

Hospital for Extrapulmonary Tuberculosis, Juelsminde, Denmark.

ACTINOMYCOSIS OF THE SPINE

Report of two cases

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The first description of *human* actinomycosis was given by Israël in 1878, and Israël & Wolff (1891) isolated anaerobic actinomyces in pure culture from patients and obtained takes on inoculation into animals.

Pus obtained from patients with actinomycosis contains one or more of four actinomyces strains. In about 50 per cent of all cases, *Actinomyces Israël* is present. However, most pus samples also contain other micro-organisms, especially *Bact. actinomycetem comitans*, anaerobic streptococci and *Esch. coli* (Holm 1950, 1951).

The portal of entry is the oral cavity, the respiratory or gastro-intestinal tract (then frequently the appendix).

The infection spreads in the directions where the tissue resistance is least, often along vessels and nerves, to the prevertebral space of the neck, the posterior mediastinum, and to the retroperitoneal space.

The skeletal system may become involved, mainly by contiguity, but apart from the jaw this is a relatively rare occurrence.

Grässner (1929) found osseous involvement in 15 per cent of 486 patients with the following distribution: spine 37 per cent; mandible, 25 per cent; ribs, 10 per cent; maxilla, 8 per cent.

The first comprehensive description of actinomycosis of the spine was given by Boström in 1891. His description is accompanied by a picture which in an excellent way illustrates the patho-anatomy of the disease which forms the basis of the radiographic findings.

Erving (1902) reported that the frequency with which actinomycosis involves vertebrae and ribs is 2 per cent. So far, 68 cases of spondylitis caused by actinomycosis have been published in the world literature.

A search of the Scandinavian literature has revealed four Danish cases, of which the first three had been subjected to autopsy (Ammen-torp 1893, Eiken 1942, Glahn 1950, Zacho 1942).

The patho-anatomical findings in actinomycosis of the spine have been described in detail by numerous authors (Parker 1923, Simpson & McIntosh 1927, Beitzke 1934, Meyer & Gale 1935, Cope 1951).

The spine is attacked by propagation of a paravertebral phlegmon. Erosion of the surfaces of several vertebral bodies occurs once the periosteum is penetrated. It is a feature characteristic of the individual vertebrae that the pedicles, transverse processes, and other parts of the arches are involved. The medial ends of the ribs may become eroded on account of propagation from the pleura or spine.

Only in rare cases does the propagating osteomyelitis give rise to destruction of intervertebral discs (Parker 1923, Haselhorst 1928, Flynn & Gillies 1938, Dixon 1939, Lubert 1944, Brett 1951, Young 1960) or to the development of external pachymeningitis.

Multiple ostia from abscesses may occur on the skin of the nape of the neck or back.

Even though, in the autopsy reports, the vertebral bodies are described as being soft on section, collapse with gibbus formation and medullary compression is rare. The explanation is probably that the osteoclastic destruction of the bony tissue is accompanied by a reactive formation of new bone, imparting to the vertebrae a moth-eaten, spongy, or honeycomb-like appearance.

Symptoms. Actinomycosis of the spine, as a complication of the cervicofacial, pulmonary-mediastinal or abdominal variety, is at first characterised by a remarkable stiffness of the involved vertebral segments, later possibly followed by neurological manifestations. In advanced cases, sinus tracts will be present especially on the nape of the neck and the back.

The diagnosis is based on the demonstration of the pathogenic actinomyces and the accompanying bacterial flora already mentioned. However, since the pathogenic strains of actinomyces are highly sensitive to penicillin and streptomycin, pus from patients treated with these drugs will reveal growth only of the accompanying bacterial flora.

Histological examination of a biopsy specimen may be helpful. Radiography must also be regarded as a valuable aid in the diagnosis; this applies, in particular, to tomography (Lubert 1944).

Radiographic Findings. The number of publications of the radiographic appearance of spinal actinomycosis is small as compared with those considering its pathological features (Parker 1923, Tabb & Tucker 1933, Lubert 1944, Young 1960).

