

Polymyositis with biological false-positive serological test for syphilis

A case report

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Summary

A young female came to the clinic with polymyositis and a biological false-positive serological test for syphilis (BFP reaction). Polymyositis, like other connective-tissue diseases, should be considered in the study of BFP reactors.

Introduction

The diagnosis of chronic biological false-positive reaction (BFP) is established in a person when the lipoidal serum tests for syphilis are positive for 6 months or more, the specific treponemal tests remain negative, and there is no clinical or epidemiological evidence of syphilis (Harvey and Shulman, 1974). The chronic BFP phenomenon has been recognized in autoimmune diseases, ageing, narcotic addiction, leprosy, and macroglobulinaemia (Shulman and Harvey, 1964; Harvey and Shulman, 1974; Drusin, Litwin, Armstrong, and Webster, 1974). Besides the well-defined association between the BFP reaction and systemic lupus erythematosus (SLE) (Harvey and Shulman, 1974), and also between it and Hashimoto's thyroiditis (Shulman and Harvey, 1964), the reaction has occasionally been encountered in other diseases with a definite or possible immunological basis, such as Sjögren's syndrome, rheumatoid arthritis, Evan's disease, haemolytic anaemias, pernicious anaemia, and myasthenia gravis (Harvey and Shulman, 1974). The present report concerns a young female with primary inflammatory polymyositis and the BFP phenomenon.

Case report

A 19-year-old single female was seen in July, 1974, when she had had muscle weakness for 2 years. The weakness had been progressive and associated with proximal muscle aches and tenderness. By May, 1974, she had difficulty climbing stairs and combing her hair. She had no fever, arthralgia, Raynaud's phenomenon, skin rash, photosensitivity, alopecia, mouth ulcers,

dysphagia, or pleurisy. She also denied any sexual exposure. The erythrocyte sedimentation rate (ESR) was persistently above 70 mm/1st hr. One physician whom she had seen, suspecting that she might have a collagen disease, requested many L.E. cell and antinuclear antibody tests (ANA); all proved negative.

When seen at this hospital she looked thin but was in no distress. The skin showed no thickening, erythematous rash, telangiectases, or splinter haemorrhages. The oropharynx was clear, and the fundi were normal. No adenopathy or visceromegaly were detected. The cardiovascular system was normal and the chest was clear. None of the joints showed signs of acute or chronic inflammation. The proximal muscles of both upper and lower extremities were weak, atrophic, and minimally tender, especially over the deltoids. Neurological examination was negative. The blood pressure was 130/75.

Laboratory investigations

Haemoglobin: 12.2 g per cent., packed cell volume 35 per cent., white blood cells; 5100/cu.mm, neutrophils 56 per cent., lymphocytes 43 per cent., eosinophils 1 per cent.

Urine showed no cells and no protein.

ESR 80 mm/1st hr. Serum electrolytes normal. Total serum proteins 6.8 g per cent. (Albumin/Globulin ratio 3.6/3.2).

BUN: 18 mg per cent., SGOT 19; SGPT 6; LDH 370; CPK 3; Aldolase 5 units (all normal). ANA (using mouse liver and human white bloodcells as substrates), CH₅₀, C₃, L.E. cell (×3), anti-DNA titre, RA latex, and test for cryoglobulins were all normal or negative. T₄, T₃ resin uptake, prothrombin time, and partial thromboplastin time normal. Toxoplasma titre normal. VDRL positive (titre 1/4). TPI, RPCFT, and FTA-ABS tests negative. All serological tests were done on duplicate samples. Schirmer's test showed normal tearing. Electromyography showed a myopathic pattern with predominant polyphasic potentials. Muscle biopsy (from the deltoid) was compatible with inflammatory myositis.

Treatment

She was started on 40 mg prednisone/day as a single morning dose along with physiotherapy. In 40 days, the ESR dropped to 21 mm/hr and the VDRL became negative. When seen in November, 1975, and while on 12.5 mg prednisone/day, her muscular strength was normal. The VDRL, determined on her brother and sister, was negative.

Discussion

The diagnosis of polymyositis in this patient is well established. Her normal muscle enzymes are not remarkable and can be explained on the basis of long-standing inflammation and atrophy of the muscle mass. The duration of the positive results to the VDRL before her admission to our hospital could not be determined. However, in the absence of non-treponemal infections that are known to be associated with an acute BFP reaction, and because of the presence of polymyositis, a disease associated with immunopathology, the BFP phenomenon was probably chronic.

None of 500 cases of chronic BFP reaction in the literature had polymyositis (Berglund and Carlsson, 1966; Wuepper, Bodily, and Tuffanelli, 1966; Putkonen, Jokinen, Lassus, and Mustakallio, 1967; Catterall, 1972; Harvey and Shulman, 1974). However, it is conceivable that some of these reactors whose basic disease was SLE, Sjögren's syndrome, or 'possible connective-tissue disease', could have had myositis as part of their primary autoimmune disease.

The demonstration of the BFP phenomenon in a young female should suggest SLE (Harvey and Shulman, 1974). In our patient, the absence of cutaneous and major organ involvement, constitutional symptoms, and the repeatedly negative tests for L.E. cells and antinuclear antibodies over more than 2 years virtually excludes SLE. Likewise, Sjögren's syndrome seems unlikely considering the lack of the sicca symptoms, and the normal Schirmer's test.

In contrast to its defined relation to SLE, the BFP reaction is not recognized as a feature of polymyositis (Logan, Bandera, Mikkelsen, and Duff, 1966; Pearson, 1972). Of 21 cases of polymyositis seen at our hospital and for whom serological data are available, only our patient had a BFP reaction. The occurrence of the BFP phenomenon in polymyositis may not differ from its occurrence in the presumably normal population. However, there is convincing evidence of an immunological pathogenesis of polymyositis suggesting that the occurrence of the BFP reaction in this disease may be significant. Deposits of immune complexes (IgM, IgG, and C₃) in intramuscular blood vessel wall (Whitaker and Engel, 1972), cell-mediated

cytotoxicity to muscle fibres (Dawkins and Mastaglia, 1973), lymphotoxin formation of lymphocytes (Johnson, Fink, and Ziff, 1972), mononuclear cell infiltration in muscles, and the responsiveness of the inflammatory process to steroids and other immunosuppressive agents indicate an immunological basis for the muscle inflammation.

Although the exact nature of the BFP reaction is not known, there is evidence that it represents a state of immunological hyper-reactivity (Catterall, 1972). Its association with autoimmune disorders, and the demonstration in BFP reactors of a variety of autoantibodies and other abnormal serum proteins (antinuclear, antimitochondrial, and antithyroidal antibodies as well as cryoglobulins) (Harvey and Shulman, 1974) support this concept. Thus the combination of the BFP phenomenon, an immunological reaction, with polymyositis, a disease with immunological features, seems more than a chance occurrence. In a young female with chronic BFP reaction, and in the absence of other features of SLE, polymyositis should be considered.

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