American Journal of Ophthalmology Case Reports 32 (2023) 101889

Contents lists available at ScienceDirect

American Journal of Ophthalmology Case Reports

journal homepage: www.ajocasereports.com/

Tattoo-associated lacrimal gland enlargement and sarcoidosis

Amritha Devi Kanakamedala, Robi N. Maamari, Steven M. Couch *

John F. Hardesty Department of Ophthalmology and Visual Sciences, Washington University School of Medicine, 660 S. Euclid Avenue, Campus Box 8096, St. Louis, MO, 63110, United States

ARTICLE INFO

Keywords:
Tattoo
Sarcoidosis
Lacrimal gland

ABSTRACT

Purpose: To describe a case of tattoo-associated sarcoidosis presenting with cutaneous findings of tattoo granulomas and ophthalmic manifestation of isolated bilateral lacrimal gland enlargement.

Observations: A 35-year-old female presented with bilateral upper eyelid swelling for over a year. She reported no associated episodes of ocular pain or visual decline since onset of eyelid edema. On examination, the lacrimal glands were firm and enlarged bilaterally. Slit-lamp examination demonstrated no evidence of active or prior ocular inflammation. Further systemic examination revealed multiple raised papules within a 4-year-old chest/shoulder tattoo. Histopathology from a lacrimal gland biopsy showed non-caseating granulomas consistent with sarcoidosis.

Conclusions and importance: The authors report a rare case of a 35-year-old presenting with isolated dacryoadenitis and tattoo granulomas found to be a tattoo-associated sarcoidosis. Although uveitis is a commonly described ocular manifestation in tattoo-associated sarcoidosis, few reports have described lacrimal gland enlargement as a presenting ophthalmic feature in tattoo-associated sarcoidosis.

1. Introduction

Sarcoidosis is an inflammatory condition of unknown etiology, resulting in formation of non-caseating granulomas that can affect any organ system, most commonly the lungs and lymph nodes. Ocular involvement is seen in 25% of patients with systemic sarcoidosis, and females have a higher rate of ophthalmic disease compared to males (56% vs. 23%).[1,2] While uveitis is the most common ocular manifestation, sarcoidosis can involve almost any ocular and periorbital structure, including the lacrimal glands.[1] Mikulicz disease was first described in 1888 and is characterized by xerostomia with parotid and lacrimal gland enlargement without a known underlying etiology. When these symptoms are attributed to a known underlying cause, including Sjogren syndrome, sarcoidosis, lymphoma, and tuberculosis, this condition is termed Mikulicz syndrome (MS).[3] While the causes of sarcoidosis are not well understood, tattoos have been associated with systemic and ocular sarcoidosis. This case report describes a patient with Mikulicz syndrome resulting from tattoo-associated sarcoidosis. Collection and evaluation of protected patient health information were HIPAA compliant. The report adhered to the ethical principles outlined in the Declaration of Helsinki as amended in 2013.

2. Case report

A 35-year-old previously healthy East Asian female presented with bilateral upper eyelid fullness and swelling for more than one year (Fig. 1A). She reported initial onset of right upper eyelid swelling associated with a superotemporal mass with subsequent development of similar findings of the left side a few months later. After 3 months with no observed improvement of the presumed chalazia using warm compresses and an oral antibiotic course, the patient was referred to the oculoplastic surgery service for evaluation. On examination, visual acuity, intraocular pressure, ocular motility, and anterior slit lamp and posterior segment examinations were normal without evidence of active or previous anterior or posterior chamber inflammation. She demonstrated bilateral upper eyelid fullness with enlarged lacrimal glands that were firm and tender to palpation. Further systemic examination was notable for multiple, circular, elevated skin papules overlying a 4-year-old chest/shoulder tattoo, concerning for granulomas (Fig. B). MRI of the face confirmed the bilateral dacryoadenitis and demonstrated bilateral parotid gland enlargement. Chest radiograph showed hilar adenopathy. Laboratory testing identified elevated angiotensin converting enzyme levels (76, range 10–55 units/L) and normal lysozyme, comprehensive metabolic panel, and complete blood count. Thyroid

* Corresponding author.
E-mail address: couchs@wustl.edu (S.M. Couch).

https://doi.org/10.1016/j.ajoc.2023.101889
Received 14 December 2022; Received in revised form 20 June 2023; Accepted 4 July 2023
Available online 13 July 2023

function panel was not obtained as thyroid related review of systems was normal. She underwent right orbitotomy with lacrimal gland biopsy, and histopathology identified non-caseating granulomatous inflammation consistent with sarcoidosis.

