FISTULOUS RHEUMATISM AN UNUSUAL COMPLICATION OF RHEUMATOID ARTHRITIS

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THE term 'fistulous rheumatism' was given by Bywaters (1953) to an uncommon complication of rheumatoid arthritis in which chronic skin sinuses with low grade inflammation occurred near the affected joints. His description of two cases appears to be the first record of this condition. Another patient with chronic arthritis who developed suppurating tenosynovitis was reported by Arlet (1961). The histological appearance of the tissue from the sinus resembled that described by Bywaters but the presence of a tophus in the ear led the author to postulate an association between the fistulae and gout. The apparent rarity of fistulous rheumatism has prompted us to report a further case.

Case Report

A 78-year-old spinster was transferred to the Geriatric Department of University College Hospital in 1955 with progressive polyarthritis of several years' duration. The joints of the left hand, then the right hand and wrist were first affected, followed by involvement of knees and ankles. Since 1957, she has been unable to move from a chair, and now has instability of her knees and little movement of the wrists and shoulders. She has been under continuous observation for the past six years in the Geriatric Unit. In 1944, she had a right mastectomy for a colloid carcinoma of the breast but there has been no recurrence.

Initial investigation in 1955 revealed Hb. 77%; ESR 72 mms./hr. (Westergren); plasma albumin 3.7 g./100 ml.; globulin 3.4 g./100 ml., gamma-globulin increased. The X-ray appearances were those of a fairly active rheumatoid arthritis with partial destruction of the radio-carpal joints and the metacarpo-phalangeal joints. There was some destruction of cartilage and erosion of the articular margin of the bones.

In 1957, small yellow lesions appeared under the skin of the proximal interphalangeal joints of the left index and middle fingers and over the medial aspect of the right hallux. Further X-rays showed gross destruction of all the metacarpo-phalangeal joints with large punched-out areas in the heads of the metacarpals and in the heads and bases of the phalanges. In some of the joints there were discrete fragments which had apparently separated from the parent bone. Four months later, fluctuant swellings developed around both elbows. There was no pyrexia, but the white cell count showed a moderate leucocytosis (15,000/cu. mm.). Aspiration of the swelling over the right elbow yielded purulent but sterile fluid. No antibiotic was given and the abscess pointed and discharged sterile fluid one month later.

During the last three years, sinuses have appeared on the dorsal surfaces of the hands and feet (Fig. 1). These have all discharged sero-purulent fluid for several weeks or months and usually healed with local antiseptic treat-

ment, leaving depressed scars. Repeated cultures from these sinuses were either sterile or yielded *staph. pyogenes*.

At present, there are discharging sinuses over the metatarso-phalangeal or interphalangeal joints of the first and second left metatarsal bones. Scars of healed sinuses are related to the interphalangeal joints of her left four fingers and two of the fingers of the right hand.

Further Investigations

The serum uric acid has been estimated on several occasions during this time, and has never exceeded 9 mg./100 ml.; Hb. is now 74%; WBC 6,700/cu. mm.; blood urea 40 mg./100 ml.; WR negative. LE cells have not been found on repeated examinations. The Rose-Waaler test was weakly positive at a titre of 1/8 and the Latex test was negative. Serum cholesterol 185 mg./100 ml.; plasma proteins were 9.0 g./100 ml.; alpha₂ and gamma-globulins increased.

Recent X-rays (Fig. 2) show more loss of density, with further disorganisation of the joints of the wrist and hands. In several of the fingers there is well marked calcinosis circumscripta of the soft tissues surrounding the inter-phalangeal and metacarpo-phalangeal joints.

The nature of one of these areas of calcinosis has been studied by surgical exploration. This revealed a thin-walled cystic swelling, arising from a proximal interphalangeal joint, containing cheesy white material. Histology of the cyst wall showed fibrous tissue with patchy calcification but with no specific changes of rheumatoid arthritis. Acid-fast bacilli were not seen in the amorphous debris in the cyst and the murexide test failed to show the presence of urates.

Injection of one of the sinuses on the dorsum of the foot with radio-opaque material showed a track extending to the first metatarso-phalangeal joint.

Biopsy of the wall of a sinus showed that, under the epidermis, there was an area of eosinophilic necrotic tissue surrounded by a zone of proliferating fibroblasts with a few small giant cells and lymphocytes. A palisade appearance of the fibroblasts was present in some areas adjacent to the necrotic tissue. These findings were considered to be consistent with rheumatoid disease. A small fragment of necrotic bone was also present in the tissue excised (Fig. 3).

Discussion

The outstanding features shown by this patient are a progressive mutilating arthritis, recurrent skin sinuses near joints and the radiological appearance of bone cysts. One of the sinuses has been shown by injection of the track to be connected with a neighbouring joint.

