted for weeks during their childhood, leaving pitted scars over the thorax, knees, and thighs. Unknown medications had been taken.

The boy was admitted with an impression of mandibular osteomyelitis and intravenous Clindamycin at 20mg/kg and Povidone Iodine mouthwash were started. Purified protein derivative skin test yielded a positive result of 5mm after 48 hours. Chest radiographs showed hazy opacities in the right upper lobe, likely consolidation due to pneumonia or pulmonary mass with minimal fluid layering on right lateral decubitus position. The CRP was elevated at 12.64 (<10) and ESR was high at 70 (0-15) indicating systemic infection. Peripheral blood smears revealed microcytic anemia, white blood cell counts were normal with no blast cells seen and platelets were adequate.

Head and Neck contrast CT scans showed a peripherally enhancing hypodense mass with lytic expansion from the right angle down to the left body of the mandible. *(Figure 3A, B, C)* There were enhancing lymph nodes along the submental, submandibular, jugular, and

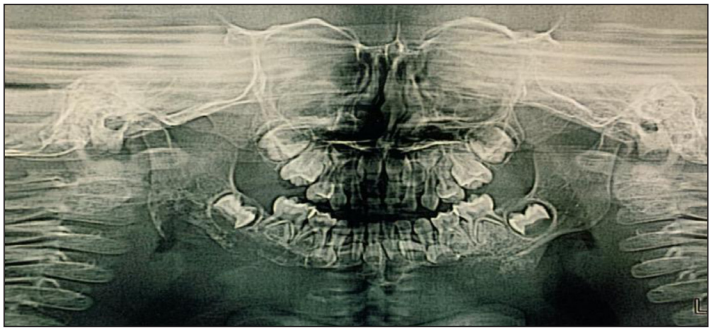

Figure 1. Panoramic radiograph showing osteolytic lesion from the right mandibular angle extending up to left mandibular body.

Figure 2. Gingival swelling from right mandibular second molar to canine with pinpoint fistula draining purulent discharge admixed with blood.

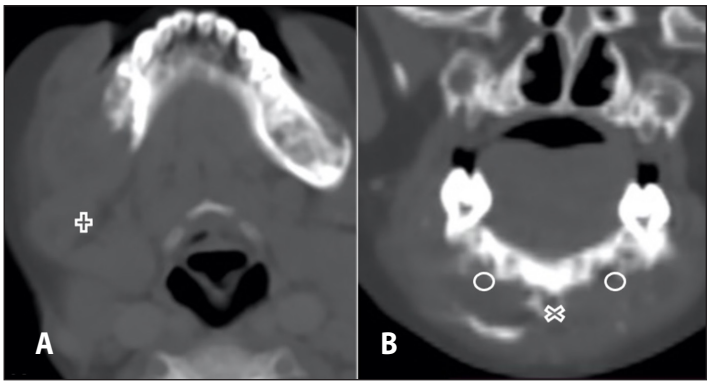

Figure 3. Head CT with contrast, **A.** axial and **B.** coronal bone window cuts showing peripherally enhancing hypodense mass with lytic expansion involving the right angle (+), symphysis (X), bilateral parasymphysis and bilateral mandibular body (O); **C.** 3D reconstruction showing lytic expansion involving the right mandibular angle, symphysis, bilateral parasymphysis and bilateral mandibular body.

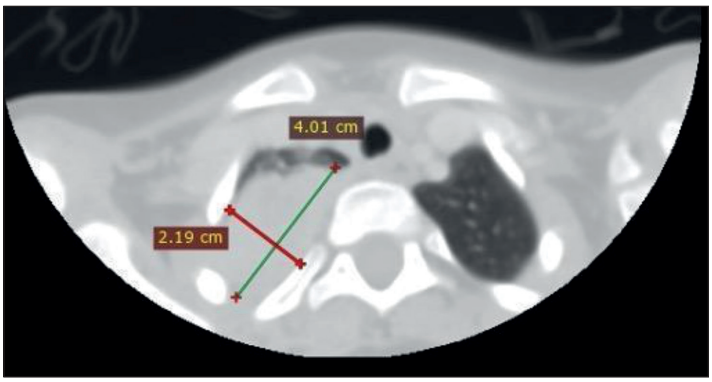

Figure 4. Neck CT scan with contrast, axial view, with incidental finding of an enhancing mass in the right lung apex measuring 4.01 cm x 2.19 cm.

