Cutaneous plasmacytoma adjacent to Bowenoid actinic keratosis on the scalp: Is there a link?

A. Mosea*, M. Millwaters

The Princess Alexandra Hospital, Harlow, United Kingdom

A R T I C L E   I N F O

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A B S T R A C T

INTRODUCTION: Cutaneous extramedullary plasmacytoma without bone marrow involvement is very rare. We present a plasmacytoma on the scalp with an adjacent Bowenoid disease.

PRESENTATION: An 86 year old man presented to our unit with an ulcerated lump on the vertex of the scalp. Excisional biopsy showed plasmacytoma with adjacent Bowenoid actinic keratosis. Blood tests did not show any systemic multiple myeloma. However, skeletal survey showed possible osteolytic lesions in some areas. Six months afterwards, the patient remains well on follow up.

DISCUSSION: As far as we know, this is the first reported case of a cutaneous plasma cell tumour next to an area of Bowenoid actinic keratosis. Relevant literature is investigated here for possible correlation.

CONCLUSION: Within the limitations of this study, solitary primary cutaneous plasmacytoma can be treated surgically with a favourable outcome. A hypothesis of correlation between Bowenoid actinic keratosis and plasmacytoma is investigated here. Further research is needed to confirm this finding.

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1. Case report

We present an 86 year old man, who had a sore, weeping and rapidly growing ulcerated plaque on the vertex of the scalp. Otherwise the patient was well in himself without any weight loss or night sweating. Past medical history includes hypercholesterolaemia for which he is on statins. He denies smoking or drinking alcohol. There is no family history of any blood disorders.

On presentation, there was a 2.0 cm ulcerated lump on the vertex of the scalp. There was no lymphadenopathy, abdomen was soft, spleen and liver were impalpable. Initial punch biopsy showed actinic keratosis with epithelial dysplasia. Therefore, a bigger and deeper incisional biopsy was performed, which showed Lambda light chain monoclonal plasma cell infiltrate on immunostain.

Multiple myeloma screen followed. Results showed normal full blood count, renal profile, liver function test, serum M protein, total protein, albumin, globulin, SFLC was high (43.04) so as KL ratio (1.971) and B2 microglobulin (4.4). IgG, IgA and IgM were all normal. However, skeletal survey showed possible osteolytic lesions in different areas. The significance of this was not determined at that time so it was decided to repeat the above tests regularly to monitor the condition. Subsequent blood tests remain largely unchanged.

Earlier, the case was discussed at the MDT meeting in our hospital. The lesion in the scalp was excised under local anaesthetic (Fig. 1), the defect was repaired with a full thickness skin graft from the shoulder.

Definitive pathology confirmed a completely excised Iga/Lambda plasmacytoma with adjacent Bowenoid actinic keratosis (Fig. 2).

The patient had wound infection following the surgery, and he regularly attended wound dressing clinics. He remains well in himself sixteen months since the lesion started, with no evidence of recurrent disease or systemic involvement.

2. Discussion

Plasma cells are produced in the bone marrow and are found in the blood, skin and throughout the body. Plasma cell tumours have been reported over 100 years ago. Five types of which have been described: multiple myeloma (MM), solitary myeloma (plasmacytoma of bone), extramedullary plasmacytoma (EMP) and plasma cell leukaemia.

EMP applies to monoclonal proliferation of plasma cells outside the bone marrow. The upper airway passage is the most common site for EMP [1,2].

Cutaneous extra medullary plasmacytoma (cEMP) without evidence of bone marrow involvement (primary cEMP) is very rare [2,3]. They usually present as a solitary or multiple red nodule or a dome shaped skin elevation which may ulcerate [3–6].

EMP in general occurs between the fourth and the seventh decade of life and is more common in men [1–4]. Cutaneous involvement in a patient with MM (secondary cEMP) indicates

* Corresponding author at: The Princess Alexandra Hospital, Hamstel Road, CM20 1QX, United Kingdom.
E-mail address: akeel.mosea@pah.nhs.uk (A. Mosea).

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increased tumour burden in the body, hence an ominous sign. On the other hand, primary cEMP has a much more favourable outcome [2,3]. The size of the tumour and the clinical presentation (solitary vs multiple lesions) are the main prognostic factors in primary cEMP [4,6].

According to the International Myeloma Working Group [7], primary cEMP can be diagnosed by showing monoclonality of the plasma cells aggregate on immunohistochemical studies, absence of M-protein in serum and/or urine (although a small M-component may sometimes be present), normal bone marrow, normal skeletal survey and no related end organ damage.

Solitary primary cEMP may be treated conservatively by surgical excision with/out local radiotherapy [3,6,8].

3. Could have Bowens disease in our case contributed to the development of the plasma cell tumour?

Skin malignancy arising from sites of trauma, irritation, burns and old scars is a well known phenomenon first described in 1828. Marjolin's ulcers represent malignant degeneration arising in a pre-existing scar tissues, mainly old burn scars. These are usually squamous cell tumours but can be basal cell or melanoma [9].

Wiltshaw [1] explained the predilection of EMP for the submucosal tissue of the upper airways passages to recurrent local infections. Also implantation of foreign materials in susceptible animals can induce EMP in the affected animal [1].

Cases of cutaneous plasmacytoma developing in sites of local trauma have been described in literature [8,10,11]. Most of these tumours arise from the scars of previous surgery in patients with multiple myeloma (secondary cEMP) [10,11]. It is thought that plasma cells expressing CXCR4 cytokine receptor can interact with the inflammatory cascade released due to tissue injury, facilitating the migration neoplastic plasma cells into the skin [11,12].

Li et al. [8] reported a case of primary cEMP developing over the pacemaker implantation site one year after insertion. Initial investigations ruled out MM in that patient. The lesion was treated with radiotherapy after extraction of the pacemaker and the implantation of a new one on the opposite site. The patient subsequently developed additional cutaneous plasmacytoma over the new pacemaker insertion site and eventually died of progressive MM.

One can argue that the existence of Bowens actinic keratosis immediately adjacent to the plasma cell tumour in our case was a mere co-incidence, as this type of keratosis is common over sun exposed sites like the scalp. However, the irritation and skin inflammation associated with Bowens disease with all the surrounding ulceration and scarring could have had a tropical effect in attracting neoplastic plasma cells from the blood or bone marrow (secondary cEMP) or promoting neoplastic monoclonality of a local plasma cell population if there was no MM in the background (primary cEMP). Further research is needed to verify this hypothesis.

4. Method

This work has been reported in line with the CARE criteria.

Conflict of interest

The author has conflict of interests.

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Ethical standards

The manuscript does not contain clinical studies or identifiable patient data.

Consent

No identifiable patient information is included.

Authors contribution

A. Mosea Jordanian Board (OMFS), FFDRCSI (OSOM), MFDRCSI, BDS (writer).
M. Millwaters MBBS, BDS, FDS RCS Eng, FRCS (OMFS) (second writer).

Guarantor

Mr. Akeel Mosea, Dept. of Oral and Maxillofacial surgery, The Princess Alexandra Hospital, Hamstel Road, Harlow.

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