The anterior and lateral surfaces of the vertebral bodies have an irre-

gular, saw-toothed appearance caused by periosteal new bone formation.

The central parts of the vertebrae are characterised by a honeycomb-like structure produced by destructions surrounded by newly formed sclerotic bone.

Very often the pedicles, processes, and arch, as well as the interarticular joints, are also involved. Compression of the vertebral bodies and destruction of the intervertebral discs rarely occur, and there are no sequestra or calcifications in the accompanying paravertebral abscesses.

In the radiographic differential diagnosis, tuberculosis should particularly be considered. Not infrequently, spinal actinomycosis is for a long time misdiagnosed as tuberculosis.

More remote diagnostic possibilities are: metastases in the spine, myelomatosis, osteitis deformans (Paget), syphilitic spondylitis and other bone mycoses.

Since a correct diagnosis is a prerequisite for adequate treatment, and as it appears from the literature that the differential-diagnostic possibilities are numerous, we find it of interest to report two cases of actinomycosis of the spine encountered in the Hospital for Extrapulmonary Tuberculosis, Juelsminde.

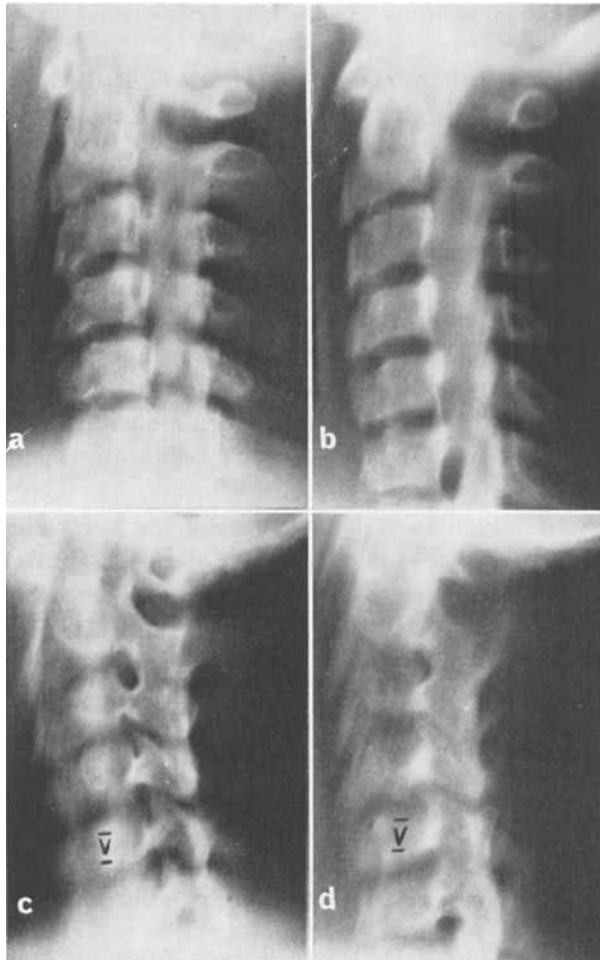
Case reports

Case 1. The patient was a girl, aged 17. For four years she had suffered from dental caries of increasing severity. During the last two years she had a number of healing but recurrent abscesses of varying localisation on the neck. At the same time increasing painless stiffness of the neck developed. A skin biopsy specimen from the margin of one of the abscesses revealed inflammatory infiltrations and actinomycetes grains.

On admission to the Hospital for Extrapulmonary Tuberculosis, Juelsminde, in November 1968, the patient was almost cachectic; weight 38.5 kg; height 154 cm. In the nape of the neck and on the upper part of the back there were scars of four healed abscesses and four fresh abscess perforations. There were no enlarged axillary or cervical lymph nodes. The neck was stiff, held in a slightly stooping position. E.S.R. 115 mm/h; Hgb 90 &; W.B.C. 10,950; temperature 37.2°C. No cough or expectoration.

