Recurrent pseudolymphomatous reaction to ear piercing: 20-year history

To the Editor,

Complications of ear piercing are varied and a pseudolymphomatos reaction is amongst the rarest presentations. A 48-year-old female presented with bilateral earlobe swellings of some years duration, clinically thought to be possible keloids following ear-piercing. A history of earlier surgery was not disclosed. She had mild hypertension and no significant medical history. Full thickness excision and primary closure was performed. Re-piercing was performed 5-months following surgery and 2 weeks following this, she represented with apparent acute infection with expressible pus. Studs were removed and flucloxacillin antibiotics administered, orally and topically along with topical 1% hydrocortisone. Repeat re-piercing was advised against.

Fleshy skin samples were received from left (measuring 20×12×10 mm) and right (17×10×8 mm) pinnae (Fig. 1). Histologically, sections of both were similar and displayed hair bearing skin and subcutis with a florid lymphoplasmacytic infiltrate, and non-caseating granulomata variably containing psammomatous calcification and a little black pigment. There was some scarring and a dilated hair follicle and/or squamous lined tract. A grenz zone was noted. Stains for microorganisms (Gram, periodic acid–Schiff with diastase, Grocott and Ziehl–Neelsen) were negative. Immunomarkers were performed. There was an admixture of T-cells (CD3+) and B-cells (CD20+, CD79a+) with T-cells predominating. Histiocytes including forming granulomata were prominently highlighted by CD68. S100 highlighted frequent scattered Langerhans cells, also highlighted by CD1a, and dendritic cells. There were scattered small numbers of CD30 positive activated lymphoid cells. T-cells had a CD4+CD8 ratio of approximately 1:1. There was expression of pan T-cell markers CD2 and CD5, but CD7 expression appeared reduced, most notably on the left. B-cells were arranged in primary and secondary follicles. The appearance was interpreted as reactive/pseudolymphoma, which was logical given the bilateral presentation at this site and with a foreign body type reaction to apparent metallic debris derived from ear piercing studs.

Multiplex PCR and differential fluorescence detection was subsequently performed on the larger left sample and was negative for the detection of clonal immunoglobulin heavy chain and kappa light chain gene rearrangements and clonal T-cell receptor beta chain gene rearrangements. However, it was positive for the detection of clonal T-cell receptor gamma chain gene rearrangements. Review was performed by a haematopathologist who concurred that given the mixed infiltrate, with scattered collections of foreign-body giant cells. There was no scarring noted. On review, the appearance was similar to the more recent samples.

The external ear comprising the auricle and ear canal may be affected by a variety of different skin lesions and dermatological conditions. They can be either solitary lesions which are locally limited to the ear or be part of a generalised dermatological condition. They may afflict skin and adnexa, cartilage and vessels. Potential complications of ear piercing may include an allergic reaction, auricular perichondritis, embedded earrings, infection, keloid formation, perichondral abscess and traumatic tear, pyogenic granuloma and pseudolymphoma.4-6

Cutaneous pseudolymphoma (CPL)/cutaneous lymphoid hyperplasia, is described in the literature as a reactive lymphoid proliferation that histopathologically and/or clinically imitates cutaneous lymphoma. However, it is suggested that use of the term CPL should be restricted to cases that histopathologically simulate cutaneous lymphomas and do not fit into any other diagnosis after clinical correlation. Clinicopathological correlation is essential. There are a wide range of aetiological agents: infection (notably Borrelia species), a broad range of drugs and foreign agents (including vaccination, piercing and tattooing), and miscellaneous causes such as insect bites, medicinal leech exposure and UV radiation, yet a number of cases remain idiopathic.7 Pseudolymphoma most commonly occurs in skin but indeed may occur in any organ. In skin, the most common aetiological agents are drugs followed by tattoo reactions. Clinically it may present as papules, infiltrated plaques and nodules and less frequently as persistent erythema or exfoliative erythroderma. In distinguishing CPL from lymphoma, multiple nodules or plaques would more suggest a suspicion of lymphoma, although the latter may also occur in mixed type B- and T-cell CPL.8 Histological features that suggest lymphoma over CPL include: bottom heavy infiltrate (deep infiltrate down to the subcutaneous fat); monomorphic infiltrate; or high-grade atypical lymphocytes (changes in nuclear morphology, changes in nuclear density, changes in nucleus/cytoplasm ratio). Immunohistochemistry and clonality studies may aid in diagnosis, such as demonstration of mixed B- and T-cells, and polyclonality of plasma cells, B- and T-cells, although the diagnosis may remain inconclusive.9

