Case report

Destructive arthritis associated with acne fulminans: a case report

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SUMMARY Arthralgias have previously been reported in association with acne fulminans, but the following case is the first report of osseous lesions associated with this disease. The destructive bony lesion was rapid in onset but self-limited.

Arthralgias associated with a fulminant ulcerative type of acne were first described by Burns in 1959. Since then other cases have been reported.2–4 Patients typically have had mild cases of acne vulgaris for about a year before the onset of their acute symptoms. For reasons unknown at present this chronic acne suddenly becomes extremely aggressive, manifesting itself as an acute ulcerative process associated with haemorrhage into the dermis with full-thickness loss of skin and also with a systemic disease, including fevers, weight loss, increased sedimentation rate, myalgias and polyarthralgias, and occasionally microscopical haematuria.6 7

Arthralgia is typically asymptomatic, self-limited, and nondeforming in nature. However, in the case presented here, in addition to the arthralgias typically seen in this disease and associated with normal radiographs, the patient had a painful, rapidly destructive albeit self-limiting aseptic inflammatory lesion of the cervical spine.

Case report

The patient was a 13-year-old male with approximately a 1-year history of mild acne which began to increase in severity in August 1977. He was seen at the Dermatology Clinic at the University of Michigan in the following month and was treated with local skin care and systemic antibiotics. Over the next month despite his therapy he developed a low-grade fever, and his acne lesions worsened.

On admission his physical findings were confined to his skin and neck. His skin lesions were described as extremely painful, confluent, violaceous ulcers with crusting. He complained of pain in his neck, had a limited range of cervical spine motion, and was sensitive to palpation over the midcervical spine posteriorly. Laboratory tests on admission included a leucocyte count of 16.9 x 10⁹/l, a haemoglobin of 13.4 g/dl, a haematocrit of 39.9%, an erythrocyte sedimentation rate (ESR) of 44 mm/h, an alkaline phosphatase of 142 IU (normal 30–115), and a normal protein electrophoresis. No blood, protein, or glucose was found in his urine. The patient had no prior history of significant systemic illnesses and had no known allergies. His chest x-ray on admission was normal, but films of the cervical spine demonstrated a questionable defect in the anterior body of C4 (Fig. 1). A diagnosis of acne fulminans was made and a corticosteroid was added to the patient’s treatment regimen of systemic antibiotics and topical skin care with local antibiotic solutions, drying and debriding agents.

However, on his 17th day after his admission to hospital the patient became acutely ill, with temperatures of 104–105°F (40–40.5°C). The ESR was 59 mm/h, leucocytes 16.9 x 10⁹/l, haemoglobin 11.8 g/dl. His skin lesions became increasingly painful and more ulcerations appeared. He complained of myalgias and arthralgias, especially of his hip and neck. An ASO titre of 150 and a positive monospot test were obtained. Tests for rheumatoid factor and antinuclear antibodies were negative.

Multiple urine and multiple blood cultures were negative. Lesions of the skin yielded only a few coagulase-negative staphylococci. His chest x-ray remained clear. Films of his hips were normal. Repeat cervical spine films showed progression of
the lytic area of C4 with loss of the C5 body and decrease in the height of the C4–C5 disc space. No instability was noted on flexion and extension films. His haemoglobin fell to 9.8 g/dl. Multiple cultures of blood, urine, and throat continued to be negative. A needle aspirate of the C4–C5 disc space done under radiographic control was negative.

Over the next few days the patient remained extremely toxic and developed upper extremity weakness, with an electromyogram showing fibrillations and positive waves in the anterior and posterior myotomes of the C5 through T1 area on the right and with normal motor and sensory conduction studies in the right upper limb. A myelogram was done with clear cerebrospinal fluid (CSF) found and no blockage to the flow of the dye. Cultures of the CSF were negative. A bone scan showed increased uptake in the cervical spine.

All systemic medication was discontinued by the second day of his febrile attack, but local wound care of his acne was continued. The patient remained toxic for several weeks, and then despite having no new medication or alteration in the local wound care for his acne lesions he slowly began to improve, his skin lesions began to clear, and his constitutional systems resolved. However, the radiographic defects in this cervical spine persisted. To rule out the possibility of a subacute infection, open biopsy of the C4 body and the C4–C5 disc space was performed.

For biopsy the surgeons used an anterior cervical approach but found no purulent material at the site of the lesion. They felt confident that the biopsy of the bone of C4 and the disc space of C4–C5
represented an adequate sampling of the tissues involved. The pathological diagnosis of this material was: 'foci of chronic inflammation in dense fibrous connective tissue ... fragments of hyaline cartilage.'

The patient was discharged on 23 October 1977. Over the next 18 months he had progressive healing of his skin condition and spontaneous fusion of C4–C5 vertebrae with probable fusion of the body of C3 to C4. He has had no recurrence of his cervical pain, and his strength and sensation in his right upper extremity is now normal.

Discussion

The aetiology of this destructive process in the boy's cervical spine is unclear. With negative cultures and a pathological report of chronic inflammation from what clinically was thought to be an adequate section of both diseased bone and disc space one can make the diagnosis of aseptic inflammatory disease of the cervical spine. Since the occurrence of this boy's cervical lesions was correlated with the rapid development of his other systemic symptoms in association with the change of his skin disease from a moderate form of acne to a rapidly destructive lesion, his arthritis probably represents another manifestation of acne fulminans. This case extends the range of musculoskeletal involvement seen in this devastating disease. In acne fulminans the host appears for some unknown reason to begin to destroy his own tissues—his skin (ulcerative full-thickness skin loss), his kidneys (haematuria), his marrow (anaemia), and his bones. Perhaps if muscle biopsies were done on these patients, necrosis of muscle tissue associated with an intense inflammatory response would be found to correlate with the symptoms of myalgia.

Authors have suggested that acne fulminans may be an autoimmune disease or a hypersensitivity reaction much akin to the Arthus or Schwartzmann reaction, with the trigger antigen being either part of the bacteria that colonise the skin in acne or altered skin antigens secondary to a persistent low-grade infection. Factors in favour of an autoimmune aetiology of acne fulminans are the rapid response of the patient to systemic steroids, the increase in gamma globulins, and the decrease in C3 complement levels in several patients. The condition appears to benefit from debridement of necrotic material from the lesions, as if this manoeuvre helps to decrease the initiating factors. The lack of improvement on systemic antibiotics and the absence of an organism on multiple cultures make a diagnosis of sepsis unlikely.

This boy's course is interesting not only because it represents the first report of bony destruction associated with acne fulminans, but also because the patient improved dramatically over 3 to 4 weeks without the help of systemic antibiotics or steroids. Treated only with local wound care for his acne, he showed improvement in his musculoskeletal state and radiographic evidence of stabilisation of the destructive osseous process.

References

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