The patient was then referred to the rheumatology service and prednisone 40mg daily was initiated over a 3-month taper. She responded well with complete resolution of ocular and cutaneous manifestations (Fig. C, D). Approximately 4 months later, the patient developed worsening cough and skin granulomas, with chest radiograph showing persistent hilar adenopathy, requiring initiation of steroid-sparing immunosuppression using methotrexate and azathioprine to achieve long-term control of the pulmonary involvement. At her last office visit, 2.5 years from symptom onset, her ocular, cutaneous, and pulmonary sarcoidosis were all well-controlled.

3. Discussion

Cutaneous manifestations of sarcoidosis involving tattoos are well documented in the literature.[4] While the precise pathogenesis of sarcoidosis and the mechanistic link with tattoos remains unclear, case reports have identified several associated ink colors and compositions.[5] The soot contained in black tattoo ink may contain carcinogenic, toxic, or mutagenic compounds, and animal models in mice have shown that these particles induce inflammation.[6] In addition to black ink pigment, case reports of tattoo-induced sarcoidosis with uveitis have been described in patients with blue or red tattoo ink.[6] While the majority of cases described in the literature reported ocular inflammation with cutaneous sarcoid within the first year after acquiring the inciting tattoos, the patient in the present case exhibited a delayed onset of sarcoidosis several years after tattoo placement, as reported in other atypical case reports[7,8] Although the etiology of tattoo-related sarcoidosis remains largely unknown, it has been suggested that tattoo pigment may cause a chronic antigenic stimulation that drives a cell-mediated immune response in genetically predisposed patients.[9] This is supported by the 4-year delay in cutaneous manifestation from the time of the tattoo placement. As such, tattoo excision is not typically recommended since the cutaneous lesions are considered manifestations of systemic sarcoidosis. Though the granulomas in the patient's tattoo could have represented a different process from the lacrimal gland inflammation and a biopsy may have provided a more definitive diagnosis, treatment with steroids resolved the granulomas, and the association of tattoo related sarcoidosis was thus made.

This case report describes a unique presentation of tattoo-associated sarcoidosis. The most common presenting ocular symptom with tattoo-associated sarcoidosis is uveitis.[10] While lacrimal gland involvement has been reported in cases of sarcoidosis, lacrimal gland involvement in tattoo-related sarcoidosis has rarely been documented in the literature.[11,12]

4. Conclusions

In summary, physicians should be aware that in addition to uveitis, dacryoadenitis is a possible ophthalmic presentation of tattoo-associated sarcoidosis. Ophthalmologists should consider evaluating tattoo appearance in patients with ocular and orbital inflammatory disease. Further research is required to better understand the association between ocular and cutaneous sarcoidosis and tattoos.

Patient consent

Consent was obtained for publication of this report and image from the patient.

Institutional Review Board Approval.

This study was exempt from Institutional Review Board Approval.

Financial Support

This work was supported by the following grants:

1. Research to Prevent Blindness unrestricted grant to the John F. Hardesty MD Department of Ophthalmology and Visual Sciences.