Mitteldorf and Kempf1 proposed a classification which divides CPL into four main groups based on histopathological features and clinical presentation: (1) nodular CPL, the largest group categorised by solitary or multiple nodules, including B-cell, Borrelia-associated, T-cell and mixed, and CD30+ CPLs; (2) CPLs simulating mycosis fungoides or other cutaneous T-cell lymphomas; (3) other, comprising distinct clinical entities such as T-cell rich angiomatoid pseudolymphoma; and (4) intravascular pseudolymphoma. Histological presentation can be divided according to different patterns: (nodular, epidermotropic, dermal diffuse, (measuring 15×10×5 mm) and right (10×8×3 mm) were received with a history of lesions associated with ear-piercing, keloids. Histologically, they were reported as showing a dense dermal lymphoplasmacytic inflammatory cell infiltrate, with scattered collections of foreign-body giant cells. There was no scarring noted. On review, the appearance was similar to the more recent samples.
subcutaneous, intravascular); principal cell morphology (anaplastic, centoblastic, centrocytic) and lymphocyte size (small, medium, large); immunophenotype (B-cell, T-cell, CD4+, CD8+, CD30+); and composition of infiltrate (admixed plasma cells, histiocytes, eosinophils, neutrophils). Molecular clonality studies require interpretation in clinical context because T- and B-cell clonality occurs not only in lymphoma, but may also arise in inflammatory conditions, infection and in pseudolymphoma. Diagnostic work-up includes a full medical history including drug, infective and foreign agent exposure and blood assessment which may include *Borrelia* and viral serology. Staging is
reserved for ambiguous cases. The most effective treatment is to remove the causative agent and to avoid re-exposure. Clinical course varies from a duration of weeks to years and it may resolve spontaneously and recur unpredictively. Treatment of CPL should be tailored to any known aetiological agent and may include intralesional steroid injection and surgical removal. The current case is mixed pattern pseudolymphoma, with a strong clinical association with ear piercing. Earlobe pseudolymphoma secondary to ear-piercing was first described in 1989 by Zilinsky et al. in a female with a 9-year history of bilateral earlobe masses arising soon after piercing. Allergic contact dermatitis secondary to metals is well recognised and more common than rare CPL. Laftah et al. described two cases of bilateral earlobe swelling with persistence in one case despite cessation of earring use. As in our case, both were mixed B- and T-cell infiltrates with accompanying polytypic plasma cells and eosinophils and had a differential diagnosis of cutaneous marginal zone lymphoma. The presence of histologically identifiable metallic debris (like in our case) was not mentioned, but x-ray microanalysis detected iron, nickel and titanium in one and gold and zinc particles in the other. Persistence of dermal metallic fragments may be directly immunogenic and trigger a delayed direct hypersensitivity reaction and lymphoid infiltration. Purported bilateral earlobe lymphoma has been documented in case reports and the presence of nodal or systemic disease would more support this. Although environmental factors may trigger disease (e.g., Helicobacter pylori and extranodal marginal zone lymphoma), equally, environmental factors may induce pseudolymphoma; therefore, monoclonality should not be taken as evidence of lymphoma as is demonstrated in the current case.

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Mark James Wilsher, Joe Marais

1 North West London Pathology, NWLP, Imperial College Healthcare NHS Trust (ICHNT), Department of Histopathology, Charing Cross Hospital, London, UK; 2 Unilabs HIS, London, UK; 3 Clementine Churchill Hospital, Sudbury Hill, Marlow, UK

Contact Dr Mark James Wilsher.
E-mail: markwilsher@gmail.com


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