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ON CYSTIC TUBERCULOSIS OF BONE

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It is to be expected in a place like Singapore where the rate of tuberculosis is still high that there will be a big variation in the individual resistance to this disease, and consequently the clinical and radiological appearances will vary as well. Cystic tuberculosis in bones is best regarded as either an expression of a high resistance already present where the lesion is walled off early, or of resistance rapidly acquired after that period in the spread of the disease which *Wallgren* (1948) has called the allergic phase, and where the disease shows up as a condition called multiple pseudocystic tuberculosis of bones. That the disease normally has a strong tendency to invade the neighbourhood is clearly shown by *Cholmeley* (1939), who studied the results of treatment in the pre-antibiotic period 1927–1937 of 55 cases with tuberculous lesions close to the hipjoint. 43 cases were treated conservatively and 12 with excision. Only 3 escaped with a mobile joint, 2 treated conservatively and 1 with excision. That surgery of localised lesions also under the pre-antibiotic time could be successful has been shown by *Ratliff* (1957), who has described 3 cases of tuberculosis of the femoral neck in childhood treated with drainage and curettage, and which all had full movement in the hip joint when seen 10 years later. The introduction of the antituberculous drugs has, of course, made radical surgical treatment much safer.

In this article 3 cases of solitary tuberculous lesions and 3 cases of multiple pseudocystic tuberculosis of bones will be presented. All are young children from 2½ to 4 years of age, and they are all Chinese.

The solitary cysts were preoperatively diagnosed as Brodie's abscess. This diagnosis was based on the history combined with the radiological picture which showed a cystic lesion well bordered with a sclerotic zone, and in one case containing a sequestrum. Sclerosis which means osteogenesis, in the neighbourhood of a tuberculous lesion occurs, as *Girdle-*

Fig. 1.

Fig. 2.

Fig. 1. Well defined cyst in the medial lower part of the neck of right femur.
Fig. 2. Caput and collum bigger than on the left side (Coxa magna). No subluxation or destruction. Time of observation 5½ years.

stone & Somerville (1952) has put it, "seldom except at a distance in space or time from an active tuberculous focus". In these cases, two of which occurred in the femoral neck and one in the lower tibial metaphysis, the length of the history is 5–12 months and the osteogenesis is in the immediate vicinity of the lesion.

Case 1. Male. 2½ years of age.

Pain and limp for 12 months. Fever the night before symptoms started. Better after 10 days, but limping persisted all the time. Since 1 month relapse of pain and also night-cries.

12.8.1953. Limping on the right side. Movements in the right hipjoint restricted in all directions.

Blood: BSR. 37 mm., Hb. 102 %. T.R. 4.75, T.W. 10.500.

Urine: Alb. +. P.C. 1–2, R.B.C. 1–2.

Mantoux positive 1/10.000.

Chest X-Ray: Right hilum is sligthly heavy with some calcifications.

X-Ray of right hipjoint: Well defined cyst in medial lower part of collum. Epiphysis slightly irregular. Joint space normal. (Fig. 1).

17.9.1953. *Curettage.* Performed through lateral approach.

Biopsy: Positive for tuberculosis.

Postoperatively treated with streptomycin, PAS and INH for 6 months combined with fixation, 2 months in hip spica and 4 months on abduction frame. This period of immobilisation was followed by 11 months in a leather support.

Last seen 5½ years after operation. There was then no limp or any complaints. Full movements in the hipjoint. The X-Ray showed that