Radiography of the spine, including tomography, revealed: a periosteal reaction of the spine and at the external lamina of the occiput; a honeycomb-like structure of the vertebral bodies in the cervical and the thoracic region as well as in the transverse processes and the upper ribs; ill-defined demarcations of several intervertebral spaces; obliteration of the joint space of the interarticular joint C₂/C₃ and a slight compression of the body of Th₃. Anteroposterior and lateral views demonstrated a paravertebral shadow.

Chest radiography revealed a large infiltration in the upper right lung field with



*Figure 1. Case 1, girl aged 17 years.
Cervical spine, tomography, lateral view.*

- a. Before chemotherapy. Jagged, periosteal new bone formation particularly at the anterior margins of the vertebral bodies of C₄-C₆.*
- b. After treatment. Organised ossification of the periosteal new bone formation. The trabecular structure of the bodies is normal.*
- c. Left apophyseal joints of the cervical spine. Before treatment. Articular margins irregular.*
- d. Same after treatment. Ankylosis between C₂/C₄ and C₅/C₇.*

Figure 2. Case 1, girl aged 17 years, before treatment. Thoracic spine, tomography, lateral view.

Irregular bone structure of the bodies, with rarefactions of varying size. Periosteal calcifications along the anterior margins of the upper part of the thoracic spine. No wedging of the vertebral bodies and no narrowing of the discs.

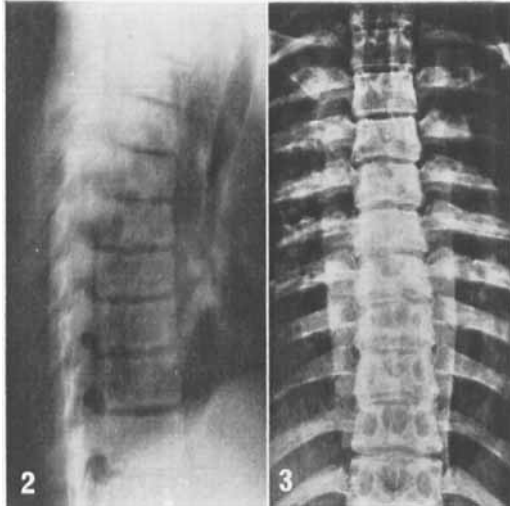


Figure 3. Case, 1, girl aged 17 years.

Thoracic spine, a-p view. Before treatment. A bilateral paravertebral abscess is visible.

a central radiolucent area and a paramediastinal infiltration in the left apex. On both sides, paratracheal lymph nodes were visible.

The dental status was poor, with extensive and deep-seated caries and periapical rarefactions in 6 + 1,2,6.

Cultures from a skin biopsy specimen and a resected rib yielded growth of actinomyces in one of the samples (type not determined).

Treatment. All teeth were extracted. Cultures gave growth of *Actinobacillus actinocyctem comitans* and *Bacteroides corrodens* from 6 + ,1,5,7.

The patient was treated with 0.75 g dihydrostreptomycin and 2 × 2 mill. units of penicillin daily for three periods each of 8 weeks at 10-day intervals.

The patient recovered. The abscesses healed, and the general condition improved steadily. Weight 51 kg; E.S.R. 11 mm/h.

Radiographs (April 1969) showed that the honeycomb-like structures in the spine and ribs had decreased. A periosteal bony bridge was visible along the anterior margins of the cervical bodies. Posteriorly, ankylosis between the small joints of several of the cervical vertebrae had developed, so that two blocks, each of three vertebrae, had formed.

Only remnants of the paravertebral abscess shadow was revealed. Lung markings normal.

As might be expected, the mobility of the spine did not improve. The patient is, and always will be, stiff in the neck and the upper part of the back.

Case 2. A male agricultural worker aged 42. In 1949, at the age of 24, he was hospitalized for two periods; the first time with a diagnosis of endocarditis, which was treated with penicillin and streptomycin.

