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Clinical Record

Primary B-cell lymphoma presenting as bilateral ear lobule swelling

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Abstract

We report a rare case of primary B-cell lymphoma presenting as bilateral ear lobule swelling. A 56-year-old white man presented with a one-year history of painless swelling of both ear lobules. An excision biopsy confirmed B-cell lymphoma. Detailed systemic investigation confirmed the primary nature of the tumour. This tumour is rare in the ear lobule. A review of the English literature revealed no previously reported case of bilateral primary ear lobule involvement. Clinicians should be aware that this tumour can present as a primary in the ear lobules.

Key words: B-Cell Lymphoma; External Ear; Head and Neck Neoplasms; Diagnosis

Case report

A 56-year-old white man from north-west England presented to his general practitioner complaining of bilateral ear lobule swellings, which had developed over the previous 12 months (Figure 1). The swellings were painless and were not associated with any other ear symptoms. The patient did not have any symptoms of B-cell lymphoma at the time of presentation. The past history was insignificant.

On examination, both ear lobules had solitary, firm swellings approximately 2 cm in diameter. There were no signs of inflammation. The skin of the left ear lobule was irregular over the swelling and was not freely mobile, but there was no ulceration (Figure 2). The ear examination was normal. A differential diagnosis of fibroma or lipoma of the ear lobule was considered and excision biopsy of the nodules was planned.

In February 1999 (14 months after onset of the condition), excision biopsies were carried out on both ear lobules, and the specimens were sent for histological examination.

Histologically, both specimens shared morphological changes, demonstrating overlying skin with an underlying diffuse dermal infiltrate (Figure 3). This infiltrate consisted of small B-lymphocytes. These demonstrated strong positivity with the B-cell markers CD20 and CD79a, while staining negatively with markers for epithelial and Hodgkin's cells. The features were typical of a B-cell small lymphocytic lymphoma, according to the revised European–American lymphoma classification (Figure 4).

Detailed systemic investigation was performed in order to rule out involvement of any other areas. Blood counts and peripheral smear were normal. The patient had no lymphadenopathy or evidence of any focal lesion in the lungs or liver.

The patient was followed up. In June 1999 (18 months from onset), he developed a recurrence in the left ear lobule, and a repeat resection and reconstruction was performed. This was followed with combination chemotherapy, including 12 cycles (every four weeks) of

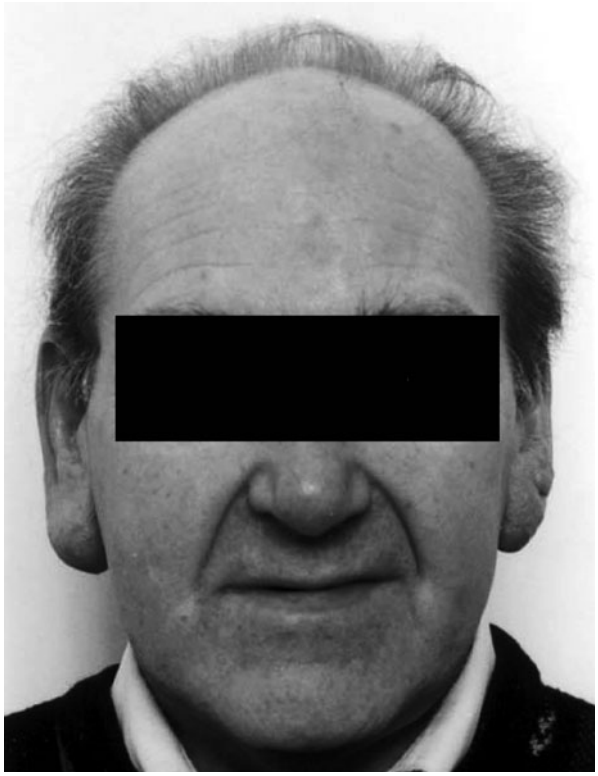

Fig. 1

Front profile of the patient, showing the bilateral ear lobule swellings.