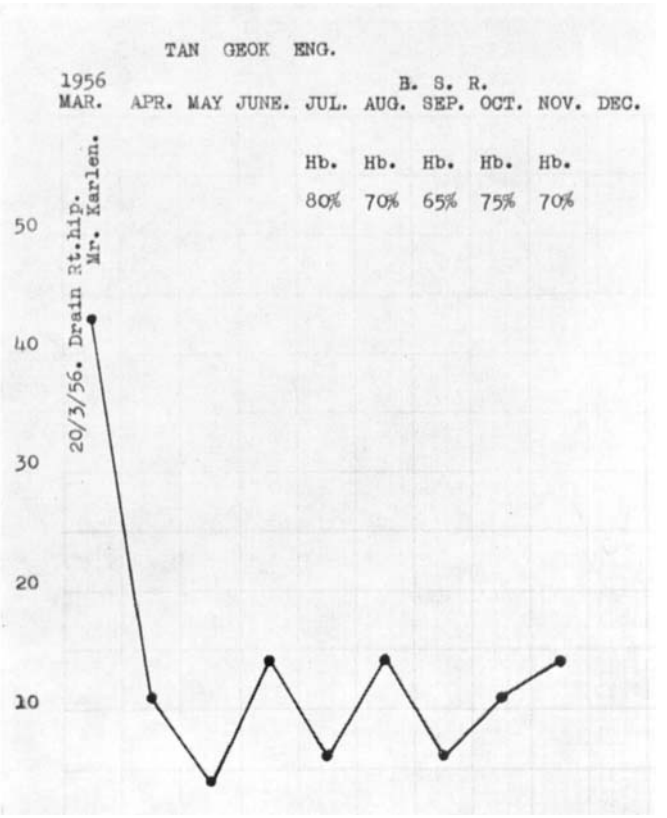

Fig. 3. BSR-chart in Case 2. Good reaction to treatment. The same applies to Case 1 and 3.

collum and caput were bigger than on the left side (Coxa magna), but no subluxation or destruction (Fig. 2).

Case 2. Female. 4 years of age.

Had a fall 6 months ago. Limping on the right side for 5–6 months.

12.3.1956. General condition good. Walks with a limp. The right hip is in flexion—abduction—external rotation contracture. Some muscle-wasting.

BSR. 43 mm. (Fig. 3).

Chest X-Ray: Primary complex.

X-Ray of the right hipjoint: Cystic area in medial lower part of collum with sequestrum. (Fig. 4).

20.3.1956. Curettage + Sequestrectomy through lateral approach.

Biopsy: Positive for tuberculosis.

Fig. 4. *Fig. 5.*

Fig. 4. Cystic area in the medial lower part of collum of the right femur with big sequestrum.

Fig. 5. Cystic disappeared. Head and neck enlarged and in valgus position (Coxa valga magna). Time of observation 3 years.

Postoperatively treated with streptomycin, PAS and INH for 6 months and fixation for 5 months (3 months in hip spica and 2 months on abduction frame).

Last seen three years after operation. No complaints. Fit and well. Excellent range of movements. The X-Ray of the right hipjoint showed that the cyst had practically disappeared. Head and neck enlarged and in valgus position (Coxa valga magna) but no subluxation or destruction. (Fig. 5).

Case 3. Male. 4 years of age.

Swelling and pain in the right ankle 6 months ago. Subsided after 10 days and the boy was gradually able to walk again. Swelling recurred off and on, more marked 10 days before admission. No fever or cough.

30.4.1957. General condition good. Marked swelling of the right ankle, fluctuant in the upper part.

Blood: B.S.R. 47 mm., Hb. 82 %, T.R. 4.03, T.W. 10.100.

Urine: Alb. +, P.C. 2–3.

Chest X-Ray: Primary complex.

X-Ray of the right tibia: Big cystic area in the medial half of the lower metaphysis juxta-epiphyseal. Fibula thinned through pressure (Fig. 6).

3.5.1957. *Curettage* through posterior approach.

Biopsy: Positive for tuberculosis. Postoperatively treated with streptomycin, PAS and INH for 6 months combined with fixation (plaster of paris 7 months and leather garter 6 months).

Fig. 6. *Fig. 7.*

Fig. 6. Big cystic area in the medial half of lower end of right tibia, justa-epiphyseal. Fibula thinned through pressure.

Fig. 7. Cyst disappeared. Time of observation 1¾ years.

Last seen 1¾ years after operation. No complaints. Fit and well. Walking well without limp. Hardly any swelling. Full movements. The X-Ray of the lower end of tibia showed complete healing with disappearance of the cyst (Fig. 7).[1]

The remaining three cases are thought to belong to the type of cystic lesions called multiple pseudo-cystic tuberculosis of bones. This name was suggested by *Komins* (1952) to avoid confusion of certain sarcoid lesions of bone. It applies to characteristic multiple cystlike bone lesions seen in tuberculosis of children; it differentiates these lesions from those due to sarcoidosis which *Jungling* (1920) had grouped under the term Osteitis multiplex tuberculosa cystica.

Multiple pseudo-cystic tuberculosis of bone is uncommon. *Young & Clinton-Thomas* (1955) collected 29 cases from the literature and added one of their own. *Murray* (1954) has pointed out the following criteria

[1] Since this article was written a further 3 cases of single cysts have been seen:
1 in the femoral neck
1 in lower end of femur and
1 in upper end of tibia.
The follow-up time is, however, too short, hence they have not been included.

