

Kaposi-like manifestations in a newly diagnosed AIDS transgendered patient with silicone embolism syndrome and disseminated tuberculosis

Chiara Picarelli¹, Alberto Borghetti¹, Simona Di Giambenedetto^{1,2}

¹Fondazione Policlinico Universitario A. Gemelli IRCCS, Institute of Clinical Infectious Diseases, Rome, Italy;

²Università Cattolica del Sacro Cuore, Institute of Clinical Infectious Diseases, Rome, Italy

SUMMARY

The use of liquid silicone for cosmetic procedures can yield serious sequelae including embolization and pneumonia. We describe a recent case of silicone embolism syndrome occurring together with systemic tuberculosis in a transgendered patient newly diagnosed with AIDS. She presented with fever, hematochezia, lymphadenopathies, purple nodular lesions and lower limb edema. HIV test was positive. A chest X-Ray showed interstitial infiltrates and a tomography showed necrotic lymph nodes and pulmonary nodules with blurred borders, suggesting Kaposi sarcoma. Psychomotor impairment then occurred in the absence of tomographic signs of acute neurological events. The *Mycobacterium tuberculosis* genome was isolated from stool and bronchial washing samples. Histological examination of a necrotic lymph node showed lymphadenopathy due to silicone accumulation. Moreover, the patient presented fever and swelling of lower limbs; a tomography showed multiple foreign body granulo-

mas. After starting antitubercular, antiretroviral and antibiotic treatment she reported symptomatic improvement including a mild recovery of motor-slowness. There are few reports about silicone-induced pulmonary disease in HIV-1 infected patients and, as far as we know, none of them describes an overlapping pulmonary involvement due to *Mycobacterium tuberculosis* infection. Even if extensive clinical and radiologic evidence is suggestive of Kaposi sarcoma (fever, severe immunodeficiency, multiple cutaneous nodules, hematochezia, diffuse lymphadenopathies), it is possible to see Kaposi-like manifestations in patients with systemic silicone embolization. With this article we wish to stress the attention on the possible overlap of more than one concurrent disease in an immunocompromised host.

Keywords: silicone embolism syndrome, disseminated tuberculosis, HIV, AIDS, Kaposi sarcoma

■ CASE PRESENTATION

A 36-year-old Brazilian transgendered male-to-female patient presented with asthenia, weight loss of about five kg in the last three months, fever up to 38 degrees, tenesmus and hematochezia. She had undergone several injections of a silicone fluid mixture into each hip and gluteus over a period of several years up to two

years before. Mild swelling of the injection site would occur right after the procedure, as well as progressive lower limbs oedema due presumably to the migration of the silicone itself. For this reason the patient had self-administered intra-muscular injections of betamethasone for the previous two years, with reported benefit on the pain and swelling of the lower limbs. She was not taking any medication and affirmed not to be on drugs. Initial laboratory findings showed: increased C Reactive Protein (187 mg/L) and procalcitonin (1.41 ng/mL), LDH (326 UI/L), hemoglobin, 11.5 g/dL; total lymphocyte count, 260 cells/mm³. Peripheral blood lymphocyte typing showed CD4

Corresponding author

Chiara Picarelli

E-mail: chiara.picarelli1989@gmail.com

6/mm³, 10%; CD8 410/mm³, 64%; CD4/CD8 ratio 0.2. A chest X-Ray showed interstitial infiltrates (Figure 1A). The research of anti-human immunodeficiency virus (HIV) antibodies was positive. Urinary toxicology screen was negative. Physical examination showed low blood pressure (90/50 mmHg), tachycardia (140/min); fever (38°C), hepatomegaly and diffuse lymphadenopathy. Rectal examination revealed bloody stools in the rectal ampulla, but no palpable vegetative lesions. Skin examination showed multiple purple nodular lesions localized to the lower limbs and to the chest. Severe lower extremity non-pitting edema was shown too. Urinary *Legionella pneumophila* and *Streptococcus pneumoniae* antigens were negative. Antibiotic therapy with vancomycin 1g q12h and piperacillin/tazobactam 4.5 g q8h was

assessed for a 7 days cycle, owing to the possible bacterial pulmonary co-infection. Meanwhile, the patient developed psychomotor impairment in absence of focal neurologic deficits. A total-body Computed Tomography (CT) with contrast showed several thoracic and abdominal necrotic lymph nodes; in both lungs, predominantly in the upper lobes and in the para-hilar area, multiple little nodules with blurred borders and a faint halo of enhanced peripheral parenchymal density, and thickening of inter-lobular septi and of the big fissure were described: these evidences were reported as suggestive for Kaposi sarcoma (KS) (Figure 1B). Compressive atelectasy of the right lung parenchyma concomitant to an extended pleural effusion and pericardial effusion were present too. Diffuse peritoneal thickening with