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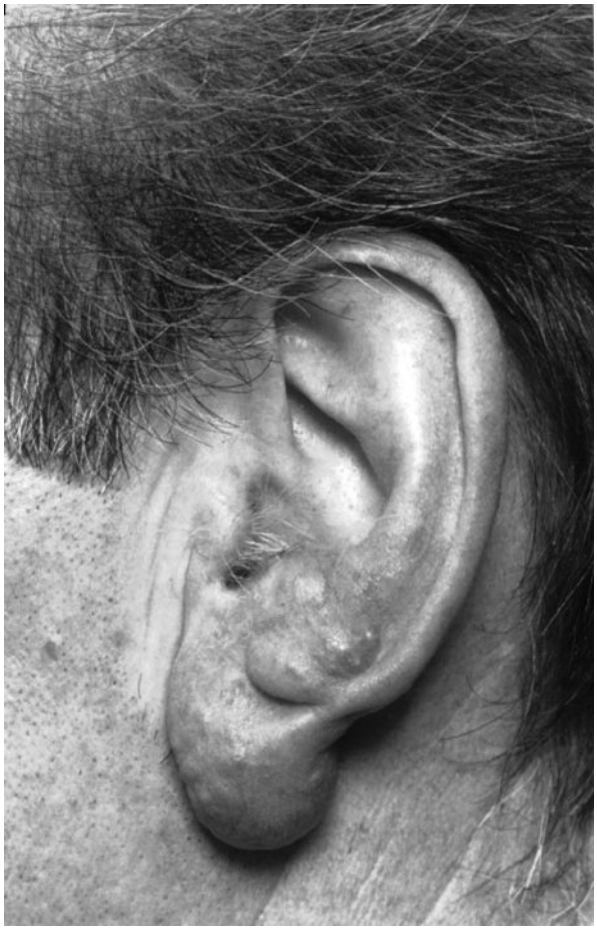

FIG. 2
Left ear, showing rounded, lobulated swelling of the ear lobule (note the similarity to otophyma).

chlorambucil with 5-fluorouracil. The patient was then disease free for three years (48 months from the time of onset of disease).

In November 2003 (12 + 57 months from onset), the patient again presented with a recurrence in the left ear lobule associated with skin ulceration. A single course of radiotherapy was given to the left ear lobule in December

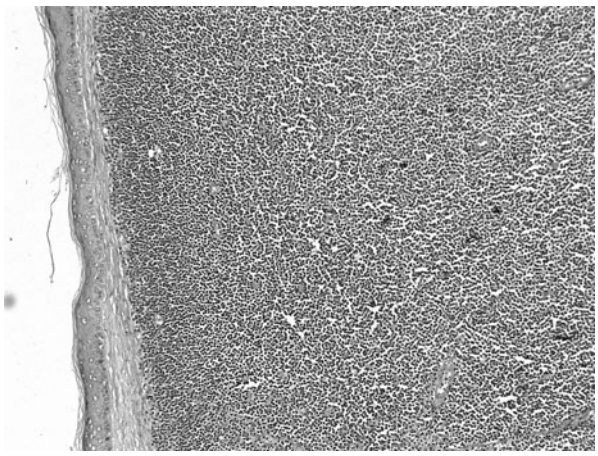

FIG. 3
Photomicrograph of the tissue, showing skin with lymphoma (H&E; ×100).

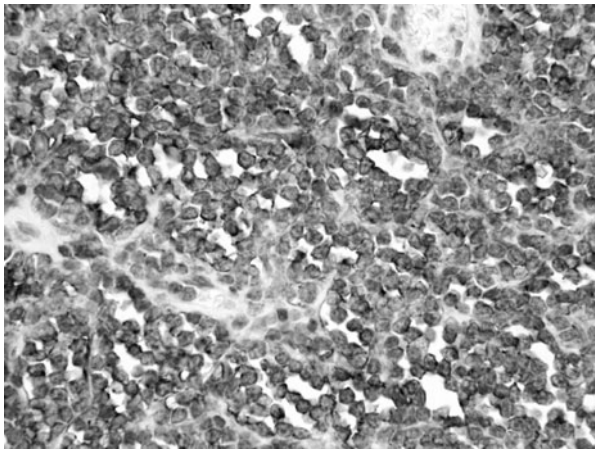

FIG. 4
Photomicrograph showing results of immunocytochemical analysis with B-cell marker L26 (×600).

