Sarcoidosis Following Rhinoplasty: Report of Four Cases

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Abstract- Sarcoidosis is a systemic idiopathic disease, characterized by non-caseating granulomas, primarily affecting the lungs and the lymphatics. Skin involvement is common and lesions may appear in scar tissues. In the present report, four cases of scar sarcoidosis following rhinoplasty are presented which were diagnosed based on serology tests, radiography imaging and histopathology findings. Three women and one man between 32 to 54-year-old are discussed in this report. Skin lesions on their nose were found after an average of 3.8 years of their septorhinoplasty surgeries. Two of the patients had systemic signs such as fever, cough and arthritis. Patients with diagnosed sarcoidosis were treated appropriately. Rhinoplasty is a common procedure today, and the fact that rhinoplasty may lead up to scar sarcoidosis in susceptible people can be an item for surgeons for consideration.

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Introduction

Several granulomatous diseases have a predilection to involve tissue in the airways. They include WG, Churg-Strauss syndrome, and sarcoidosis (1).

Sarcoidosis is a chronic, systemic granulomatous disease described by the presence of noncaseating granulomas in organs and tissues, such as the skin, lymph nodes, lung, eyes, brain, joints, kidneys, and heart. Cutaneous lesions may present with a variety of morphologies, including papules, nodules, plaques, and infiltrated scars (2).

The etiology of sarcoidosis remains unknown, although infectious agents and environmental exposures and also, genetic factors have been proposed (3,4).

Skin involvement is common and often overlooked or misinterpreted, given the lesions' variability (5).

Scar sarcoidosis denotes to lesions of cutaneous sarcoidosis that appear in preexisting scars. This condition may be caused by mechanical trauma such as skin cuts or venipuncture, and scars caused by infection such as herpes zoster (6). Scar sarcoidosis is a rare but specific manifestation of cutaneous sarcoidosis occurring in 2.9-29% of overall cases (7). In these cases, further investigation on systemic involvement must be performed (8). Patients with sarcoidosis may demonstrate the Koebner phenomenon, resulting in new lesions at the trauma site from tattoo (9).

The diagnosis of cutaneous sarcoidosis is supported by the recognition of compatible clinical features, the detection of classic histopathological findings, and the exclusion of other granulomatous disorders (6).

There are a plenty of evidences in the English literature about sino-nasal manifestations of sarcoidosis and also about surgical considerations in sarcoidosis cases who are candidate for rhinoplasty; but to our best knowledge, although rare, sarcoidosis may occur in scar tissues in skin or osteotomy sites after a rhinoplasty surgery, and this may be considered as a surgical complication or "consequence" of rhinoplasty. So, here, four cases of scar sarcoidosis following rhinoplasty are presented. In all cases, the diagnosis of sarcoidosis was made following evaluation for some new cutaneous/sub-cutaneous lesions occurring in facial/peri-nasal area a few months to years after rhinoplasty surgery.

Case reports

Case 1

A 51-year-old woman presented with a 2.5×1.5 cm stony mass (without erythema or tenderness) from 3-months ago on the right lateral nasal wall that obscured

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the nasofacial groove (Figure 1). The patient had no pain or any rhinological or other systematic complaints. In exception of rhinoplasty six years ago, the patient had not any history of trauma on the mass site. Cone Beam Computed Tomography (CT) showed a radiolucent mass, and Fine Needle Aspiration revealed non-inclusive inflammation.

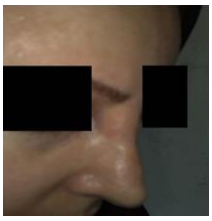

Figure 1. A woman presented with a stony, non-painful mass on the right lateral nasal wall, six years after rhinoplasty

Two pieces of tissue measuring 3×1.2×1 cm were excised completely. No bone erosion was encountered. Histology revealed striated muscle fibers with infiltration of inflammatory cells forming many small granulomas, aggregates of epithelioid histiocytes, plasma cells, lymphocytes and few multinucleated giant cells within the background of fibrotic deposition. Striated muscle with chronic non-caseous granulomatous inflammation was considered compatible with sarcoidosis (Figure 2). Other diagnoses, such as Wegner's granuloma were ruled out according to negative serological tests for instance c-ANCA and p-ANCA and negative histopathologic features of WG include vasculitis of medium and small vessels with intramural, eccentric, necrotizing granulomatous lesions (10).