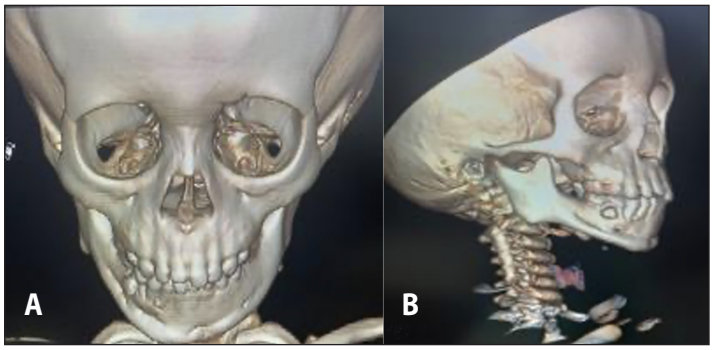

Figure 5. Facial CT scans, 3D reconstruction. **A.** anterior and **B.** right lateral oblique views showing longitudinal groove along the body signifying post infection changes.

posterior cervical chains with the largest measuring 2.2 cm x 1.68 cm and an incidental finding of an enhancing right upper lung mass. *(Figure 4)* Chest CT scan with contrast showed a 4.6 cm x 5.4 cm x 5.9 cm heterogenous enhancing mass in the right upper lobe and a 2.1 cm x 5.2 cm x 2.3 cm mass on the right pleura at the level of T7-T8 with lysis of adjacent ribs, and a lytic bone lesion in the right scapula.

Incision and punch biopsy intraoperative findings revealed granulation tissues admixed with yellowish cheesy materials. Fluid samples taken for Gram stain with aerobic culture sensitivity (GS/CS), KOH and AFB yielded negative results. Histopathologic examinations of the mandible and lung specimens showed caseous and non-caseous granulomas consistent with Koch's infection. Gastric aspirate sent for TB PCR yielded very low detection of MTB.

He was discharged on an anti-tuberculosis regimen under Direct Observed Treatment (DOT) with instructions for monthly follow-up. Chest x-rays requested for all household members were negative for TB.

Four months after starting treatment, there was still swelling of the mandible and intraoral pus with minimal blood. By the 7th month, he was asymptomatic, with good facial symmetry and no mandibular swelling. However, there were still hyperemic areas along the right mandibular gingival mucosa from canine to second molar. Repeat panoramic x-ray showed reduction in the bony defect with progressive bone deposition.

Eight months post treatment, facial CT scans showed bone deposition and remodeling of the body and angle of the right mandible and longitudinal groove along the body signifying post infection changes. *(Figure 5 A, B)* Chest CT scans showed decrease in the pleural based mass to 1.74 cm x 4.69 cm x 2.10 cm still associated with lysis of the adjacent T8 rib. The previously seen right upper lobe mass now measured 2.91 cm x 3.91 cm x 4.19 cm. No lytic lesion was seen in the scapula.

Currently, he is compliant with follow-up, with good nutritional status (Z-score of 0, based on WHO weight-for-age and height-for-age, 2-5 years old) and regularly taking his medications to complete a 12-month course.

DISCUSSION

Around 1 million children (<15 years) suffer from TB worldwide and more than 136,000 die each year with 75% occurring in low income countries.[4] The Philippines has one of the highest TB incidence rates in the region with around 554 cases per 100,000 in 2016.[3] Children under 5 years of age are at high risk of developing clinical TB after infection and are prone to developing severe forms of TB such as meningitis and disseminated TB particularly if not protected by Bacillus Calmette-Guerin (BCG) vaccination.

Extra pulmonary TB diagnosis is usually made based on history, clinical presentation, radiography, sputum analysis, histopathology, and serological investigations. We presented the case of a child with a slow, progressively enlarging, non-tender mass on the mandible with associated on and off low-grade fever and bloody to purulent discharge from the crown of the mandibular 2nd molar. In the two published cases of the EPTB of the mandible, the 33-year-old man presented with preauricular swelling associated with trismus[6] and the 10-year-old child presented with painless mandibular swelling and draining sinuses.[5]

Laboratories revealed elevated CRP, high ESR, and microcytic anemia pointing to a systemic infection. Gold standards for detection of tuberculosis are culture sensitivity, histology, serological testing, and TB PCR. Gastric aspirate TB PCR showed very low detection for MTB, and GS/CS, KOH and AFB also yielded no growth. Several factors may contribute to a negative result such as poor collection technique, an aerobic bacterium not isolated from the specimen or a different pathogen mimicking bacterial infection.

Radiologic workups exhibited an ill-defined radiolucency with sclerotic borders and osteolytic changes on panoramic x-ray, and CT scan findings of a peripherally enhancing hypodense mass with lytic

expansion involving the right angle down to the left body of the mandible suggestive of a nonspecific osteomyelitis, with an incidental finding of a lung mass leading us to perform tissue biopsy. Results showed caseating and non-caseating granuloma consistent with Koch's infection.