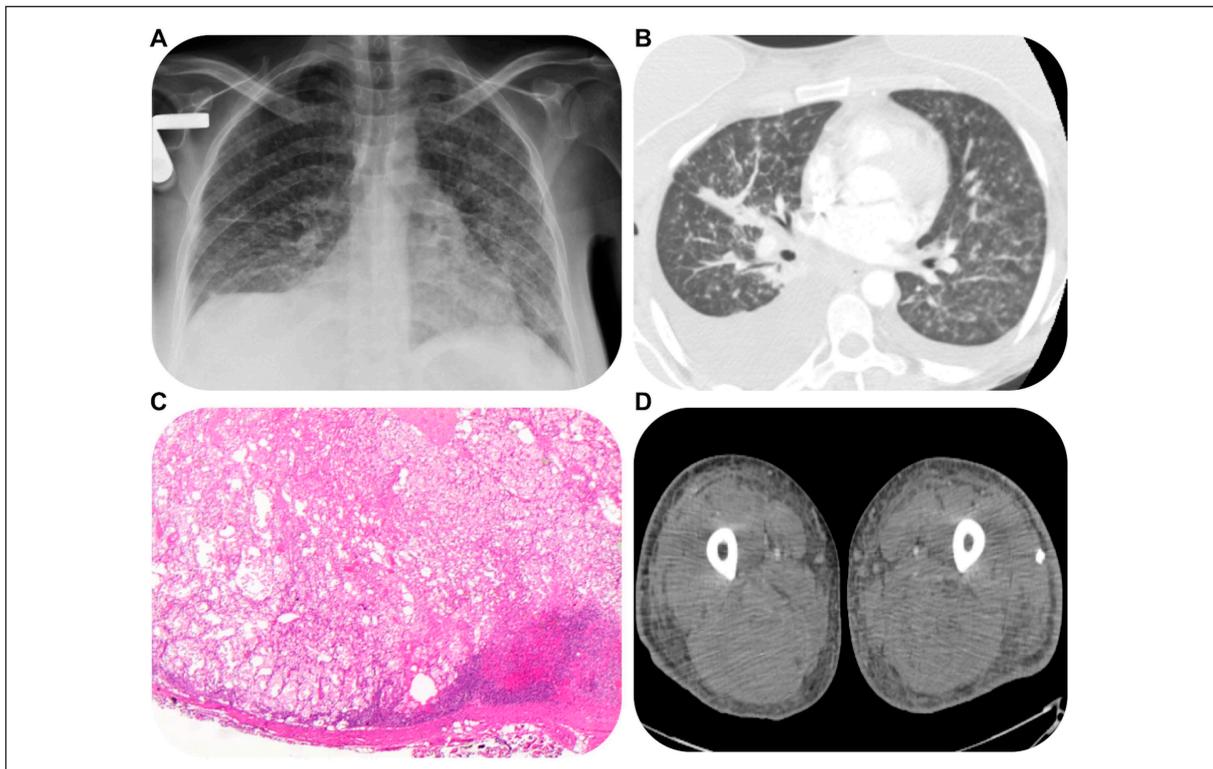


Figure 1 - Radiological findings and histologic exam of a necrotic lymph node

A: Chest X-Ray at presentation, showing bilateral diffuse interstitial infiltrates; **B:** Computed tomographic scan demonstrating multiple little nodules with peri-lymphatic and peri-broncho-vascular distribution were observed; the biggest of them showed blurred borders with the evidence of a faint halo of enhanced peripheral parenchymal density, as it is usually observed in the diseases characterized by peri-nodular micro-haemorrhage and several thoracic necrotic lymph nodes. **C:** Histologic exam performed after the biopsy of an axillar necrotic lymph node showed lymphadenopathy due to silicone accumulation. **D:** Computed tomographic scan demonstrating multiple and partly calcific foreign body granulomas with associated inflammation of the near tissues.

