Case series of extra pulmonary tuberculosis presenting as facial swelling

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IN BRIEF

- Discusses three cases of tuberculosis that initially presented as facial swellings.
- Demonstrates the importance of considering tuberculosis as a differential diagnosis.
- Gives an overview on the pathology, management and epidemiology of tuberculosis.

In England there were 7,290 cases of tuberculosis (TB) reported in 2013. The area with the highest incidence of the disease in England is London, with hotspots in other urban areas. TB affecting the head and neck is rare. We present three such cases of TB presenting as pre-auricular swelling. Two of the patients were initially misdiagnosed as having dental infection, demonstrating the importance of taking a good history and considering the differential diagnosis of TB when appropriate. TB remains a potentially fatal disease that should be considered in patients presenting with facial swelling where common causes have been excluded.

INTRODUCTION

Tuberculosis (TB) is an infectious disease caused by Mycobacterium tuberculosis and is transmitted from an affected individual via air droplets. Initial symptoms include fever, night sweats, weight loss, fatigue and persistent cough. Primary infection usually occurs in the lungs,1 although can more rarely be in the pharyngeal tonsils.2 TB can then spread to other areas of the body via the blood stream. Bone TB affects approximately 1% of patients and is usually secondary to either renal or pulmonary infection.1 Presentation of TB can be with a 'cold abscess' where abscess formation is not accompanied by the usual signs of inflammation, such as heat and erythema.

Despite a decrease in the global incidence of TB since 2002, there were estimated to be 8.6 million new cases of TB in 2012, 13% of which were in HIV seropositive patients.³ In the same year there were 1.3 million deaths attributable to the disease, one quarter occurring in HIV seropositive patients.³ In the UK there were around 9,000 cases per year in 2005–2011.⁴ In the two years to 2013 the incidence of TB in England reduced to 7,290,⁵ 74% of UK TB patients were born outside of the country, and 39% of all UK cases in 2011 were in London.⁴ Urban areas account for most UK TB cases,

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Refereed Paper Accepted 8 April 2015 DOI: 10.1038/sj.bdj.2015.342 British Dental Journal 2015; 218: 519-522 with incidence more than three times the national average in London, Manchester, Birmingham, Leicester, Luton and Coventry.⁵

TB is a notifiable disease which needs to be reported to communicable disease control with subsequent contact tracing.6 Immunisation with the Bacillus Calmette-Guerin (BCG) vaccination in the UK is targeted at infants who have an increased risk of TB, for example those born, or with relatives born, in areas with a high incidence of the disease, or those with a positive family history within the last five years.6 Immunisation is also recommended for all healthcare workers who have direct patient contact who are not found to be immune.6 Although the BCG vaccine provides adequate protection for paediatric TB, it is ineffective for adult pulmonary TB, and is problematic in HIV seropositive individuals due to the risk of infection.3 Vaccination for adults migrating to the UK is therefore not the routine. To combat this ineffectiveness in adults, new vaccines are being developed,3 with investment from the UK government being targeted towards this goal.5 Treatment in TB is usually with a combination of three antibiotics, though there is increasing tendency to drug resistance,3,4 which is particularly problematic in HIV seropositive patients.5

Clinical manifestations of TB in the head and neck region may include non-healing sockets, abscess formation and chronic ulceration of the buccal mucosa, gingivae, lips and palate.⁷⁻¹⁰ When presenting as oral ulceration this can clinically mimic squamous cell carcinoma.⁹ An Indian study of 300 pulmonary TB patients found that only one patient had tuberculous ulceration of



Fig. 1 Case one MRI showing abscess

the tongue, demonstrating that this is a rare presentation. ¹⁰ TB has also been isolated in the mandible and cervical lymph nodes ⁹ and the tempromandibular joint (TMJ). ^{11,12} We describe three presentations of extrapulmonary TB causing facial swelling and discuss their relevance to practice.