Tuberculosis infection may disseminate through direct transfer from infected sputum, regional extension of soft tissue lesion to involve the underlying bone or through hematogenous spread,[7] resulting in involvement of the mandible in our case. The mandible is said to be the second most common site of TB infection.[8] Tuberculosis of the mandible is rare because the mandible contains less cancellous bone, and the infection is thought to begin in the cancellous portion.[8] Involvement of the mandible may be due to its blood supply and bone density. The mandible is a dense bone and is solely supplied by the inferior alveolar vessels. Infection can cause decrease in blood supply leading to necrosis which makes it susceptible for bacteria to grow.[7] Tuberculosis of the mandible could present with a variety of signs, including periodontitis with horizontal bone loss, apical osteitis and widespread destructive lesions and sequestrations which were observed in our patient.[9]

In our case, the patient was fully immunized but still developed disseminated TB. Factors in developing disseminated TB include malnutrition, poor dental hygiene, and family history of unknown abscess formation. The disease may have started in the lungs as is more common and since there were dental caries, these may have been the route of infection for spread in the mandible.

Our patient showed drastic improvement by the 7th month of treatment with a DOT anti-tuberculosis regimen. He remains compliant with the treatment regimen which will be completed for 12 months.

In summary, tuberculosis can manifest in various ways, and diagnosing extrapulmonary TB remains a great challenge, hindering early detection. Multifocal TB can present as simple mandibular swelling, and a thorough workup should look for other involved sites. Early diagnosis in children may prevent debilitating sequelae and improve long-term treatment outcomes. A thorough history, complete physical examination, and appropriate diagnostics are key to diagnosis and treatment. The proper anti-TB therapeutic regimen is important and long term follow up is warranted especially in children because TB can remain inactive for years.

REFERENCES

1. Sambyal SS, Dinkar AD, Jayam C, Singh BP. Primary tuberculous osteomyelitis of the mandible in a 3-year-old child. *BMJ Case Rep.* 2016 Sep 21;2016:bcr2016216854. DOI: http://dx.doi.org/10.1136/bcr-2016-216854; PubMed PMID: 27655878; PubMed Central PMCID: PMC5051373.
2. World Health Organization. Tuberculosis. 14 October 2021. [cited 2022 February 27] Available from https://www.who.int/news-room/fact-sheets/detail/tuberculosis.
3. Snow K, Yadav R, Denholm J, Sawyer S, Graham S. Tuberculosis among children, adolescents and young adults in the Philippines: a surveillance report. *Western Pac Surveill Response J.* 2018 Nov 9;9(4):16-20. DOI: 10.5365/wpsar.2017.8.4.011; PubMed PMID: 30766743; PubMed Central PMCID: PMC6356044.
4. Kiat MO. Clinical profile and treatment outcomes of childhood extra-pulmonary tuberculosis in a children's medical center. *PIDSP Journal.* 2017 Jul-Dec; 18(2):3-15.
5. Romualdez J, Dualan M. Mandibular osteomyelitis: An unusual form of tuberculosis. *Philipp J Otolaryngol Head Neck Surg.* 1999 April-June;14(2):36-43.
6. Santos JM, Reala ET. Tuberculosis of the temporomandibular region. *Philipp J Otolaryngol Head Neck Surg.* 2018 Jul-Dec;33(2):41-4. DOI: https://doi.org/10.32412/pjohns.v33i2.275.
7. Karjodkar F, Saxena VS, Maideo A, Sontakke S. Osteomyelitis affecting mandible in tuberculosis patients. *J Clin Exp Dent.* 2012 Feb;4(1):e72–e76. DOI: 10.4317/jced.50588; PubMed PMID: 24558529; PubMed Central PMCID: PMC3908814.
8. Towdur GN, Upasi AP, Veerabhadrappa UK, Rai K. A rare, unusual presentation of primary tuberculosis in the temporomandibular joint. *J Oral Maxillofac Surg.* 2018 Apr;76(4):806-811. DOI: 10.1016/j.joms.2017.09.010; PubMed PMID: 29031528.
9. Kakisi OK, Kechagia AS, Kakisis IK, Rafailidis PI, Falagas ME. Tuberculosis of the oral cavity: a systematic review. *Eur J Oral Sci.* 2010 Apr;118(2):103-109. DOI:10.1111/j.1600-0722.2010.00725.x; PubMed PMID: 20486998.