CASE REPORT

A case of disseminated tuberculosis with increased tumour markers and testicular involvement

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KEYWORDS
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Summary
We report a patient having disseminated tuberculosis (TB) with negative smear and culture for Mycobacterium tuberculosis, left testicular mass, and an increase in tumour markers; alpha-fetoprotein (AFP), CA-125, and CA-19-9. The patient was a 53-year-old Caucasian male, presented with night sweat, weakness, and weight loss. Radiological findings were compatible with tuberculosis, however, sputum, bronchoalveolar lavage and biopsy materials were negative for malignancy or tuberculosis. A testicular mass was detected during the physical examination and orchidectomy material revealed acid-fast bacilli containing caseating granuloma. The patient responded well to anti-tuberculous therapy; clinical and radiological improvements observed at the end of the treatment period. Tumour markers were also decreased.

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Description of case

A 53-year-old male was presented with a 1-month history of night sweat, weakness, and 2-months’ history of weight loss (15 kg). He was slim, pale and weak, and his body temperature was 37.8 °C on the day of admission. He was a heavy smoker and reported using approximately 20 cigarettes per day for the previous 40 years.

A 1 × 2 cm. left testicular mass was detected by physical examination, which was also confirmed with ultrasonographic examination. No abnormality was found during the bronchoscopic examination.

White blood cells were found to be increased (12,300/μL) in complete blood count. Erythrocyte sedimentation rate was 23 mm/h, and LDH level was 298 U/L. Urinalysis was normal. HBsAg, HCV and HIV antibodies were negative. Alpha-fetoprotein (AFP) was marginally elevated (9.6 ng/mL). CA-125 (412.7 U/mL), and CA-19-9
(93.33 U/mL) levels were obviously high (Table 1). Sputum, bronchoscopic lavage, and transbronchial biopsy materials were negative both for microorganisms and malignancy.

<table>
<thead>
<tr>
<th>Marker</th>
<th>Before anti-tuberculous therapy</th>
<th>After 2 months of therapy</th>
<th>After 6 months of therapy</th>
</tr>
</thead>
<tbody>
<tr>
<td>CA-125 (U/mL) (Normal: &lt;35)</td>
<td>412.7</td>
<td>89.7</td>
<td>6.4</td>
</tr>
<tr>
<td>CA-19-9 (U/mL) (Normal: &lt;37)</td>
<td>93.33</td>
<td>35.4</td>
<td>14.45</td>
</tr>
<tr>
<td>AFP (ng/mL) (Normal: &lt;8.1)</td>
<td>9.6</td>
<td>8.6</td>
<td>7.6</td>
</tr>
</tbody>
</table>

Bilaterally diffuse reticulo-nodular infiltrations were observed in posteroanterior chest radiograms (Fig. 1). Abdominal computed tomography (CT) indicated para-vertebral, paraaortic, and abdominal multiple

Figure 1 Initial posterior-anterior chest radiography showing bilaterally diffuse reticulo-nodular infiltration (A). Becoming almost normal following a 6-month-anti-tuberculous therapy (B). Initial thorax CT scan image showing bilaterally reticulo-nodular miliary infiltration (C). Almost all infiltrations being cleared at the end of anti-tuberculous therapy (D).
lymphadenopathies. Chest CT illustrated bilaterally diffuse reticulo-nodular pattern (Fig. 1).

After consulting the patient, with the department of urology, left orchidectomy was performed for histopathological diagnosis of the testicular mass. Histologic examination of the orchidectomy specimen revealed extended necrosis with caseous granulomatous inflammation of possible tuberculous etiology. Ziehl–Neelsen staining for acid-fast bacilli was positive.

An anti-tuberculous therapy, with isoniazid, rifampicin, ethambutol supplemented by pyrazinamide for the first 2 months, was started on the basis of pathologic findings. The patient became symptom free in the second month of the treatment. The abnormalities observed in chest radiography and in high resolution CT were almost completely resolved after 6 months (Fig. 1). Thus, the response to the treatment also confirmed the diagnosis as disseminated tuberculosis. Tumour markers decreased to the normal ranges at the end of the therapy as well (Table 1).

Discussion

Tuberculosis is the leading cause of death due to a single infectious agent worldwide. Although the term “miliary TB” has been traditionally used for a pathological and radiological description, currently all forms of progressive widely disseminated haematogenous TB are being called as miliary TB, even the classical pathological or radiological findings are absent. Many adult patients with progressive tuberculosis have accompanying medical conditions that cause their specific immune responses to wane. Our patient was not immunocompromised and he had no high risk condition associated with progressive tuberculosis.

Generally, when M. tuberculosis is identified in any specimen, other systems of patients with suspected tuberculosis are not tested. As a result of this inclination, involvement of multiple organs with TB is probably much more common than it is recognized; therefore, most of them are overlooked. Abdominal involvement is a rare form of TB. CA-125 may be used as a marker for diagnosis, and in the follow-up of the patients with abdominal tuberculosis. CA-19-9 is expressed in mucous cells of the bronchial gland and surface of the bronchial surface epithelium cells in benign pulmonary diseases. It has been reported in several case reports that the level of CA-19-9 increased in various body fluids in patients with tuberculosis and decreased after anti-tuberculosis therapy. The authors suggested that CA-19-9 level may reflect the activity of pulmonary tuberculosis. CA-125 and CA-19-9 levels were found to be increased in our case. Plasma levels of these markers were started to decline after 2 months, and found within the normal levels at the end of anti-tuberculous therapy (Table 1). AFP level was only marginally elevated to 9.6 ng/mL and then decreased to 7.6 ng/mL over a six-month period. Therefore, it may not reflect the disease activity or response to therapy in tuberculosis. It has been known that tumour markers significantly decrease following orchidectomy in testicular tumour cases. In our case the elevation of tumour markers was considered as a reflection of disseminated tuberculosis, so tumour markers did not decreased immediately after orchidectomy.

Existence of a testicular mass was a coincidence that helped us to make the accurate diagnosis by an orchidectomy. It is not clear whether the actual cause of elevated levels of tumour markers is either abdominal or testicular involvement. To our knowledge this is the first in the literature where both tumour markers have been investigated simultaneously and found to be elevated in a patient with disseminated TB. We suggest that measurement of these markers may achieve a useful non-invasive tool in the diagnosis of disseminated tuberculosis. It may also be a useful to screen the response to anti-tuberculous therapy. However, it is sure that further studies in prospective design are needed to claim this hypothesis.

Conflict of interest statement

There is no conflict of interest for any of the authors of this case report.

References