2. The following authors have no financial conflicts of interest: ADK, RNM, SMC.

Authorship

All authors attest that they meet the current ICMJE criteria for

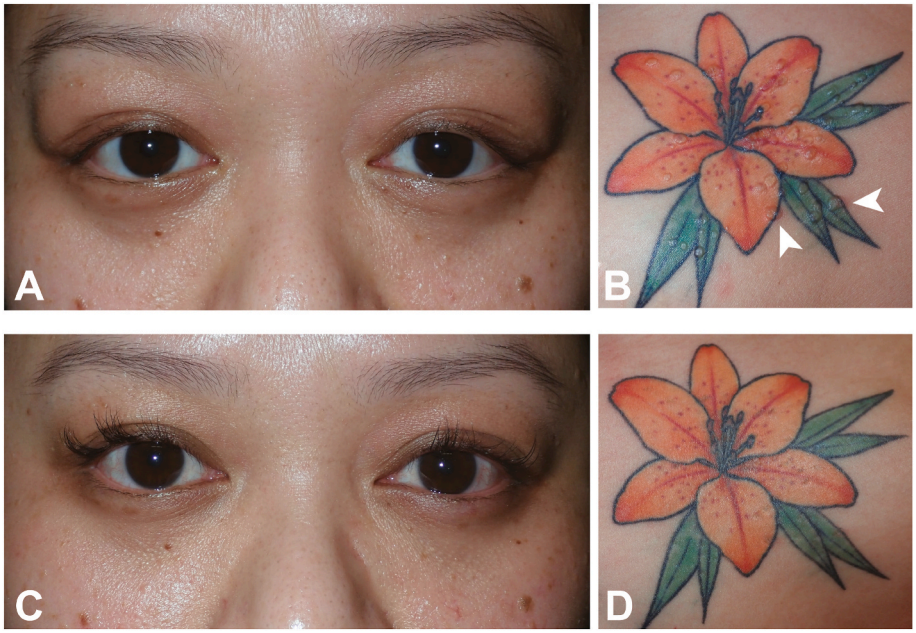

Fig. 1. A. Initial presentation demonstrating bilateral superotemporal eyelid swelling and fullness. **B.** Multiple, circular, elevated lesions resembling granulomas (white arrowheads) overlying the patient's tattoo on initial presentation. **C.** Resolution of lacrimal gland enlargement following systemic treatment. **D.** Improvement in cutaneous granulomas overlying the tattoo following systemic treatment.

Authorship.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Acknowledgements

None.

References

1. Prabhakaran V, Saeed P, Esmaeli B, al e. Orbital and adnexal sarcoidosis. *Arch Ophthalmol*. 2007;125(12):1657–1662.
2. Rothova A, Alberts C, Glasius E, Kijlstra A, Buitenhuis H, Breebaart A. Risk factors for ocular sarcoidosis. *Doc Ophthalmol*. 1989;72(3-4):287–296.
3. Rao DNP, Fernandes R, Wang ZB, Sandhu SJS. A case report of Mikulicz syndrome. *J Radiol Case Rep*. 2017;11(7):1–7.
4. Fernandez FE, McDonnell J. Cutaneous sarcoidosis: differential diagnosis. *Clin Dermatol*. 2007;25(3):276–287.
5. Administration USFaD. Think before you ink: are tattoos safe? FDA. https://www.fda.gov/consumers/consumer-updates/think-you-ink-are-tattoos-safe. Accessed October 13, 2021.
6. Ostheimer T, Burkholder B, Leung T, Butler N, Dunn J, Thorne J. Tattoo-associated uveitis. *Am J Ophthalmol*. 2014;158(3):637–643.
7. Tran D, Ashton R, Cotterill J. Sarcoidosis presenting as tattoo granuloma inadvertently treated with laser therapy. *J Cutan Laser Ther*. 2000;2(1):41–43.
8. Murdoch S, Fenton D. Sarcoidosis presenting as nodules in both tattoos and scars. *Clin Exp Dermatol*. 1997;22(5):254–255.
9. Lo Schiavo A, Ruocco E, Gambardella A, O'Leary RE, Gee S. Granulomatous dysimmune reactions (sarcoidosis, granuloma annulare, and others) on differently injured skin areas. *Clin Dermatol*. 2014;32(5):646–653.
10. Robert A, Rajni M, Jr FAG. Sarcoidal tattoo granuloma. *Dermatol Online J*. 2010;16(11).
11. Agarwal A, Chandak S. Sarcoidosis presenting as lacrimal gland enlargement: eyes speak the truth. *Taiwan J ophthalmol*. 2020;10(3):227–230.
12. Bingöl Kızıltunç PÇF, Hoşal B, Kaygusuz G. Bilaterally diffuse lacrimal gland involvement: initial presentation of systemic sarcoidosis. *Turkish J Orthod*. 2017;47(3):165–168.