2003 (60 months from onset). The lesion disappeared and the overlying skin healed.

Thereafter, the patient was regularly followed up. At the most recent follow up appointment, 96 months after initial presentation, he was disease free.

Discussion

B-cell lymphoma is a common condition affecting the head and neck region. However, the present case is the first reported incidence of B-cell lymphoma presenting bilaterally in the ear lobules as primary lesions. A few cases have been reported of B-cell lymphoma presenting as a primary in the pinna,[1–3] external auditory canal,[4–6] internal auditory canal,[7] temporal bone,[8–10] middle ear,[11] mastoids[12,13] and tympanic membrane.[14] Usually, such cases have been associated with symptoms referring to the ear. However, in the present case, the patient was symptom free apart from the local swelling, and there were no systemic features suggestive of lymphoma.

Non-Hodgkin's lymphomas of the pinna have been reported in both human immunodeficiency virus (HIV) positive and HIV-negative patients.[3,15] There have also been reported cases of HIV-positive patients with non-Hodgkin's lymphoma arising in the mastoid[12,13] and involving the tympanic membrane.[14]

The head and neck is the second most common site of extranodal lymphomas, after the abdomen. These can mimic other commonly occurring tumours in this region. The diagnosis of such extranodal lymphomas may require special techniques, such as the use of immunohistochemistry.

Generally, B-cell lymphomas show a good response to chemotherapy. However, it is possible for these tumours to relapse even after a complete cycle of chemotherapy, and for local radiotherapy to be required, as in our case. Thus, we recommend that such cases be followed up for any recurrence, even after chemotherapy.

Non-Hodgkin's lymphoma is treated with chemotherapy and radiotherapy, but the initial presentation is generally to other specialities. In cases of non-Hodgkin's lymphoma of the head and neck region, initial presentation to an otorhinolaryngologist is highly likely. Otorhinolaryngologists should thus consider non-Hodgkin's lymphoma in the differential diagnosis of atypical lesions of the head and neck.

Such symmetrical (malignant) lesions may easily be mistaken for otophyma. Thus, we recommend that all such lesions be subjected to biopsy in order to ascertain their

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histological diagnosis. In addition, the clinician should be prepared to repeat the biopsy if the clinical picture does not correlate in any way with the histological diagnosis.

- **This paper reports a rare case of primary B-cell lymphoma presenting as bilateral ear lobule swellings**
- **The symmetrical presentation of this malignant lesion may mean that it is easily mistaken for an otophyma**
- **All such lesions should be subjected to biopsy in order to ascertain the histological diagnosis. The clinician should also be prepared to repeat the biopsy if the clinical picture does not correlate in any way with the histological diagnosis**

Such cases present a challenge when planning the treatment regime. In our patient, radiotherapy was not initially a good option (because of bilateral involvement), and he therefore underwent chemotherapy. However, despite 12 complete cycles of chemotherapy, the patient suffered a recurrence and therefore underwent radiotherapy. Hence, it is important in such cases that an early diagnosis is made, followed by a suitable treatment regime, to enable a better prognosis.

Conclusion

Bilateral involvement of the ear lobules may be caused by otophyma or a keloid following fibroma excision. Otophyma can be diagnosed clinically by the thick, sebaceous discharge expressed from the swelling. Otophyma is part of acne rosacea, a general facial skin condition. A patient with keloid usually has a history of trauma. The absence of such features should prompt the clinician to perform a biopsy for histological examination. The finding of lymphoma in our case highlights the need for histological confirmation of all atypical, bilateral ear lobule swellings.

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