

# Still's disease in patient with silicone breast implants: case report

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**Abstract:** Still's disease is a systemic manifestation of juvenile arthritis that rarely occurs in adults and therefore is often forgotten in differential diagnosis of fever of unknown origin. The case presented here shows diagnostic difficulties in a patient with silicone breast implants and fever with Still disease. The cause-effect relationship between breast implants and Still disease is not certain, however, this possibility should be considered when a patient with fever resistant to antibiotics and with silicone implants is admitted to the hospital.

**Key words:** breast implants, silicone, Still disease

## CASE REPORT

A 41-year-old woman was admitted to the hospital because of prolonged fever (up to 40°C with 2 peaks in the morning and in the evening). During the 2-months' course of the disease prior to hospitalization, pneumonia, endocarditis, sinusitis, urinary tract infection as well as inflammatory foci in the oral cavity, pharynx and reproductive organs have been excluded. Fever did not respond to several antibiotics, but was resolving temporarily after administration of antipyretic drugs (metamizole).

The patient had had the silicone breast implants, inserted in 1993 and removed two years before admission due to inflammatory reaction around the implants with consecutive spill of silicone in the subcutaneous tissue. Three months before admission both breasts were reconstructed with liquid expanders. Considering the presence of a foreign body (breast implants) as a cause of fever, sonography of the breasts has been performed a few times before admission, however no pathology could be seen around the implants.

On admission she was febrile but alert and cooperative. Physical examination revealed salmon-pink rash on the patient's shoulders (which was also described during the prior hospitalizations and was considered an allergic reaction to the

administered antibiotics) and slightly enlarged cervical and axillary lymph nodes.

Laboratory results disclosed elevated inflammatory parameters (white blood cell count 16.9 G/l, C-reactive protein 43.7 mg/l, fibrinogen 528.7 mg/dl, sedimentation rate 70 mm), lactate dehydrogenase 672 U/l, aspartate aminotransferase 129 U/l, alanine aminotransferase 71 U/l,  $\gamma$ -glutamyl transpeptidase 198 U/l with normal bilirubin level. Hepatitis B and C markers were negative. Anti-cytomegalovirus and anti-toxoplasma IgM antibodies were negative. A total IgE level was elevated (268.1 IU/l). Mild hypothyreosis with the thyreotropic stimulating hormone level (5.87 uIU/ml) and elevated anti-thymic globulin and anti-thyroid-peroxidase antibodies was observed.

The standard procedures to establish the diagnosis of fever were performed (chest X-ray and computed tomography [CT], abdomen sonography and CT, thyroid hormones status and thyroid gland biopsy, sonography and histopathology of enlarged lymph nodes, otorhinolaryngological and gynecological examination, titers of antistreptolysin O, anti-nuclear antibodies and anti-neutrophil cytoplasmic antibodies-screening). All those examinations revealed only a vague hepatosplenomegaly and enlarged cervical and axillary lymph nodes with normal structure. All the cultures (sputum, urine and blood) taken on admission were negative. However, in the blood sample taken later during the hospitalization, the presence of methicillin-susceptible coagulase-negative *Staphylococcus* was stated. The bacterium was sensitive to gentamycin and trimetoprim, which were initiated immediately. Nevertheless, no relevant fall of the body temperature was observed. As an inflammatory reaction to the breast implants and sepsis originating from this place was suspected, breast implants removal was proposed to the patient but she refused.

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**Table. Diagnostic criteria for adult Still disease. To establish the diagnosis, at least 5 criteria, including at least 2 major ones, are required [1]**

**Major criteria:**

- 1) fever of 39°C or higher, lasting 1 week or longer
- 2) arthralgia lasting at least 2 weeks
- 3) macular or maculopapular nonpruritic salmon-pink eruption, appearing during fever
- 4) leukocytosis >10,000 G/l, neutrophil count >80%

