Widespread Paraffinoma

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Paraffinoma (sclerosing lipogranuloma) is a distinct foreign body reaction caused by the introduction of mineral oils. Widespread paraffinoma is rare and, to our knowledge, has never been reported in Taiwan. We herein report a 34-year-old man with widespread self-inflicted paraffinomas involving multiple anatomic sites. The patient had self-injected vast amount of baby oil and other oily substances (more than two liters) into these lesions in an apparent attempt to cure his muscle inflammation. Histopathologically, multiple empty cysts were seen in the sclerotic dermis and subcutis with the characteristic "Swiss-cheese" appearance; oil red O stain demonstrated the presence of lipids within the cysts. We excised three painful nodular lesions and no recurrences were noted after more than three months of follow up. (Dermatol Sinica 20 : 268-274, 2003)

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INTRODUCTION

Paraffinoma is caused by the presence of paraffin (mineral oil) or other lipophilic substances under the skin. It gives a peculiar histologic pattern of "Swiss cheese" — cystic spaces of various sizes replacing the dermis and subcutaneous tissue. Paraffinomas can be factitious or iatrogenic. They have been described in various anatomic sites including male genitals, female breasts, orbital region and peritoneum. However, widespread, multiple-site occurrences are unusual.

CASE REPORT

A 34-year-old man presented with a two-month history of multiple yellowish brown nodules on the chest, shoulders, buttocks, and bilateral lower extremities. The lesions were firm, and some became painful and tender a few months prior to the medical visit. On examination, there were yellowish-tan nodules on the chest and bilateral upper arms (Fig. 1A). The lesions on both legs were yellowish brown confluent plaques covering thighs, buttocks, calves, ankles, and popliteal fossae (Fig. 1B). Enlargement of left inguinal lymph node was also noted. The physical examination elsewhere and systemic review were basically normal. The clinical differential diagnoses included foreign-body granuloma and deep fungal infection.

Laboratory workup revealed mildly elevated uric acid (10.6 mg/dL; normal range 2.6 ~ 7.8). The serology workup including HIV and VDRL was negative. The chest X-ray was normal.

A skin biopsy performed on the right thigh revealed multiple various-sized cystic spaces in the dermis and subcutaneous tissue (Fig. 2). Foamy cells were noted around some of these empty cysts. Lymphoid aggregates and multinucleated giant cells were present. PAS and acid-fast stains were negative. Tissue sent for bacterial, fungal and mycobacterial cultures also failed to grow any microorganism. Polarized microscopy showed no bifrangent material. Oil red O-stained sections prepared from frozen tissue showed that the cystic or empty spaces contained lipid material (Fig. 3). The pathologic picture was characteristic of a paraffinoma or...
so-called sclerosing lipogranuloma.

Upon query for occurrences of oil or liquid injections, the patient reluctantly admitted a revealing history. While in the military, he suffered from muscle aches on the thighs, buttocks, chest, and arms, which he believed was muscle inflammation. He applied several topical agents including antifungal and corticosteroid ointments, but they were ineffective. Two months prior to the visit, he started to inject baby oil (composed of mineral oil and fragrance) into these areas in an attempt to eradicate this inflammation and also to improve his dry skin condition. Within a period of one week, he injected over two liters of baby oil into multiple sites over the body where he thought was inflamed or dry. A few months after the injection, he began to worry about the persistent yellowish indurated plaques on the injection sites. He tried to massage them away while continuing to inject various oil substances, only to find the lesions more extensive.

Coupling with earlier clinicopathological findings, a final diagnosis of paraffinoma was made. Three symptomatic nodules on the shoulders and legs were subsequently excised over a period of two years. Histopathology of the excisional specimens showed a more fibrotic stroma with thick, eosinophilic collagen bundles compared to the earlier biopsy. Some of the cysts were encircled by concentric layers of collagen bands (Fig. 4). The lymphoid aggregates and multinucleated giant cells were less prominent. Calcification was occasionally noted.