Although the diagnosis of gout has been suggested, there has never been any evidence of urate deposits in the sinuses or in the skin nodules. The

FIG. 1.—Left hand showing scars of healed sinuses and left foot showing discharged sinuses.

serum uric acid has never been raised. A prolonged course of Probenecid neither influenced the skin lesions nor the serum uric acid level; nor did it provoke an acute exacerbation of arthritis. The possibility of disseminated lupus erythematosus cannot be excluded, but the general well-being of the patient after six years, the absence of the L.E. cell phenomenon and a persistently normal blood urea, make this unlikely. The ubiquitous nature of the sinuses and the failure to isolate tubercle

FIG. 2.—Recent X-ray showing disorganized joints, sub-articular cysts and calcinosis circumscripta.

FIG. 3.—Biopsy from wall of sinus, showing fragment of necrotic bone. $\times 44$.

bacilli from the joint aspirates rule out a tuberculous arthritis.

The clinical picture is consistent with rheumatoid arthritis, although the Rose-Waaler test is only weakly positive. The continued pain and elevated ESR suggest that there is persistent inflammatory or destructive activity in the joints. Radiological deterioration in the joints has been associated with an increase in the number of subarticular cysts. Another unusual radiological feature is the occurrence of areas of circumscribed opacities in the soft tissues of the fingers. These lesions consisted merely of amorphous debris but no urate deposits were detected.

These findings are in many ways similar to those described in Bywaters' patients. As in our patient, staphylococci were occasionally isolated from the sinuses. He postulated that bone cysts were formed by the propulsion of synovial fluid into the cancellous bone under the inflamed joint surfaces which were denuded of cartilage. Necrotic fragments of bone resulting from stress microfractures were extruded from the cysts and were eventually discharged through the skin, forming sinuses. The track, at first sterile, might become secondarily infected with staphylococci. In the case described above, bone cysts are associated at some sites with thin-walled sacs beside the shafts of the bone. It is

possible that debris from the joint or from destroyed subarticular bone has remained in the subcutaneous tissue and has become surrounded by a fibrous wall.

We conclude that these chronic sinuses and multiple bone cysts are manifestations of the destructive features of rheumatoid arthritis. Steroid therapy has not been given to the above patient because of her already immobile state, her age, and the relative lack of severe symptoms, and it is not yet known whether the chronic destructive process could be suppressed by such treatment.

Summary

A patient with rheumatoid arthritis is described in whom fistulae have developed near affected joints. Investigations have shown that the fistulae communicate with neighbouring disorganised joints. Rheumatoid changes and necrotic bone were demonstrated in the wall of such a fistula.

It is postulated that bony sequestra, detached from severely involved joints, are discharged to the surface through these fistulae.

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PASTEURELLA SEPTICA MENINGITIS WITH SURVIVAL

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HUMAN infections with *Pasteurella septica* have been divided by Talbot and Sneath (1960) into two groups, namely, those with and those without a history of animal bites. The majority fall into the first group and usually show local infection and adenitis (Allott, Cruikshank, Cyrlas-Williams, Glass, Mezer, Straker and Tee, 1944). Infections in the second group involve the respiratory tract (Cawson and Talbot, 1955) or take a systemic form with meningitis or bacteraemia (Schwartz and Kunz, 1959; Bearn, Jacobs and McCarty, 1955).

Meningitis due to *P. septica* is extremely rare. We have been able to trace only 14 reported cases (Regamey, 1939 (four cases); Le Chuiton, Bideau and Pennaneac, 1939; Kapel, 1942; Fölmer and Have, 1943; Tomic-Karovic and Ivanonic, 1944; Zeller and Lepper, 1950 (two cases); Lewis, 1953; Ewan, 1955; Swartz and Kunz, 1959; Talbot

and Sneath, 1960). Six of these cases were fatal. With the exception of the case mentioned by Talbot and Sneath (1960), there has been no report of *P. septica* meningitis in this country.

In this paper a case of *P. septica* meningitis with recovery in a Mongol infant is reported.

Case History

A Mongol girl aged six months was admitted to Lewisham Hospital on 2.11.61, having fed poorly and vomited for 24 hours. She had had five convulsions in the 12 hours prior to admission. The mother's pregnancy and delivery were normal, the child was bottle fed and her weight on admission was 14 lb. 10 oz.

On examination the temperature was 102.5°F., pulse 160/min., the anterior fontanelle was full and the fauces slightly reddened. There was doubtful reddening of the left ear drum. Lumbar puncture was performed at once. The CSF was turbid, there were 19,500 poly-