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Second Opinion, Please

Ankylosing Spondylitis

T. R. CULLINAN, BASIL CHRISTIE

British Medical Journal, 1972, 2, 400-401

Sydenham House, Ashford, Kent

Dear Basil,

This boy, E. F. aged 19, came to see me last week complaining of increasing pain in both tibial heads, which had been coming on for several weeks. He has no relevant personal or family history and, apart from minor complaints, has always been well.

As you will see, both tibial heads are clinically swollen anteriorly and tender to touch, though they are not markedly hot. X-ray shows no radiological change but I do not think we can escape a presumptive diagnosis of Osgood-Schlatters disease. There is no joint involvement.

The pain is now such as to affect his work as a carpenter's apprentice.

Kent and Canterbury Hospital, Canterbury

Dear Tim,

Thank you for asking me to see this patient who, I agree, appears to have bilateral Osgood-Schlatters disease. This is unusual at this age, though the upper ossification centre of the tibia may not join until the twentieth year.

X-rays of the knees show no abnormality, so I am treating him symptomatically with some short-wave therapy.

Sydenham House, Ashford, Kent

Dear Basil,

You will recall treating E.F. a few months ago for Osgood-Schlatters disease. He responded fairly well to short-wave

therapy and was left with residual stiffness only, which has been helped by indomethacin.

He now complains, however, of left hip pain which is not entirely related to the stiffness in his knees. Hip movement was full and painless today, and his hip x-ray shows no signs of Perthes's disease.

Kent and Canterbury Hospital, Canterbury

Dear Tim,

I think this boy's pain felt in his left hip, which has been present three weeks, almost certainly is referred from his back. The pain is aggravated by lumbar movements and there is some pain on full straight leg raising on the left. The hip movements are entirely normal.

The x-rays you ordered of the left hip are normal; I have had some films taken of his sacroiliac joints and these are also normal.

I am arranging some lumbar traction for him, as I think his pain must be coming from his back.

Sydenham House, Ashford, Kent

Dear Basil,

The lumbar traction you gave E.F. four months ago solved his backache for a time, but now it has returned and he also complains of a right shoulder joint pain.

Although his E.S.R. was only 15 last week, I very much fear we may be dealing with a more generalized disease than at first we thought.

Kent and Canterbury Hospital, Canterbury

Dear Tim,

This boy now has pain in his right shoulder and a recurrence of his backache. The only abnormality on examination was some tenderness of his tibial tuberosities. The remainder of the skeletal system was normal. I repeated his blood picture

Ashford, Kent

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which was normal apart from an ESR of 34. An x-ray of his right shoulder was normal but films of his sacroiliac joints show changes now of ankylosing spondylitis.

I would suggest putting him on to phenylbutazone, 100 mg three times a day.

Sydenham House, Ashford, Kent

Dear Basil,

It is now nine months since last you saw this boy and during that time he has been able to remain at work though sometimes in considerable pain.

He was not able to tolerate phenylbutazone, even with the addition of alkalis, but found Tanderil very helpful for several months. Five months ago, his E.S.R. was only 8, with a normal blood count, but now it has risen to 58.

He can now no longer tolerate Tanderil because of gastrointestinal symptoms, and indomethacin suppositories have failed to help him. His back, until now, has been more troublesome than his knees, but these too have now flared up again and he has had to stop work. I hesitate to put so young a lad on steroids, but feel we are being forced towards the point where these must be considered.

Do you think ACTH would be better and safer than adrenal corticoids?

Kent and Canterbury Hospital, Canterbury

Dear Tim,

In view of this boy's gastric intolerance to the butazones and the increasing activity of his ankylosing spondylitis, I agree that we shall have to take stronger measures. Personally I am against irradiation as it carries a definite risk, and its results are uncertain.

I would certainly prefer ACTH to oral steroids and would suggest 20 units daily for one week, then reducing to 10 units daily.

Sydenham House, Church Road, Ashford, Kent

Dear Basil,

This boy has now been on ACTH for four months with almost complete remission of his knee symptoms and some definite improvement to his back.

He now, however, has begun to have diarrhoea with occasional blood and pus, and I am fearful of the possibility of ulcerative colitis. Ordinary rectal examination reveals a small fissure-in-ano and I can see no evidence of proctitis with the proctoscope. I feel, however, that sigmoidoscopy is indicated.

Dear Basil,

You will recall that a barium enema and sigmoidoscopy by your surgical colleague two months ago revealed no evidence of ulcerative colitis. He did, however, find a fistula-in-ano.

E.F.'s admission at that time, away from the anxieties that were building up at home, helped the healing process to start and now, though he still has occasional bloody diarrhoea, his symptoms are certainly much better.

Dear Basil,

It is now nearly nine months since this boy has been in serious difficulty, and he is presently at a residential Government Training Centre. The short spell of rest in hospital, together with the gentle attention of the nurses, certainly speeded the healing of his bowel.

He continues on ACTH and his back, though still stiff, is much looser than a year ago. His knees give virtually no trouble and his bowels act normally.

Looking back over this case, one or two interesting questions arise.

Does ankylosing spondylitis often present as, or with, Osgood-Schlatters disease, because I am certain he had no back symptoms when first he presented with tibial pain? Also, I am sure there was no peripheral arthritis.

Secondly, do you think it is significant that the bowel complications eventually resulting in fissure-in-ano, appeared only after he had started on ACTH? I suppose that in the years to come he may well appear with more varied bowel disease—perhaps true ulcerative colitis or Crohn's disease.

Thirdly, what are the current thoughts on immunosuppressive therapy in this disease?

Kent and Canterbury Hospital, Canterbury

Dear Tim,

Thank you for your letter. I think E.F. is an instructive case and shows how ankylosing spondylitis can present other than in the back. Looking back through his x-rays I see the first was of his feet in 1966, when he was aged 16. On the request form at that time, you noted that he had "long-standing pain in his right foot." The x-ray was normal. This was eighteen months before he presented with his painful tibial tuberosities. It was on his second attendance in 1969 that my suspicions were aroused, as I had an x-ray of his sacroiliac joints. These x-rays were normal but when repeated a year later showed definite changes of ankylosing spondylitis. Whereas the classical presentation is of low lumbar pain with associated morning stiffness in a young person, it is not uncommon for painful peripheral joints, especially the heels and feet, to be the first presentation. Because these may precede the backache by several years, their real significance may be lost. This is the first time I have seen a case presenting as an Osgood-Schlatters disease.

I do not think that the ACTH can be incriminated for the bowel symptoms, but ulcerative colitis and regional enteritis, as you know, are seen in association with ankylosing spondylitis. It could well be that in the future he will develop more overt signs of enterocolitis. The other organs which may be affected are the iris which may be subject to recurrent iritis, and the heart and aorta giving signs of aortic insufficiency with signs of a prolonged P-R interval on the E.C.G.

I think too little work has been done so far on immunosuppressive drugs in ankylosing spondylitis to comment on this point. In rheumatoid arthritis the immunosuppressives have produced modest improvement, but they are drugs not without toxic effects and work on them in the rheumatic diseases is still at the experimental level.

It is a pity that ACTH was needed to control the situation in this case, but when other measures had failed we were really left with only the alternatives of this or irradiation. The latter gives symptomatic relief in about 75% of cases, but the relief is often short-lived and further irradiation adds to the danger of marrow aplasia.

I am pleased that E.F. is so well at the moment and re-training to be an instrument fitter.

Glossary

E.S.R.	Erythrocyte sedimentation rate.
Tanderil	Oxyphenbutazone.
ACTH	Adrenocorticotrophic hormone.
E.C.G.	Electrocardiogram.