multiple nodules, and other nodules with blurred borders in the subcutaneous tissue of the thighs bilaterally were also suggestive for KS. Brain CT was normal. An echocardiogram was performed and showed pericarditis. In the suspicion of disseminated tuberculosis, the patient was isolated. Sputum samples were negative for microorganisms and for bacterioscopic research of *Mycobacterium tuberculosis* as well as serum assays for *Chlamydia pneumophila*, *Legionella pneumoniae*, *Mycoplasma pneumoniae*, *Cytomegalovirus*, *Cryptococcal* antigen and Beta-D Glucan. Polymerase Chain Reaction (PCR) for *Mycobacterium tuberculosis* genome from a stool and a bronchial washing sample was positive. Antitubercular therapy with isoniazide, rifampin, pirazinamide and etambutol was started, with prompt benefit on the fever; two weeks after, combination antiretroviral therapy (cART) with tenofovir/emtricitabine and dolutegravir (at the dose of 50 mg BID) and prophylaxis with azitromycin and trimetoprim/sulfamethoxazole were also started. Histologic examination of one of the nodular skin lesions biopsy was positive for foreign body granulomas; histologic exam of an axillar necrotic lymph node showed lymphadenopathy due to silicone accumulation (Figure 1C). A proctologic examination showed internal hemorrhoids. Afterwards, the patient presented fever, swelling and erythema of the lower limbs. A CT scan with contrast of the legs showed multiple and partly calcific foreign body granulomas with associated inflammation of the near tissues (Figure 1D). Antibiotic therapy with teicoplanin with loading dose of 400mg q12h for three doses and maintenance dose of 400mg q24h was assessed for a 14-days cycle. Within days, she reported symptomatic and psychomotor impairment improvement and was discharged with the indication to be followed-up at the infectious diseases outpatient service of our clinic.

■ DISCUSSION AND CONCLUSIONS

Liquid silicone is utilized for cosmetic procedures by physicians as well as illegally by unlicensed personnel; it plumps tissue volume and, moreover, it's durable and presumably weakly antigenic and can be used for therapeutic purposes [1, 2]. A recent review of the literature, though, focuses on the potential antigenicity of some polymers [3, 4]. However, silicone injection is not risk free: some

potential complications include local infection, tissue necrosis and migration from the former site of injection, causing foreign body giant cell reactions, regional lymphadenopathy and acute systemic illness [1, 2, 5-8]. The embolization of silicone to the pulmonary vasculature can cause a presentation similar to the one of Fat Embolism Syndrome. Many reported cases of silicone embolic-related acute pneumonitis with dyspnea, fever, cough, chest pain, hypoxia, hemoptysis, diffuse alveolar damage, acute granulomatous pneumonitis and lymphadenitis with subsequent deaths are described in literature [1, 7-10]. The onset of respiratory disease is often seen within days of the procedure, though a case of chronic granulomatous pneumonitis with septal thickening and micro nodules was also described [1]. Another common manifestation of silicone embolism is rash. Neurologic dysfunction is present in up to one-third of all patients following silicone embolism syndrome, and is associated with rapid clinical deterioration and increased mortality [9]. Management of this kind of embolic syndrome is supportive. Silicone pulmonary embolism is proposed to result from increased tissue pressures at injection sites, local massage of the site or the inadvertent direct intravenous injection of silicone particles [10, 11]. There are few reports about silicone induced pulmonary disease in HIV-1 infected patients and none of them describes an overlapping pulmonary involvement due to *Mycobacterium tuberculosis* infection [1]. Moreover, in our experience, many confounding factors were present: the patient was severely immunocompromised and presented multiple purple nodular lesions localized to the lower limbs and to the chest, hematochezia and tenesmus and a CT scan was suggestive for nodal, pulmonary and peritoneal KS. Necrotic nodes described in CT as part of the systemic involvement of KS were likely caused by foreign body accumulation together with tubercular caseous necrosis. Hypoxia and a compromised pulmonary imaging were also a consequence of systemic silicone embolism and tubercular disease. Data about the burden of HIV infection in transgender in Italy (roughly 400000 people) are not available yet; though, a national campaign of surveillance promoted by Superior Institute of Health and aimed to the improvement of the standard of care in this population has recently been approved. With this article we want to

stress the attention on the possible overlapping of more concurrent diseases in an immunocompromised host ... when it's AIDS it's better not to rely always on Occam's razor principle!

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Conflict of interest

There are no conflicts of interest.

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