Fig. 8.

Fig. 9.

Fig. 8. Cystic lesion with expansion of the 4th left metacarpal.
Fig. 9. Cystic lesion in the upper part of right ulna.

as typical: the predilection for childhood; the tendency for the lesions to affect the long bones symmetrically and simultaneously and to progress and regress in harmony; the rarity of joint involvement and the favorable prognosis if it did occur; the lack of early bone reaction with later cortical expansion and fusiform enlargement; and the frequent occurrence of sinuses without sequestrum formation. *Alexander* (1954) has described one case in a 21 year old negro, and pointed out that in contrast to children the involvement was of the axial skeleton and not in the long bones. In his case the lesions were found in multiple places in the spine and ribs, further in the skull, scapula and ileum.

In the cases to be described there has been not only involvement of the long and the short bones, but also of the spine, the mastoid, the malar bone, the scapula, the ribs, and the skin.

Case 4. Male. 3 years of age.

"Eczema" for 1½ years. Discharging sinus in right elbow for 2 months. Low grade fever for 2 weeks.

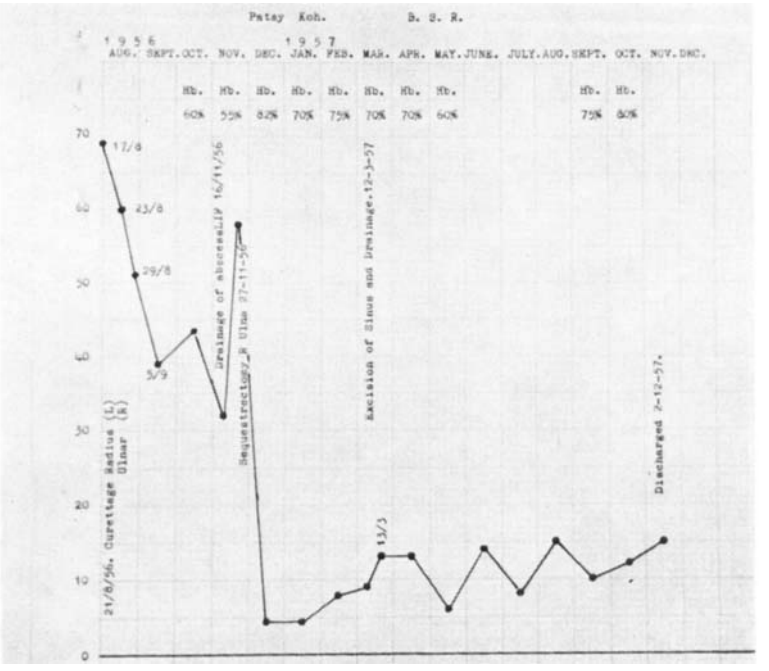

Fig. 10.
BSR—and operation—chart in Case 5.

7.9.1957. General condition good. Multiple patches of "eczema" all over the body. Discharging sinus at the posterior aspect of the right elbow. Free movement in the joint. Discharging ears.

BSR. 31 mm.

Skeletal X-Ray:

1. Cystic lesion with expansion of the fourth left metacarpal. (Fig. 8).
2. Cystic lesion in upper end of the right ulna. (Fig. 9).
3. Large cystic defect in the right mastoid.
4. Destruction of the posterior half of the right 7th rib.

25.9.1957. *Curettage* of the right ulna.

Biopsy: Positive for tuberculosis.

Biopsy (skin): Positive for tuberculosis.

10.11.1957. Transferred to the E.N.T. Department for mastoidectomy. Died a few hours after operation probably in pontine bleeding. Autopsy refused.

