CASE REPORT

Subcutaneous sarcoidosis in a rhinoplasty scar

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SUMMARY

The presence of a subcutaneous hard bony-like lump at the lateral nasal wall after a septorhinoplasty procedure is an unfavourable result. The reported patient developed this complication 2 years after a revision surgery, in which percutaneous osteotomies were performed. An excision biopsy of the lump took place and the histopathological analysis revealed a granulomatous gigantocellular inflammation with absence of birefringent particles on polarised lamp and negative mycobacteria culture. After additional investigations, the final diagnosis was consistent with grade 2 pulmonary sarcoidosis associated with subcutaneous sarcoidosis. No treatment was initiated. The facial symptoms resolved without any additional treatment and the pulmonary function tests have not deteriorated after 1 year of follow-up. The polymorphism of cutaneous lesions in sarcoidosis, the absence of systemic symptoms and the unrecognised entity of subcutaneous sarcoidosis in a scar illustrate the diagnostic challenge with this patient.

BACKGROUND

Sarcoidosis is a common systemic disease of unknown aetiology specifically affecting the lungs, the lymph nodes and the skin, and is seen typically in Afro-American women in the third decade of their life. Skin is affected by sarcoidosis in at least 20% of cases and cutaneous lesions are the initial manifestation of the disease in a third of these patients.¹ The first descriptions by Besnier in 1899, followed by Boeck in 1899, were cutaneous forms of sarcoidosis. Schaumann later described non-caseating granuloma in different organs, indicating a systemic disease.² Cutaneous sarcoidosis lesions are described as specific or not specific according to the presence of non-caseating granuloma. In addition to many morphological variants, the manifestations are protean and illustrative of the great imitator nickname it deserves. It is suspected that the interplay between an extrinsic antigen and a disrupted T cell reaction in a genetically predisposed individual leads to the sarcoidal granuloma formation. Scar sarcoidosis is well documented as a frequent specific lesion. It typically presents with a raised, hard skin rash on the scar, different from the patient we present. Subcutaneous sarcoidosis is a rare entity, presenting as slightly tender subcutaneous lesions without skin surface lesions.

CASE PRESENTATION

A 34-year-old woman presented to the ear, nose and throat outpatient clinic with a slightly tender subcutaneous mass in the right paranasal region, which she first noticed 9 months ago. Her medical history was unremarkable except for chronic

rhinosinusitis and nasal deformity. She underwent functional endoscopic sinus surgery followed by a closed septorhinoplasty 4 years ago. The rhinoplasty was later revised by an open approach with percutaneous osteotomies because of an unsatisfactory result. On clinical examination, there was a bone-like mass of size approximately 4 cm located along the right frontomaxillary process region, extending within the medial aspect of the orbit ([figure 1](#)). Intranasal examination, skin inspection and eye function tests were unremarkable. No scar was visible.

INVESTIGATIONS

Full blood tests were unremarkable. The laboratory workup revealed an elevated ACE level (66 U/L, normal range 18–55 U/L). MRI revealed an unspecific subcutaneous infiltration of the premaxillary soft tissues, the lower eyelid and the internal canthus of 7 mm×3.8 cm×2 cm, without any extension into the orbit ([figure 2](#)).

A biopsy using an endoscopic approach was performed. The histological results were consistent with unspecific inflammatory changes. Excision of the mass was suggested. The mass was excised through an incision in the nasopalpebral crease down to bone ([figure 3](#)). Some granular inflammatory ill-defined tissues were exposed. Subtotal excision, sparing the skin, was performed. The histology revealed a gigantocellular and epithelioid granulomatous reaction ([figure 4](#)). There was no evidence of necrosis nor foreign body on

Figure 1 Right-sided lateral nasal wall subcutaneous mass with normal overlying skin.

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Figure 2 T1-weighted, fat suppression and postgadolinium injection images showing frontonasal aspecific tissue infiltration on the right side.

microscopic examination. The presence of infectious agents was excluded by special colouration (Gram, Grocott and Ziehl) and by mycobacterial PCR. The diagnosis of subcutaneous sarcoidosis in a scar was suspected. The performed pulmonary CT scan showed bilateral hilar lymphadenopathy associated with a discrete reticulonodular infiltrate. The analysis of transbronchial biopsies and bronchoalveolar sample confirmed the diagnosis of stage 2 pulmonary sarcoidosis. The pulmonary function tests were normal.

