

Review Series

# RHEUMATOLOGY

ARTICLE

**Silicone breast implants and  
connective tissue disorders:  
no evidence of cause  
and effect**

Hugh Zachariae, M.D., Ph.D.,  
Højberg, Denmark.

CASE REPORTS

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*The review series features published case reports from around the world, discussed in depth by the original authors. We hope that you will find these collections of case histories informative and useful in your daily practice.*

— Chief editor

## ARTICLE

# Silicone breast implants and connective tissue disorders: no evidence of cause and effect

Hugh Zachariae, M.D., Ph.D.,  
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## REFERENCES

# A 15-year exercise program for rheumatoid vasculitis

W.P. Marley, T.F. Santilli.  
Scand J Rheumatol 1998; 27: 149–51.

## AUTHOR'S INTRODUCTION

We report a rare case of rheumatoid vasculitis, which occurs in less than 1% of patients with rheumatoid arthritis, and the response to a 15-year supervised exercise program of a patient with significant impairment in mobility and physical work capacity. This disorder is characterized by chronic visceral and peripheral pain that encourages disability and historically carries a poor prognosis. Treatment requires astronomical amounts of medical care. The hallmark of our case, however, is this patient's determination to overcome disability. It is a tribute to the human spirit. We are not aware of a similar study of such length.

## Case report

This 55-year-old white male sales manager was retired prematurely on disability and referred by his rheumatologist for a supervised program of therapeutic exercise at the age of 40. He was determined to overcome multiple disabling conditions related to his disorder, which was characterized by chronic visceral and peripheral pain that encouraged disability (1–4). He had a 30-year history of rheumatoid arthritis and secondary vasculitis with inflammatory neuropathy and mononeuritis multiplex preceded by severe rheumatic fever when he was 3 years old. There was no family history of cardiovascular disease, systemic lupus erythematosus, Hodgkin's disease, or rheumatic fever. He had a 12 pack-year history of smoking, but had not smoked for 30 years.

The patient presented with significant muscle wasting, hypesthesia, and diminished proprioception in the extremities. No reflexes were elicited

in the lower extremities. Subluxation deformities were noted in both hands, but were greater on the left than on the right. There was flexion and extension limitation of the metacarpophalanges and proximal interphalanges with flexion limitation in both knees at 90°. There was decreased position, pinprick, and vibratory sense as well as severe loss of motor and sensory function in the hands and feet such that the patient required braces. He had no feeling distally from the elbows and knees. The ability to perform activities of daily living was impaired. His car had been specially adapted for entrance/exit and operation. He could not use public transport. Buttons had been replaced by Velcro. The left hand had no prehension and could not be used for daily tasks – items on the floor, shelf, or table were picked up with the right hand. He could manage stairs if there was a railing.

His medical history included a subtotal gastrectomy, bilateral vagoto-

my for peptic ulcer, and bilateral synovectomy for the knees. He had undergone surgery for repeated renal calculi. Cataract and retinal surgery was required for the right eye in 1991. Other clinical events included osteomyelitis in the left foot and surgical reconstruction of the left hand. Compromised arterial circulation developed into gangrene in four fingers and four toes and required surgical debridement. No dorsalis pedis pulse could be felt on the left, and chronic venous incompetence required the use of compression stockings in the past. Acute cholecystitis required a cholecystectomy in 1981. Hospitalization was required in 1971 for pulmonary emboli which were resolved by anticoagulant therapy. Some of the clinical problems of the patient may have been related to the side effects of prednisone, which had been taken for 20 years in doses ranging from 5 to 75 mg daily. In the words of his rheumatologist: "This patient has never had a remission, in the classic sense, of rheumatoid arthritis and no patient I'm aware of has taken prednisone for as long a period and lived."

The erythrocyte sedimentation rate ranged from 2 to 34 mm/h during the observation period, but showed a persistent elevation in recent years. A chronic, persistent anemia and moderately elevated white cell count had been present for many years. Lipid values and measures of pulmonary function were within normal limits. Body weight varied between 71.7 and 64.9 kg (height, 175.3 cm) during the observation period. Percent body fat ranged from 16.4% initially to a low of 13.5% in 1993 to a high of 17.7% in 1995. No significant electrocardiogram angles or related symptoms were observed during a multistage exercise test, and initially excessive diastolic pressor responses during exercise testing resolved.

### Discussion

Re-education in basic movement patterns was necessary before the patient could attempt formal exercise. Early triumphs were lifting his forearms,

writing, dialing a telephone, turning a doorknob, walking, and riding a stationary bike. He modified or designed splints, clothing, exercise equipment and self-help devices and learned to distinguish between neural, arthritic, and muscle pain. When sufficient progress had been made, a supervised program of cardiovascular, flexibility, and progressive resistance exercise was begun. Cardiovascular training consisted of a 6-minute warm-up, 45 minutes on a specially adapted stationary bike, a summation of three 15-minute intervals achieved in gradual fashion, and a 6-minute cool-down period. The exercise training program other than the warm-up/cool-down parts could be reduced to as little as 10 minutes when there were pain flare-ups. Training heart rate range was set at 85% of a multistage exercise test peak heart rate minus 10 beats per minute (6). The exercise program was changed when necessary to adjust for the pain threshold, impairment in the range of joint motion, and improvements in physical condition. A similar approach has been taken with flexibility exercise (7) and progressive resistance exercise (using resistance machines and free weights) (8). Resistance and flexibility exercises were also performed at home with equipment designed by the patient.