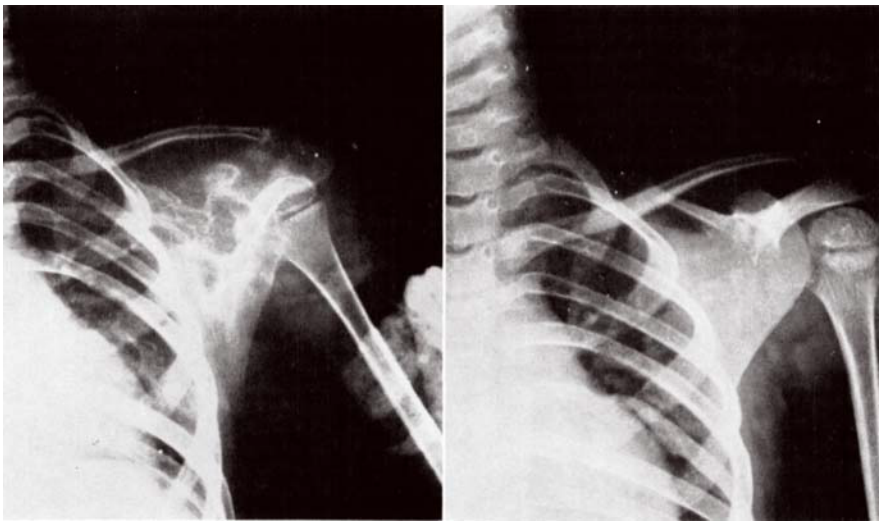

Fig. 11 a. *Fig. 11 b.*

Fig. 11 a. Destruction of upper part of left scapula and of posterior part of 6th left rib.

Fig. 11 b. No bony lesion in left scapula. The 6th left rib regenerated.

Case 5. Female. 4 years of age.

At age of 1½ years there was small swelling on left lower malar bone, growing slowly. Admitted to hospital. Gland taken out. Later treated by general practitioner—"a small piece of bone" is said to have been taken out. 1½ years ago pus started to discharge through a sinus in outer corner of left eye. No fever. Swelling of both forearms for 1 month. Cough for 2–3 years.

9.8.1956. General condition fair. Apathetic. Left malar region swollen, not tender. Discharging sinus at outer corner of lower left eyelid. Left forearm swollen on the upper half on the radial side. The swelling is diffuse, hard, not tender. Fluctuant swelling over posterior aspect of upper part of right forearm. Big fluctuant swelling in left fossa iliaca.

Blood: BSR 69 mm., Hb. 54 %, T.R. 2.6, T.W. 6.300 (Fig. 10).

Chest X-Ray: No pulmonary lesion.

Skeletal X-Ray:

1. Erosion of D.11–12 with destruction of disc.
2. Posterior part of the left 6th rib destroyed.
3. Destruction of upper part of left scapula (Fig. 11 a).

Fig. 12 a. *Fig. 12 b.*

Fig. 12 a. General enlargement of left radius with cystic destruction in midpart plus sclerosis in the distal part. Cystic destruction in upper end of right ulna and in olecranon.

Fig. 12 b. Slight angulation of left radius in the midpart. No deformity of the upper part of right ulna. Time of observation 2 years.

4. General enlargement of left radius with cystic destruction in midpart + sclerosis in distal part (Fig. 12 a).
5. Cystic destruction in upper part of right ulna + olecranon. (Fig. 12 a).

21.8.1956. *Curettage* of left radius and right ulna.

16.11.1956. Drainage of psoas abscess on left side.

27.11.1956. Sequestrectomy of right ulna.

12.3.1957. Excision of sinus + drainage of left scapula.

Biopsy: Positive for tuberculosis from right ulna, left radius, psoas abscess and left scapula.

Treated with streptomycin, PAS and INH for six months and as "umbrella" on further operations. Short periods of fixation in plaster of paris after the operations.

Last seen two years after last operation. General condition good. No sinuses. Full movement in both elbows and shoulders. No swelling. Spine quite mobile without tenderness.

B.S.R. 15 mm. The X-Ray of the skeletal system showed:

Scapula and facial bones: No bony lesion detected (Fig. 11 b).

Regeneration of the 6th left rib.

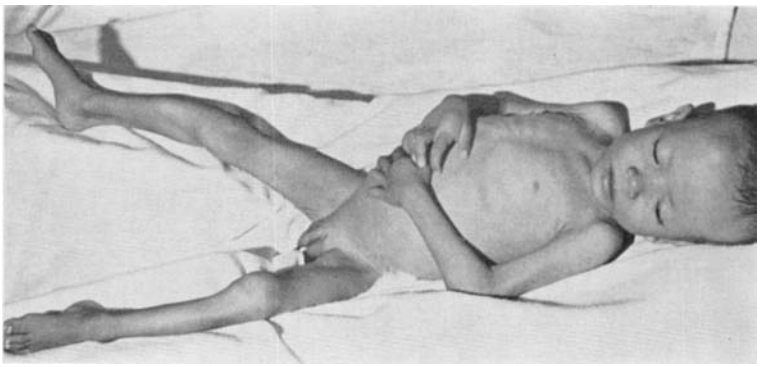

Fig. 13.
Case 6 on admission. Thin and emaciated. Note the swelling in the right side of the forehead, on both hands and on upper part of right tibia.