No recurrences of lesions were noted on the excised sites after follow up for more than three months. Upon reviewing his mental status, the patient was oriented with intact verbal communication throughout the clinical course. However, he admitted he was in a depressed state. There were no auditory or visual hallucinations. Unfortunately, the patient denied the suggestion of further psychiatric consultation.
DISCUSSION

Paraffin, known as mineral oil in its liquid form, has been used medically as vehicles for intra-muscular injections of lipid-soluble drugs since the early part of 20th century.7 Paraffins were also injected into various tissues for cosmetic purposes in order to increase the volume of a tissue or to eliminate wrinkles. The first record of its cosmetic use dates back to 1899.3, 4 Subsequently, these agents were used for a wide range of purposes including cleft palate, wrinkles, other facial deformities, baldness, as well as muscle, breast and penile augmentation.4 The adverse effects from the use of these oils were reported as early as 1906, when Heidingsfeld described disfiguring subcutaneous nodules in two patients following paraffin injections for facial wrinkles.4 The term paraffinoma first appeared in medical literature in 1920.12 Despite early warnings, the practice of paraffin injection for cosmetic purpose continued throughout the century. The therapeutic use had generally ceased by 1930.

Sclerosing lipogranuloma is a term first introduced by Smetana and Bernhard in the mid-20th century to describe a peculiar lesion in the male genitalia.12 Its histopathological descriptions were analogous to those of paraffinoma.1 Because the majority of patients denied a history of foreign body injections, Smetana and Bernhard considered it a distinct subcutaneous inflammatory response to endogenous lipid degeneration caused by various types of tissue injury.13 Due to a lack of sophisticated equipment detecting the nature of lipids and inconsistent patient accounts on their injection histories,14, 15 conflicting opinions regarding the pathogenesis of sclerosing lipogranuloma were proposed over the ensuing years.

In 1977, Oertel and Johnson studied 21 cases and indicated the presence of paraffin hydrocarbons in the lipid when analyzed by infrared spectrophotometry.16 Included among their cases were nine that previously had been reported by Smetana and Bernhard. Subsequently, newer modality such as field desorption mass spectrometric analysis had been developed and proved the exogenous origin of paraffin in paraffinoma, even though patients have denied any history of foreign body injections.17

The term paraffinoma or sclerosing lipogranuloma now refers to cases caused by exogenous oily substances specifically containing straight-chain saturated hydrocarbons.7 They form because of a lack of enzymes in humans to degrade these substances, inciting a foreign body reaction.2, 7 Therefore, modern dermatology textbooks tend to avoid the use of the term sclerosing lipogranuloma.18 Nevertheless, there are still few recent reports of sclerosing lipogranuloma arguing that endogenous lipid as the cause. They are dubbed "primary" sclerosing lipogranuloma. In these cases, no history of injection or topical application of oils is reported. Other features of primary sclerosing lipogranuloma include prominent epithelioid cells and eosinophils in the lesion, peripheral eosinophilia, and spontaneous regression following partial resection.19, 20 Because of a different etiology, some think these cases are better termed allergic granuloma.21

The histopathology of paraffinoma often demonstrates a "Swiss cheese" appearance, a picture of multiple round, empty spaces (cysts) in a fibrotic stroma.2, 18 Another occasionally seen feature is the "onion-skin" appearance, which describes larger cysts lined by concentric fibrotic rings.2 Our case exhibited both of these features. Oil-red-O stain or Sudan IV stain demonstrated the presence of oil in these cysts. As stated above, humans lack the enzyme to metabolize interstitial exogenous oil. The body tissues thereby respond with a foreign body reaction. The histological findings may vary with time and types of exogenous oil. Our case exhibited multiple biopsies and echoed the earlier report on histological evolutions, i.e. multinucleated foreign-body giant cells seen early in the inflammatory reaction while older lesions dominated by interstitial fibrosis and multiple Swiss cheese-like cystic spaces.7 The degree of fibrosis varies according to the nature of the inciting oil.7
Paraffinoma is mostly localized to one or limited anatomic sites, often occurring on male genitalia, female breasts, and faces sites patients most commonly desire to alter their contour. The most documented site is the male genitalia. Various types of oil, usually liquid paraffin, have been injected into the penis in the hopes of increasing the girth and potency. Not surprisingly, most patients deny having such injections. Paraffinoma of the breast results from paraffin injection in breast augmentation. It manifests from a painless mass to a destructive ulcer simulating breast cancer. Less common sites include orbit, scalp and peritoneum. Paraffinoma of eyelids and anterior orbit has been reported after endonasal sinus surgery, related to ointment-covered nasal packing used in the operation. Scalp paraffinoma had been reported due to grease gun injury or oily injections believed to have effect on hair regrowth. Paraffinoma of the peritoneum was due to early surgical practice that poured paraffin into the peritoneum to prevent peritoneal adhesions.