CASE ONE

A 45-year-old Asian lady was referred by her GP to the ENT surgeons with a six-month history of painful right facial swelling, which had not responded to four courses of antibiotics. Her dentist had extracted her lower right wisdom tooth suspecting an odontogenic infection; however, this only served to exacerbate the signs and symptoms of pain and swelling. An MRI scan showed a 5 cm \times 4.3 cm area in the region of right TMJ with a cystic and solid component (Fig. 1). A CT scan with contrast showed multiple focal areas of abscess

affecting the right condyle and extending into the masticatory spaces. Following discussion in a local multidisciplinary team meeting, the patient was referred to the oral and maxillofacial surgeons. On direct questioning in the maxillofacial surgery clinic, there was a history of weight loss, night sweats and evening pyrexia. The patient had a large, firm, tender 5 cm swelling in the right preauricular region extending anteriorly towards the zygomatic arch. She also had marked trismus but no lymphadenopathy. A working diagnosis of extra-pulmonary tuberculous osteomyelitis was made. An orthopantomogram (Fig. 2) revealed destruction of the right condylar head and her chest radiograph was unremarkable. An aspirate of pus from the swelling was sent for urgent microbiology culture and sensitivity, which confirmed the diagnosis of tuberculosis. The patient was referred to the respiratory team who started her on rifampicin, isoniazid, ethambutol and pyrizinamide. At a six-month review the swelling had completely resolved, although the patient continued to suffer trismus. A follow up OPG at two years demonstrated remodelling of the right TMJ with abnormal morphology (Fig. 3), and the patient's trismus had completely resolved.

CASE TWO

A 19-year-old Pakistani male, referred to the oral and maxillofacial department by his GP, had a four-week history of spontaneous onset of pain and progressive swelling in the right preauricular area. In addition, he gave a history of cough and regular episodes of pyrexia. Examination confirmed a well-localised 3 cm soft fluctuant lump, just inferior to the right zygomatic region, overlying the parotid gland (Fig. 4). Differential diagnoses included sebaceous cyst, parotid tumour and extrapulmonary TB. Ultrasound scan showed a 24 mm × 15 mm predominantly cystic lesion, consistent with a sebaceous cyst. A volume of 4 ml of thin pus was aspirated and sent for microscopy and culture. As TB was suspected, a Mantoux test and chest radiograph were also requested to investigate the possibility of pulmonary involvement. A CT scan of the neck suggested a necrotic lymph node, focal abscess or, less likely, a parotid neoplasm. Mantoux test was strongly positive and blood tests showed elevated erythrocyte sedimentation rate. His chest radiograph was unremarkable. An urgent referral was made to the Chest clinic where, following the microbiology report, a diagnosis of extra-pulmonary tuberculosis was made. The isolate was identified as M. tuberculosis. Treatment was



Fig. 2 Case one orthopantomogram



Fig. 3 Case one follow-up orthopantomogram



Fig. 4 Case two on presentation

commenced with a regime of rifampicin, isoniazid, ethambutol and pyridoxine after confirming sensitivity to these antibiotics. Six months later he had no symptoms of active TB and his facial swelling had completely resolved.

CASE THREE

A 23-year-old male of Pakistani origin was referred to the oral and maxillofacial surgery team by the accident and emergency department with a dental abscess, reporting a one-month history of a gradually enlarging swelling over his right cheek, difficulty opening his mouth and pain on biting. He also reported one day of dental pain a week



Fig. 5 Case three on presentation

before his presentation to accident and emergency. The patient's signs and symptoms had not responded to flucloxacillin prescribed by his GP. On examination he had a diffuse, tender facial swelling overlying the right mandibular ramus (Fig. 5), with mouth opening of 1 cm. There was no erythema overlying the swelling. An orthopantomogram (Fig. 6) revealed gross caries in 14, 46 and 36, and the patient was admitted for treatment of a dental abscess with intravenous antibiotics. On further review of the radiograph it was noted that the right condyle was eroded which was confirmed on a subsequent CT scan. A working diagnosis of osteomyelitis secondary to TB was made.