The patient exercised at the Human Performance Laboratory 2 to 5 times weekly. There were periods, as evidenced by the extensive medical history, when exercise was not possible. The exercise program was then modified and gradually re-established. This pattern was followed for 15 years. Figure 1 shows the pattern of multistage exercise test workload responses. Performance doubled from 400 to 800 kilopondmeters/min (kpm/min) in the period 1980 to 1983. Subsequent test responses failed to achieve this level, but all exceeded the initial test performance. Figure 2 reveals a similar metabolic pattern. Physical work capacity is displayed in metabolic units, or Mets where 1 Met = 3.5 ml O<sub>2</sub>/kg/min. Physical work capacity almost tripled in the first 3 years (2.8 to 7.6 Mets). The



reductions seen in 1988, 1992, and 1995 (7.0, 6.0, 6.3 Mets, respectively) suggest regression, and the observed reduction in metabolic status may well have been related to progression of rheumatoid vasculitis and the side effects of long-term medication. However, the patient's physical work capacity continued to exceed that observed before the start of the program. Significantly, 5 Mets is the level established by the Social Security Administration for disability classification, and although the gains observed for this patient were modest, the exercise program, in the opinion of his rheumatologist, "...has improved his exercise tolerance considerably and has benefited him emotionally."

To conclude, despite the fact that low work capacity and muscular fitness are common in these patients, exercise has been controversial as therapy for rheumatoid arthritis. Nordemar et al. (9) reported that "...As in many other diseases, the general prescription of rest in rheumatoid arthritis is not adequate" and a better disease outcome is consistently observed in active patients. Similar reports suggest that exercise frequency is not related to the activity of rheumatoid arthritis or to radiological progression of joint destruction (10-12), and it is well known that exercise counters the muscle-wasting effects of prednisone (13). Indeed, Rall et al. (14) reported that exercise can enhance protein synthesis and reverse the reduced functional status relative to muscle weakness in patients with rheumatoid arthritis. Other findings clearly show that physical activity and related lifestyle interventions can significantly reduce premature all-cause mortality and disability, improve quality of life, and increase chances for longevity in the population at large (15) and would be presumed to benefit patients with rheumatoid arthritis as well.

Figure 1

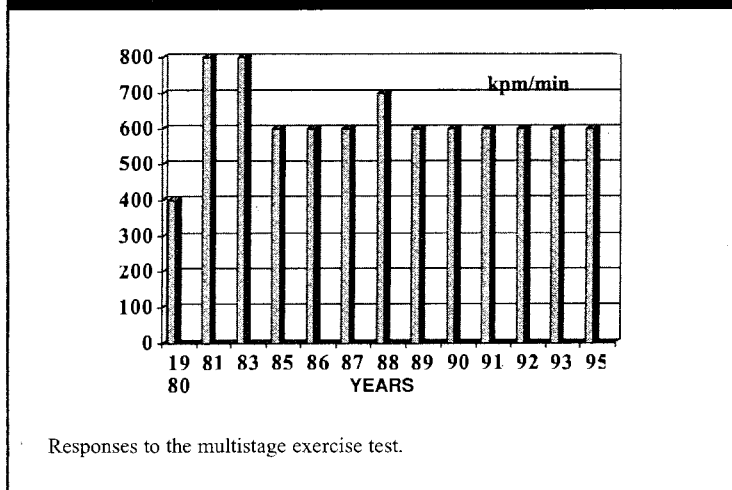
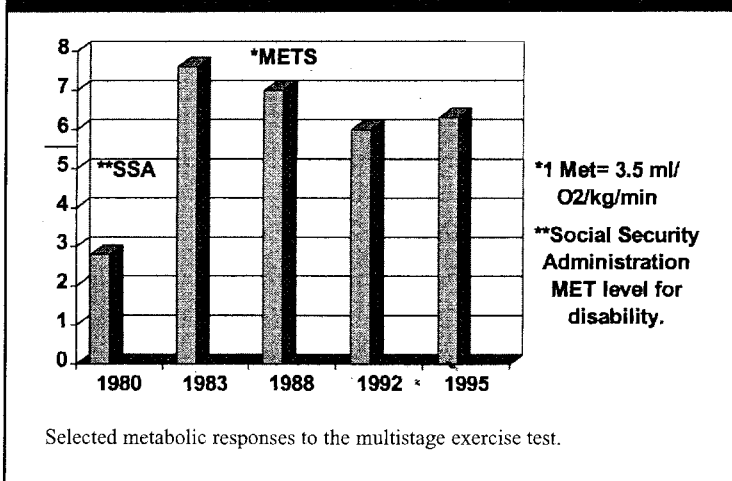


Figure 2



### EDITORIAL BOARD COMMENT

The remarkable lesson that this case report teaches is the importance of always optimizing a program of physical exercises in patients with rheumatoid arthritis. Awareness of this among physicians and health professionals is far from universal. However, it should not be neglected even in cases like this. Rheumatoid arthritis is complicated by vasculitis with motor deficiencies. In particular, mononeuritis multiplex is known to be a life-threatening condition, and apparently the patient received aggressive medical therapy including high doses of glucocorticoids. This probably left him with some degree of accelerated arteriosclerosis and caused premature cataract.