Widespread paraffinoma has rarely been reported. Only two cases were found in the literature, with completely different circumstances. One involved a dancer who sought to augment the size of her calves by injecting more than one liter of mineral oil into her legs. Physical examination showed diffuse involvement of the thighs and calves with femoral and inguinal lymphadenopathies. Initial chest X-ray revealed multiple fine, scattered densities, which later were found to be fat stain (+) foreign body granulomas on lung biopsy. Lymph node biopsy revealed a similar picture. Paradoxically, chest X-ray taken three months later showed total clearing of the fine nodular densities. No severe complications were noted except for leg pain and swelling in two years of follow up. The other case was a man with scrotal lipogranulomas from repeated self-injection of mineral oil for years. Sudden onset of pulmonary edema occurred and resulted in his death. Autopsy revealed disseminated lipogranulomas in the lungs. Vacuoles were also found in the kidneys and spleen. The patient was reported to have a cough for four years. The cause of acute pulmonary edema was attributed to formation of new emboli from inadvertent intravascular injection of a large dose of mineral oil.

Our case involved the chest, arms, buttocks, and legs. The substantial injection amount (more than two liters) and involvement of widespread anatomic sites were unusual. Clinically, there were two main types of lesions, namely, nodules (chest and arms) and plaques (predominantly the lower extremities). This different clinical morphology may be due to the different amounts of injected oils in various sites, since all the lesions were on injection sites, i.e. the patient had injected more oily substance into the thighs and legs than the chest and arms. Presumably, if injected oil is confined to a localized area, it produces a nodular lesion, while diffuse plaques could be seen if oil is distributed in a wide range of body surface. While the patient reported no shortness of breath, hemoptysis or cough and chest X-ray was normal—indicating the lung was free from involvement thus far; pulmonary granuloma may still be subclinical. Cessation of further injection is of primordial importance to prevent the potentially fatal pulmonary emboli. Another interesting note is the patient's unusual intent of curing his "muscle inflammation". This bizarre behavior raised the question of a possible altered mental state, although association of psychotic or personality disorder with paraffinoma has not been previously reported.

The lesions of paraffinomas usually appear weeks to years after administration, but some may appear as late as 42 years. The inciting factors that initiate the symptomatic inflammatory event are not known. Patient idiosyncrasies, total dose, structures and impurities of the exogenous lipid, and local trauma may be determining factors. Once initiated, paraffinomas tend to persist indefinitely. Spontaneous resolution has not been reported. The induration present clinically may be due to extensive fibrosis and obstruction of lymphatic drainage by
lymph nodes engorged with these nonabsorbable agents. In chronic cases, the lesions remain essentially unchanged, although the coexistence of sarcoma has been reported. Surgical excision remains the only effective therapy.

Although paraffinomas are not common, clinicians need to be aware of this entity as most patients are reluctant to admit to injecting foreign substances into their bodies. The clinical diagnosis may not be immediately apparent if a history of oil injection is not established. Furthermore, there can be a very long lag time before the development of clinical symptoms following such injections. Considering the unquenchable human desire to improve body appearances, paraffinomas will unfortunately continue to be seen in the new millennium. Increased public awareness must be established to prevent this physically and psychologically debilitating condition.

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