**Minor criteria:**

- 1) sore throat
- 2) lymphadenopathy and/or splenomegaly
- 3) liver dysfunction: increased activity of liver enzymes (SGOT, SGPT, LDH – after exclusion of other potential reasons)
- 4) absence of rheumatoid factor in IgM class and antinuclear antibodies (measured by immunofluorescence method)

**Exclusion criteria:**

- 1) infections (especially sepsis and mononucleosis)
- 2) neoplasms (especially malignant lymphoma)
- 3) other rheumatoid diseases (periarteritis nodosa, rheumatoid vasculitis with extraarticular features)

LDH – lactate dehydrogenase, SGOT – serum glutamic oxaloacetic transaminase, SGPT – serum glutamic pyruvic transaminase

Despite the treatment the patient was still febrile and her state was worsening. She was weak, hypotonic, developed electrolyte abnormalities. Suspecting the possibility of relative hypocortisolism (hypotonia, tendency to low sodium levels) in the patient with staphylococcal sepsis, glucocorticoid (hydrocortisone 4 × 50 mg *i.v.*) administration was started on the 14th day of hospitalization resulting in the immediate fall of the temperature, inflammatory parameters and liver enzymes as well as the rise of blood pressure. An attempt to reduce the dose of glucocorticoid resulted in the immediate rise of fever, worsening the patient condition and the appearance of rash. A higher dose of steroids resulted in the regression of all the symptoms. Such a reaction, regarding the signs that earlier were thought to be associated with sepsis, lead to the diagnosis of Still's disease. The patient was discharged from hospital with no fever on 60 mg of prednisone daily and no antibiotic treatment. Two coexistent diagnoses were regarded, i.e. staphylococcal sepsis and Still's disease induced by breasts implants.

As a relationship between silicone implants was considered plausible, and only on as high as 40 mg dose of prednisone the patient was free of symptoms, she was again advised and finally agreed to remove the implants. In the histopathological examination no signs of inflammation or abscesses were found. Cultures of expanders liquid and blood were negative. However, any attempt of reducing the high dose of prednisone after the operation resulted in high fever. Only when methotrexate and chloroquine treatment was started, the reduction of the steroid dose became possible. At present the patient is treated

with prednisone 10 mg daily, chloroquine 500 mg daily and methotrexate 7.5 mg weekly. No side effects of the prolonged steroids therapy except the weight gain have been observed in two years after initiation of the treatment.

## DISCUSSION

The cause-effect relationship between silicone and Still's disease is not certain. In the large meta-analysis of Janowsky [2], no association was confirmed between breasts implants and connective tissue disease. However, neither the meta-analysis, nor even the largest studies included did not list Still's disease. The concept of a relation between breast implants and Still's disease is based on case reports, which, however, do not prove a causal relationship [2-5]. For example, Montalto et al. [5] described a regression of the disease under corticosteroid treatment, which persisted after steroid discontinuation in spite of the fact that implants were not removed. In the presented case the disease developed 2 years after removal of broken silicone expanders, but in a short time after replacing them by liquid expanders. Such a course may suggest induction of an immunological intolerance which remained subclinical until a new exposure exacerbated the course of the disease, but another possible explanation is induction of the Still disease by concomitant staphylococcal infection (which made the diagnosis more difficult and caused a delay of it).

Diagnostic difficulties were also caused by the fact that classic diagnostic criteria for adult Still disease include sepsis as an exclusion criterion [1]. Nevertheless, taking into account that diagnostic criteria were fulfilled (3 major and 2 minor criteria – see Tab.) and that all signs and symptoms resolved after glucocorticoid treatment, the diagnosis seems to be adequate. One should also take into account that signs and symptoms of Still's disease and sepsis are in part similar. One cannot exclude that the first positive blood culture could be caused by inappropriate sampling and that signs and symptoms qualified as septic were in fact signs and symptoms of Still's disease.

In conclusion, in the presented case the causal relationship between Still's disease and silicone breasts prostheses is also uncertain. However, it cannot be excluded. Therefore, with purpose of reminding Still's disease as a potential cause of fever resistant to antibiotics, difficult to prove, we decided to describe this case.

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