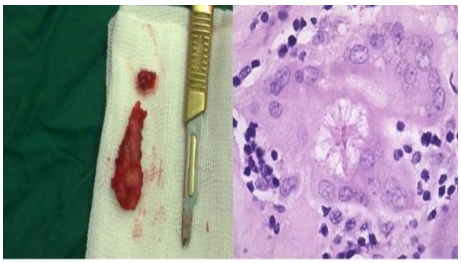

Figure 2. Gross (left) and microscopic (right) appearance (Hematoxyllin–Eosin Staining) of the tissue revealed asteroid bodies in the cytoplasm, stellate or spider like inclusions, aggregates of epithelioid histiocytes, plasma cells, lymphocytes and few multinucleated giant cells within the background of fibrotic deposition

Case 2

A 40-year-old woman, 2 years after septorhinoplasty (open approach) presented with nasal obstruction without epistaxis and a stony mass (1.3×1.8 cm) on the nasal dorsum (upper third) and on the naso-frontal angle. She gradually noticed edema and erythema on her knees, ankles, wrists and elbows, just a few weeks after beginning of lesion. She had no weight loss, cough or hemoptysis but she felt a mild dyspnea with low grade fever and sense of fatigue. ESR and CRP and Rheumatic Factor were positive, but Angiotensin Converting Enzyme (ACE) and c-ANCA and p-ANCA were negative. Chest X ray and CT scan of the thorax revealed multiple lymphadenopathies with diameters up to 11 mm in bilateral hila, right pre-tracheal, right para-tracheal, and sub-carina, and a pulmonary nodule with 4 mm diameter in right lung (Figure 3). Broncoscopic biopsy results diagnosed sarcoidosis, then she received 15 mg of prednisolone orally and almost all of musculoskeletal problems were resolved within three days, except stony lesion on her nose bridge. She received 20 mg intra-lesion triamcinolone acetonide (40 mg/ml) injection. After five days the entire lesion disappeared.

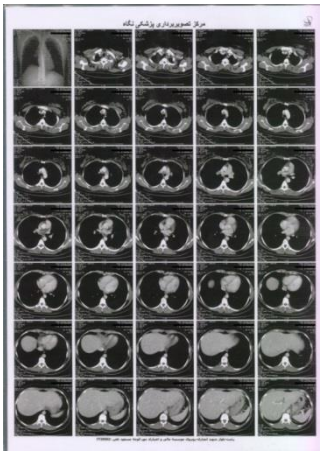

Figure 3. CT scan of the thorax, parenchymal and mediastinal windows in a post-rhinoplasty sarcoidosis woman

Case 3

A 54-year-old woman 3 years after septorhinoplasty presented with several firm masses on the nasal dorsum, supra tip and nasofrontal angle with diameters of 0.5×0.5 cm. There were also several smaller masses on sites of tattoos on eyebrows and lips. She had nonproductive cough, low grade fever and fatigue since several months ago. Chest X-Ray revealed mediastinal widening and hilar lymphadenopathies. ESR, RF, c-ANCA and p-ANCA and ACE were positive. Patient was referred to rheumatologist and the complaints and lesions were disappeared completely after systemic steroid therapy and intra lesional injection of triamcinolone in nasal dorsum and supra tip masses.

Case 4

A 32-year-old man, 4 years after septorhinolasty presented with a skin 1×0.5 cm inflammatory lesion on rightward of nasion region. There were no systemic signs and symptoms. Due to his career, location and dermatologist consultation, differential diagnosis such as sarcoidosis, leishmaniosis and tuberculosis was raised. Serological tests, Chest X-Ray, CT scan and biopsy was performed. Chest X-Ray was normal. ACE was higher than normal, but c- ANCA and p-ANCA was negative. Histopathological exam on skin biopsy a granulomatous inflammation in reticular dermis composed of back to back epithelioid non- necrotizing granulomas associated by mild lymphoplasmacytic infiltrate and separated from the epidermis by a Grenz zone that was suggestive for sarcoidosis (Figure 4).

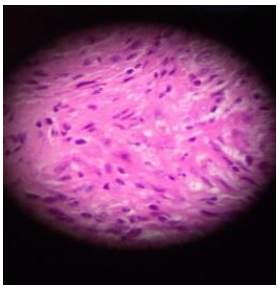

Figure 4. A granulomatous inflammation in reticular dermis composed of back to back epithelioid non- necrotizing granulomas associated by mild lymphoplasmacytic infiltrate and separated from the epidermis by a Grenz zone

Discussion

The skin is complicated in 25% of sarcoidosis cases. Although not life-threatening, the unsightly skin lesions of sarcoidosis can be emotionally devastating (5). Because of its easy accessibility, the skin biopsy is of great value as less-intensive diagnostic procedure (5,9). Only 29% of cutaneous sarcoidosis present as a scar sarcoidosis or cicatricial form in intramuscular injection sites, tattoos, and venipuncture areas, alongside the old cutaneous scars (8,10). Differential diagnosis includes Wegner's granuloma, lipoma, lupus vulgaris, leprosy, rosacea, granuloma and reaction to foreign bodies set aside by patient's history, physical examination and even Para clinic evaluations(4,8).