DIFFERENTIAL DIAGNOSIS

Foreign body granuloma, mycobacterial granuloma, subcutaneous sarcoidosis.

OUTCOME AND FOLLOW-UP

Ten months after the excision of the paranasal mass, the patient showed no clinical signs of recurrence. A control MRI revealed

Figure 3 Intraoperative view of the inflammatory tissue.

Figure 4 H&E colouration of the excised frontonasal mass. Presence of granuloma with the epithelioid and multinucleated giant cells. Note the absence of necrosis.

radiological persistence of the premaxillary soft-tissue infiltration with orbital sparing, but in a decreased form.

As the patient was asymptomatic, a clinical and radiological follow-up was decided (figures 5 and 6). To date, no sign of worsening of symptoms has occurred.

DISCUSSION

The lesions of cutaneous sarcoidosis are polymorphic in their presentation; despite scar sarcoidosis being considered as a frequent lesion, the diagnosis remains challenging. The literature reports scar sarcoidosis developing after minimal trauma to the skin, including tattoo, venipuncture, after herpes zoster virus infection and within fresh or old scars, but make hardly any note of a post-traumatic subcutaneous granuloma without skin lesion.³⁻⁶ One article reports the occurrence of a lump after a rhinoplasty in a female patient, diagnosed 12 years earlier with erythema nodosum, which they considered as a form of

Figure 5 Ten months postoperative views. Note the absence of the lump on front view. The three-fourth right-sided view showing minimal scar and redness of the overlying skin.

Figure 6 Ten months postoperative views. Note the absence of the lump on front view. The three-fourth right-sided view showing minimal scar and redness of the overlying skin.

subcutaneous sarcoidosis.⁷ They did not report precisely on the localisation of the granulomas. A second article reports the development of subcutaneous sarcoidosis with normal overlying skin, 10 months after a melanoma excision. The punch biopsy showed no histological changes of the epidermis and dermis.⁸ Whether the correct diagnosis for our reported case is subcutaneous sarcoidosis or scar sarcoidosis with subcutaneous granuloma cannot be fully answered as the skin was not excised. In the original description of subcutaneous sarcoidosis, by Darier and Roussy⁹ in 1904 and detailed in the paper of Vainsencher and Winkelmann¹⁰ in 1984, the authors emphasised that the epidermis and dermis were normal, and that the granulomatous process took place in the hypodermis, in the septa at the periphery of the fat lobule. In the largest review of 54 cases with subcutaneous sarcoidosis, the extremities were predominantly affected, with only 12 patients having lesions on their face or neck. Also, no history of trauma nor inoculation were described.¹¹

The association with systemic sarcoidosis is variable and should be looked for in any case. Some cutaneous sarcoidosis, such as lupus pernio, the verrucous and ichthyosiform variants are associated with severe pulmonary involvement.¹ Some papers report a strong association of pulmonary involvement with scar sarcoidosis as well as for subcutaneous sarcoidosis.^{10 11}

Treatment for cutaneous sarcoidosis is recommended when affecting the orbit or depending on the severity of physical deformity caused by the lesions. Topical or intralesional

corticosteroids are the first line of treatment for mild to moderate disease, whereas systemic corticosteroids are the first choice in severe cases. Intralesional triamcinolone was reported as efficient treatment for scar sarcoidosis.⁵ Conservative surgical excision is a valid alternative when a tissue sample is necessary.

A frequent complication, such as an unfavourable result after a cosmetic surgery, can indeed hide an uncommon form of a polymorphic systemic disease. Despite difficulty in ascertaining a definitive diagnosis in the present case, we would rather consider the reported patient as a form of subcutaneous sarcoidosis in the subcutaneous part of the scar.

Learning points

- ▶ Cutaneous sarcoidosis is frequent but subcutaneous sarcoidosis is rare.
- ▶ A subcutaneous lump can be the first sign of systemic sarcoidosis.
- ▶ Postoperative abnormal findings should not be regarded as the sole surgical complication but as potential systemic disease.
- ▶ Scar sarcoidosis may present without a visible scar.

Competing interests None.

Patient consent Obtained.

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