The second time he complained of pain in the neck and the upper part of the back. As urinary cultures and inoculation into guinea pigs yielded growth of tubercle bacilli of human type, in 1951 he was transferred to the Hospital for Extra-

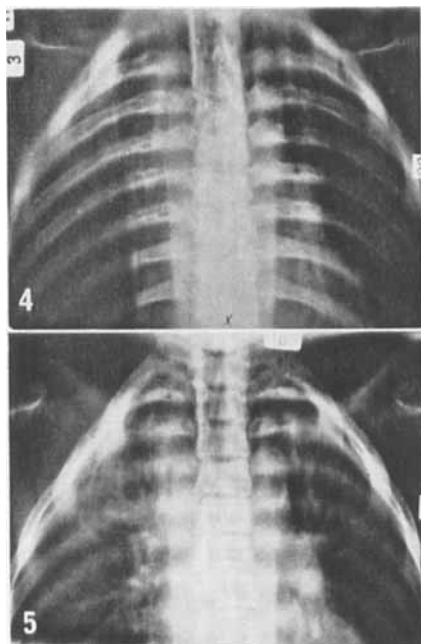


Figure 4. Case 1, girl aged 17 years. Posterior part of the ribs, tomography, a-p view. Before treatment. The medial part of several ribs shows a honeycomb-like appearance and marginal serration.

Figure 5. Case 1, girl aged 17 years. Apices of the lungs, tomography, a-p view. Infiltration and cavitation in the right apex. A para-mediastinal infiltration is seen in the left apex.

pulmonary Tuberculosis, Juelsminde, where he stayed for a year with a diagnosis of tuberculosis of the thoracic spine, the left kidney, and the lungs. Gastric washings revealed tuberculosis on four occasions, strangely enough of *bovine* type.

Radiography of the thoracic spine showed narrowing of the intervertebral discs between Th₄ and Th₈. No collapse of the bodies, but the anterior margins of Th₇-Th₁₀ were irregularly eroded. Large paravertebral abscess shadows were present.

The Albee operation was performed at the level of the middle thoracic spine, under the cover of penicillin and streptomycin.

In 1954, the patient underwent a thorough course of antituberculosis chemotherapy.

He then fared relatively well until 1964, when he was re-admitted because a recurrent abscess had appeared in the left lumbar region. Culture of pus showed growth of staphylococci, but no tubercle bacilli.

Radiographic of the thoracic spine showed ossification of the anterior longitudinal ligament at the involved vertebrae, and a large paravertebral abscess shadow. *Fistulography* through the fistulous orifice in the left lumbar region revealed entry of the contrast via the paravertebral abscess through a fistulous tract into the oesophagus.

This finding did not arouse suspicion, but merely confirmed the tuberculous aetiology.

Medication was started with Leder cyn® and Erythromycin®; later a regular course of antituberculous chemotherapy was given.

As the external fistula closed in March 1966, the patient was discharged.

In 1968, the patient was re-admitted with a new large abscess, this time in the

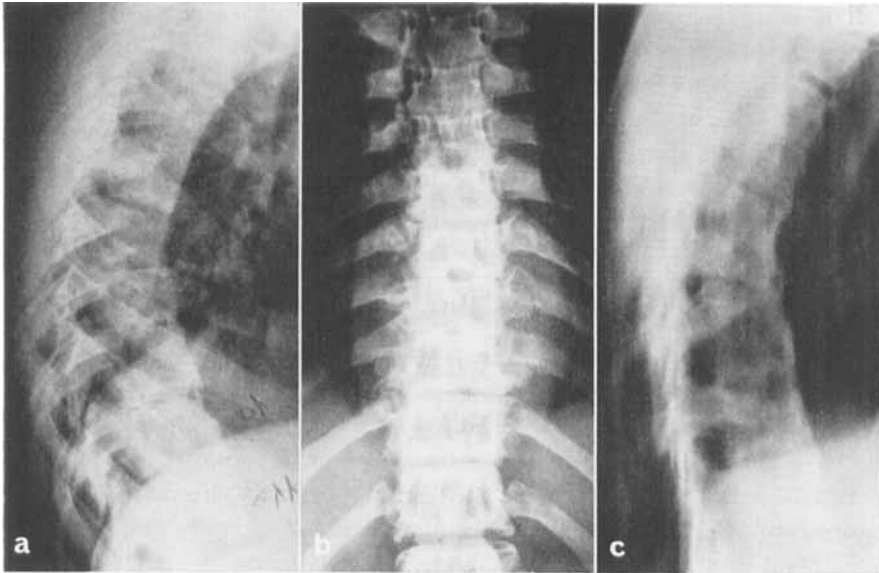


Figure 6. Case 2.
Thoracic spine (1951), lateral view.