Fig. 6 Case three orthopantomogram

A chest radiograph demonstrated hilar lymphadenopathy and the patient was referred to the respiratory team, who started antituberculous medications. An aspirate from the buccal swelling cultured *M. tuberculosis*. The diagnosis of chest and bone TB was confirmed. The patient attended clinic on several occasions for drainage from the swelling following commencement of TB treatment. Following two months of treatment on TB triple therapy (rifampicin, isoniazid and pyrazinamine) the buccal swelling reduced (Fig. 7) and the patient's condition is continuing to improve.

DISCUSSION

Although TB is resulting in fewer deaths, with a decrease in mortality of 42% between 1990 and 2012,3 it still represents a significant global health problem. The distribution of TB cases worldwide is such that the burden of disease is borne by low and middle income nations, accounting for 95% of TB deaths.13 This presents itself in the UK as an higher incidence of TB in migrant populations, as is seen in all patients in our case series. As London has the biggest migrant population in the UK, with 37% of residents being born outside the country in 2011,14 it is no surprise that 39% of all TB cases in the UK were in London in 2010.4 These factors demonstrate the importance of patient demographics and contacts when considering differential diagnoses for patients presenting with facial swellings.

None of the patients in this series had a BCG vaccination at birth, and all were born outside the UK. Although all patients in this case series gave a positive history of foreign travel, they had no recollection of TB contact. Screening questions to identify those at increased risk of latent TB infection (LTBI) is recommended for individuals coming into the UK from high-risk areas, although the implementation of screening has been recently been found to be variable and

ill-coordinated.¹⁵ Those identified as high risk by the screening then progress to have testing for TB. A recently published Public Health England document outlines strategies for improving screening to help with identification of LTBI to contain the spread of the disease.⁵ This includes dissemination of information about the disease to those who are entering the UK from a high incidence area who are not required to undergo active tests.⁶

The three cases presented show similarity; none having a previous TB diagnosis, all presenting with preauricular swelling which had no signs of inflammation evident. Two of the patients had significant trismus due to condylar destruction as a result of tuberculous osteomyelitis following likely haematological spread of the bacterium. These signs and symptoms have previously been reported in the literature in cases of TB of the TMJ, 11,12 although one of these previously reported cases was not proven. The same two cases in this series had initially been misdiagnosed as odontogenic infections, and one patient had her third molar extracted as a result of this error, highlighting that TB should be considered as a differential diagnosis in facial swellings when common causes have been excluded. The importance of dentists considering TB has been highlighted in a previous case report of oral ulceration;7 however, it is important to be aware of other head and neck manifestations of this disease, including facial swelling.

When a diagnosis of TB is made, or clinical suspicion is high enough to warrant treatment, the relevant consultant in communicable disease control⁶ (a public health consultant) must be notified within three days. Globally, under-reporting is a significant problem that hampers progress in disease control.³ In these cases the respiratory physicians who took over the management of the patients arranged notification to the relevant body. Prompt referral to our medical



Fig. 7 Case three following two months of treatment

colleagues is essential to ensure that patients are managed appropriately as TB is not a common presentation to a dentist or an oral and maxillofacial surgery department. The NICE guidance requires contact-tracing to take place for all new diagnoses, depending on their current work, school and family situation. This is in order identify any related cases, to prevent further spread of the disease, and is carried out by the local public health bodies.

CONCLUSION

These cases demonstrate extra-pulmonary TB presenting initially as facial swelling. Patient demographics should alert clinicians to consider TB as a differential diagnosis, in particular in immigrants from, and patients who have travelled to, high incidence areas. Suspicion should be raised when there is a facial swelling where common causes have been excluded, when it is refractory to antibiotic treatment or when a swelling is not accompanied by signs of inflammation ('cold abscess'). Once TB is suspected a case history for TB should be taken, although the absence of lung symptoms or a positive contact history does not exclude TB as a potential cause. Clinicians should be aware of the risk to healthcare workers of TB contact and the importance of prompt referral and liaison with other healthcare professionals when a diagnosis of TB is suspected.

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