Nowadays, rhinoplasty is a common procedure, and it may have some minor or major skin or soft tissue complications (11,12). In a study from the Mayo Clinic, 220 patients out of 2319 cases of sarcoidosis (9%) had head and neck involvement and solitary 1% had isolated nose and sinuses involvement (13). In the study of Braun JJ *et al.*, in 2004, 15 cases of sinonasal sarcoidosis were symptomatic with the majority of nasal obstruction (12 cases), rhinorrhea (9 cases), nasal crusts (6 cases), epistaxis (6 cases), facial pain (5 cases), anosmia (2 cases), nodular skin lesions (4 cases), lupus pernio (2 cases), cervical lymphadenopathy (4 cases) (14).

In Aloulah M. *et al.*, study, sinonasal involvement was noted in 30% of patients with sarcoidosis. The mean age of the patients was 52 years, with a female: male ratio of 2.8:1 (15).

In the present study, three women and one man were reported (mean age, 44-year-old). In three cases, the site of lesions was on nasal dorsum and in one case was on nasofacial angle. Furthermore, in one case, there was scar on supratip and nasofrontal angle other than nasal dorsum. None of patients had history of sarcoidosis or other rheumatological diseases or any sinonasal symptoms (rhinorrhea, anosmia, nasal obstruction and epistaxis). Two of our patients had systemic signs and symptoms. Low grade fever, non-productive cough, fatigue, mild dyspnea, edema, and erythema of joints were the systemic symptoms of studied patients. Also, two patients had multiple lymphadenopathies in bilateral hila.

In our study, all four cases of sarcoidosis were diagnosed by histopathological findings, and other disorders, such as Wegner's granuloma were ruled out according to negative serological tests such as c-ANCA and p-ANCA and negative histopathologic features of WG. As we know, negative specific antibodies in the serologic evaluation could not rule out Wegener's granulomatosis, especially in the limited form including sinonasal form of the disease. All patients were followed for at least 18 months (i.e. 1.5 to 5 years) after diagnosis of sarcoidosis, and recurrence was not seen.

Basat *et al.*, have presented ''sarcoidosis nodule formation on the lateral nasal osteotomy lines'' in a sarcoidosis patient underwent rhinoplasty (16). In the study of Marks SC and Goodman in 1998, from 6 studied patients with nasal and sinus sarcoidosis, five patients had active pulmonary disease (17).

All of our patients received intra-lesional corticosteroid injection and the lesions had been disappeared completely during 1.5 years. This finding is in concordance with the results of Braun *et al*., study who reported that corticosteroids remain the cornerstone of therapy (14). Albeit, the literature says that despite an often long and aggressive treatment, relapses and chronicity are frequent after tapering or discontinuing the corticosteroids and require a long follow-up and interdisciplinary management (15). Finally, we must clarify that, although the nasal lesions may have been the

presenting feature of sarcoidosis, it does not mean that nasal surgery 'caused' sarcoidosis or even that the systemic manifestations weren't present (but undetected) long before surgery.

As we know, proofing the causality with case series could not be possible and we have not the assumption that rhinoplastic surgery might have a "causality effect" for sarcoidosis; but we can keep in our mind "scar sarcoidosis" as a consequence or sequela of rhinoplasty, especially in endemic areas for sarcoidosis and also in areas in where rhinoplasty is done frequently. This point is much more important when we consider that according to rhinoplasty and many other surgery references, it is not necessary doing routine rheumatology serological evaluations in usual rhinoplasty candidates. Finally we emphasize importance of accurate and more detailed physical examination and history taking, and even paraclinic evaluations (if necessary) in all the patients and surgery candidates, including rhinoplasty cases.

Our findings showed that the scar of rhinoplasty can be a site for skin involvement of sarcoidosis and "scar sarcoidosis" should be considered in the differential diagnosis in patients presenting with unusual cutaneous or subcutaneous lesions/nodules in rhinoplasty scars. Rhinoplasty is a common procedure today, and the fact that rhinoplasty may flare sarcoidosis in some susceptible people can be considered by aesthetic surgeons.

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