Recurrence of Kikuchi’s lymphadenitis after 12 years

R W Blewitt, S N Kumar, J S Abraham

Abstract
A 40 year old woman developed recurrent Kikuchi’s disease 12 years after the original episode. The recurrence affected the same site (axilla) and occurred after the longest delay so far recorded in a European resident. Care must be taken to avoid misdiagnosis of Kikuchi’s disease as lymphoma.

Keywords: Kikuchi’s disease

Case report
A 40 year old woman presented with a lump in the left axilla, first noticed one month earlier. She had remained well, with no fever or other sign of systemic illness. On examination, a 2 cm mobile lymph node was found in the left axilla. There was no other palpable lymphadenopathy or hepatosplenomegaly. Investigation revealed a normal full blood count and a slightly raised ESR of 19 mm. Fine needle aspiration cytology of the lymph node showed small well differentiated lymphocytes and no malignant cells. One month later the lymph node was excised and submitted for histopathological examination.

In the laboratory, a diagnosis of high grade lymphoma was initially considered, but further examination and knowledge of the past medical history soon led to the correct diagnosis of recurrent Kikuchi’s disease.

Twelve years previously at a different hospital, the patient had had a similar lymph node removed from the same axilla. The specimen had caused some diagnostic concern, lymphoma again having been considered. Slides had been referred for expert opinion, through which a diagnosis of Kikuchi’s disease had been confirmed.

The patient has received no treatment and has remained well with no sign of further lymphadenopathy.

Histopathology
The excised lymph node was moderately enlarged (20 × 12 × 10 mm). Part of the lymph node contained reactive follicles, but the remainder was replaced by extensive areas of necrosis (figs 1, 2, and 3) around which were broad zones of blastoid lymphocytes (positive for CD3 and CD45RO). An abundance of karyorrhectic debris was present in the necrotic areas, as well as histiocytes with crescent shaped nuclei.

Review of the slides from the previous lymph node excision revealed very similar features. About two thirds of that lymph node was affected by nodular areas of coagulative necrosis, the remainder of the lymph node containing reactive follicles. The blastic cells were rather less numerous than in the latter biopsy.

These findings allowed a confident diagnosis of Kikuchi’s disease.

Discussion
Kikuchi’s disease (histiocytic necrotising lymphadenitis) is a benign self limiting condition that causes lymphadenopathy in young people, more commonly females than males. A flu-like illness frequently precedes the development of lymphadenopathy, which tends to be
localised in the cervical region. The disease has a much higher incidence in Oriental populations than in the United Kingdom, where it is quite rare.

Recurrence of lymphadenopathy is unusual, and has been estimated at about 3%. Recurrence has been recorded over a period of two to 10 years after initial presentation and there is a unique example of four recurrences over a period of 18 years in an Australian individual of Greek descent. Our case represents the longest delayed recurrence (12 years) in a European resident.

For the histopathologist, the most important point about Kikuchi’s disease is the danger of misdiagnosis as lymphoma. That Kikuchi’s disease can simulate lymphoma has been repeatedly noted since the first description of Kikuchi’s disease 27 years ago. The danger is more acute in European practice, where the disease is seldom encountered. A recent review of cases from a major British referral centre indicates that Kikuchi’s disease is still being misdiagnosed as lymphoma.

It is the sheets of blastic lymphoid cells that simulate lymphoma in Kikuchi’s disease. Careful attention to the clinicopathological features of Kikuchi’s disease, however, should be sufficient for confident diagnosis. In Kikuchi’s disease the lesions are based in the paracortex and usually involve only part of a lymph node. Frank necrosis may not always be present, but abundant karyorrhectic debris is invariably and may be seen in both blastic and necrotic areas, contrasting with an absence of neutrophils. The blastic cells are of T cell or histiocytic phenotype. The presence of histiocytes with crescentic nuclei is a helpful but not invariable feature. Kikuchi’s lymphadenitis is almost indistinguishable morphologically from systemic lupus erythematosus, which should therefore be excluded clinically.

In our case the history provided by the patient was invaluable. She was confident that the present lump would be the same as the previous one “which the doctors had found fascinating.”

In conclusion, it is important to remember Kikuchi’s disease as a rare cause of lymphadenopathy in young individuals. The disease can occasionally recur after long periods. This case, furthermore, reminds us of the wisdom of reviewing previous biopsy slides and paying close attention to the patient’s history.

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