a. Narrowing of several discs. Irregular anterior margins of vertebral bodies.

b. a-p view. Bilateral paravertebral abscess.

c. Thoracic spine, tomography, lateral view. (1968). Serrated, periosteal calcification anteriorly. Reticular structure of the vertebral bodies. Narrowing of several discs; only slight wedging of the bodies.

right lumbar region. Extirpation of the abscess was performed and a fistulous tract along the internal side of the transverse processes was revealed. *Histological examination* of tissue from the abscess showed *actinomycosis*, and a typical Wolff-Israël actinomyces was cultivated from the pus.

The patient was then given antituberculous chemotherapy for 6 months, still on the assumption that the basic disease was tuberculosis. The fistulae, both the old and the new ones, closed, and have not recurred.

A check-up examination in May 1969 showed a clinically good condition with E.S.R. of 5 mm/h. The patient was then able to work full time.

Radiographs showed normal paravertebral shadows, partial fusion of the lower thoracic segments, and a periosteal new growth. Only a slight bulging at the site of the healed fistula at the oesophagus was demonstrated.

DISCUSSION

Our first patient, the 17-year-old girl, had actinomycosis involving the cervical and thoracic spine, ribs and the lungs, and extensive dental caries, probably with actinomycotic foci in the maxilla.

The involvement of the spine must have been secondary to a paravertebral phlegmon, whose origin was the periapical infection or the pulmonary affection.

In this patient, the clinical picture was typical with stiffness of the spine and with sinus tracts to the skin. The diagnosis was secured by the demonstration of pathogenic actinomyces, biopsy with characteristic histological findings, and a radiographic appearance which is typical of actinomycosis.

Actinomyces was found in only one of the many samples studied, while the others yielded growth merely of the bacterial flora which usually accompanies this fungus. The explanation of this may be that previous chemotherapy had eradicated the highly penicillin-sensitive actinomyces, while the two accompanying micro-organisms are resistant to penicillin and sensitive only to streptomycin.

Whereas the first case must thus be regarded as elucidated, the demonstration of Wolff-Israël actinomyces in the second case gives rise to clinical and radiological diagnostic problems.

The second patient had for 18 years been conceived as suffering from a tuberculosis spondylitis, since tubercle bacilli had been recovered from both urine and gastric washings.

A biopsy from the *spine* with histological examination and culture of material, which would have solved the problem, was not performed.

As far as we know, co-existence of actinomycosis and tuberculosis has been described only once before (Glahn 1950).

In the light of the experience gained in our first case and by studies of the literature, we now believe that, in the second case, the primary disease must have been actinomycosis, which began in October 1949 with uncharacteristic symptoms. We interpret the presence of human tubercle bacilli in the urine and bovine bacilli in the lungs as a secondary infection which had gained a foothold because of the diminished resistance of the patient.

SUMMARY

On the basis of the literature a brief survey is given of human actinomycosis, with special reference to its vertebral manifestations.

Two cases are reported, making the total of published cases of actinomycotic spondylitis 70.

The first patient, a girl aged 17, suffered from typical actinomycosis of the lungs, the cervical and thoracic spine, and the upper ribs with

sinus tracts to the skin. The diagnosis was secured by culture of pathogenic actinomyces, biopsy, and characteristic radiographic findings.

The second patient, a man aged 42, suffered not only from actinomycosis of the thoracic spine, but also from renal tuberculosis of human type and pulmonary tuberculosis of bovine type.

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