Rare disease

Warthin's tumour with coexistent tuberculosis

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Summary

A 81-year-old man presented with a 2-month history of swelling of the right angle of the jaw. On histopathological examination a diagnosis of Warthin's tumour with coexistent tuberculosis of the parotid gland was made. PCR for tuberculosis was positive. Tuberculosis of the parotid gland is very rare and coexistence with Warthin's tumour is extremely rare. This case is being reported for its rarity.

BACKGROUND

Tuberculous infection of the parotid gland is rare and its coexistence with Warthin's tumour is even rarer. Only a very few isolated cases have been reported in the literature. Coexistence of tuberculosis of the parotid gland with mixed parotid tumour is more common than with Warthin's tumour. We present a case of tubercular infection of the parotid gland in a patient with Warthin's tumour.

CASE PRESENTATION

A 81-year-old man presented with a 2-month history of swelling at the right angle of the jaw. The swelling measured 5×3 cm and was firm and overlying skin was normal. Systemic examination was unremarkable. Haemogram showed haemoglobin 10 gm%, total leucocyte count 8000 cells/mm³ and differential leucocyte count P₄₀ L₅₃ E₀₄ M₀₃ B₀₀; the erythrocyte sedimentation rate was 33 mm in the first hour. Chest x-ray showed calcified hilar lymph nodes with prominent bronchovascular markings. Fine needle aspiration cytology of the parotid gland showed clusters of epithelioid cells, lymphocytes, occasional Langhan's giant cells (figure 1A) and clusters of oxyphilic cells (figure 1B). PCR for tuberculosis was positive. The swelling was excised and sent for histopathological examination.

The tumour measured 5×4.5×3 cm with an attached normal parotid gland. It was well encapsulated. The cut surface showed small cystic spaces filled with proteinaceous material.

On microscopic examination, oncocytic epithelial cells forming some papillary projections along with lymphoid aggregates were seen (figure 2). Multiple granulomas composed of epithelioid cells, lymphocytes and Langhan's giant cells were seen with focal areas of caseous necrosis. A normal parotid gland was also identified (figure 3). A diagnosis of Warthin's tumour with associated tuberculosis was made.

INVESTIGATIONS

Mycobacterial culture was positive.

TREATMENT

The patient was given antitubercular treatment.

OUTCOME AND FOLLOW-UP

The patient is responding to treatment.

DISCUSSION

Less than 3% of all neoplasms are primary salivary gland tumours. The majority of primary salivary gland tumours

Figure 1 (A) Fine needle aspiration cytology (FNAC) smear showing caseous necrosis, epithelioid cells, lymphocytes and Langhan's giant cells (Pap×250). (B) FNAC smear showing cluster of oxyphilic cells and lymphocytes (Pap×250).

Figure 2 Microphotograph showing oncocytic epithelial cells forming a papillary projection and lymphoid aggregates (H&E×125).

Figure 3 Microphotograph showing multiple granulomas composed of epithelioid cells, lymphocytes and Langhan's giant cells with focal areas of caseous necrosis. Normal parotid gland was also identified (H&E×50).

affect the parotid gland and most are benign. Some 15% of epithelial salivary gland tumours are Warthin's tumour, which is almost exclusively a tumour of the parotid gland.¹ It has a strong association with smoking^{2 3} and more

commonly affects men in their sixth and seventh decades of life. Warthin's tumour usually presents as an asymptomatic, slowly growing round or oval mass. The disease was named Warthin's tumour after the pathologist who

Table 1 Coexistent tuberculosis with Warthin's tumour

Study	Age (years)/sex	Diagnosis	Other focus of TB	Mycobacterial culture	PCR
Watanabe <i>et al</i> (2001) ⁹	75/female	Warthin's tumour with TB	Tubercular meningitis	Not available	Positive
Watanabe <i>et al</i> (2001) ⁹	78/male	Warthin's tumour with TB	No other focus	Negative	Negative
Wen <i>et al</i> (2008) ¹⁰	81/male	Warthin's tumour with TB	Bilateral lung nodules	Positive	Not available
Oh <i>et al</i> (2009) ¹¹	53/male	Warthin's tumour with TB	Pulmonary TB 25 years previously	Not available	Positive

TB, tuberculosis.

first published case reports in the American literature in 1929.

This tumour develops due to salivary gland heterotopia in peri- and intraparotid lymph nodes. During embryogenesis of the parotid gland, epithelial cells from the oral mucosa penetrate into lymphocyte-rich tissue. The late encapsulation of the parotid gland explains the occurrence of intraparotid lymph nodes and heterotopic salivary gland remnants entrapped in the lymph nodes.

Tubercular infection of the parotid gland is very rare,⁴ with the first case described by von Stubenrauch in 1894.⁵ The disease can reach the parotid gland either as an ascending infection through the salivary gland duct^{5 6} or through haematogenous/lymphatic spread from a distant primary focus.^{6 7} As Warthin's tumour contains lymphocyte-rich stroma, mycobacterium seeding can take place. Diffuse and circumscribed forms of parotid tuberculosis have been described. The diffuse form is the most common and consists of small and large caseous areas or abscesses of oedematous, indurated and friable parotid tissue. The circumscribed form is very rare and may take the form of a cold abscess or cyst.⁸ The majority of cases of parotid tuberculosis are diagnosed by histopathological examination of a surgically removed mass.

Hence, a coexistent disease in a benign lesion must be kept in mind during histopathological examination, especially in a developing country such as India where the incidence of tuberculosis is very high. A few cases of coexistent tuberculosis with Warthin's tumour are listed in table 1.

Competing interests None.

Patient consent Obtained.

Learning points

- In developing countries like India, mycobacterial infection is very common.
- This infection can affect any organ.

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