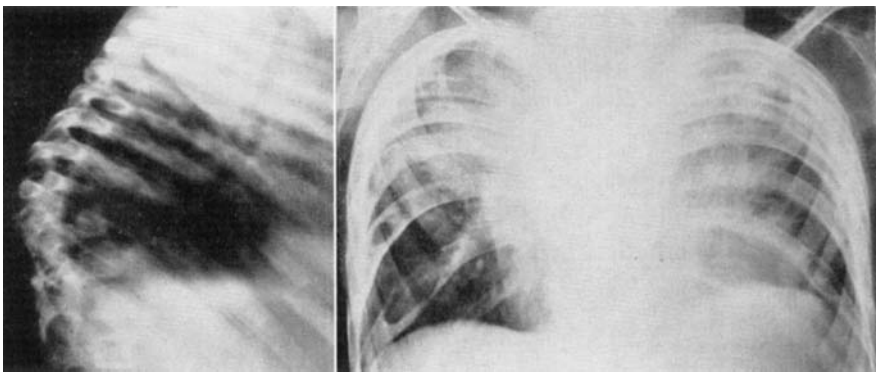

Fig. 14. *Fig. 15.*

Fig. 14. Pott's disease with destruction of D7–D9.
Fig. 15. Marked cystic lesions in 6th and 7th right and 6 left rib.

Moderate collapse of body D.11 with some narrowing of disc below it. Complete recalcification.

Slight angulation in the midpart of left radius.

No bony lesion in right ulna (Fig. 12 b).

Case 6. Male. 4 years of age.

"Not well" for two years. One week before admission there was fever, sweating and coughing. Poor appetite.

25.5.1955. Very thin and emaciated. Prominent swelling on the right frontal region of skull (Fig. 13).

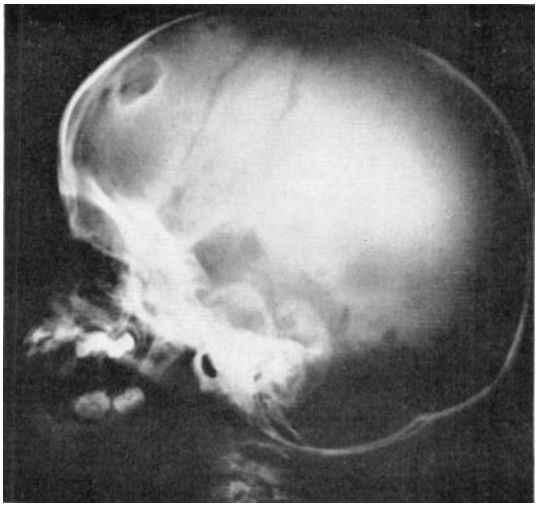

Fig. 16.
Well demarcated cystic lesion in the frontal region.

Extremities: Swelling at proximal end of right ulna and fusiform swellings of the 2nd and 4th right metacarpal. Same type of swelling over proximal end of left ulna and left second metacarpal. Some bowing forwards of right tibia with a localised swelling in the middle third.

Blood: B.S.R. 62 mm., Hb. 49 %, T.R. 2.4, T.W. 7.100.

Calcium 11 mgm%, phosphates 5.3 mgm%, K.T. negative.

Urine: Alb. +, P.C. 3–4, R.B.C. 2–3.

Mantoux pos. 1/100.

Chest X-Ray: Primary tuberculosis.

Skeletal X-Ray:

1. Kyphosis in mid-dorsal spine due to partial destruction of D.7 and D.9 and total destruction of D.8 (Fig. 14).
2. Ribs: Cystic lesions with expansion in the 6th and 7th rib on the right side and 6th rib on the left side (Fig. 15).
3. Skull: Well demarcated cystic lesion in the frontal region (Fig.16).
4. Forearms: Well demarcated cystic lesion in left olecranon, multiple cystic lesions in upper part of right forearm and olecranon surrounded by sclerosis. Small cystic lesions also seen in the lower lateral part of right humerus (Fig. 17).
5. Hands: Partial destruction combined with sclerosis of both 2nd metacarpalia and 4th right metacarpal bone.
6. Right tibia and fibula: Big cystic lesions in upper and middle part of tibia with some expansion of the bone (Fig. 18). The mid-

Fig. 17.

Fig. 18.

Fig. 17. The lesions in both ulnae, lower end of right humerus.
Fig. 18. Cystic lesions in both fibulae and right tibia.

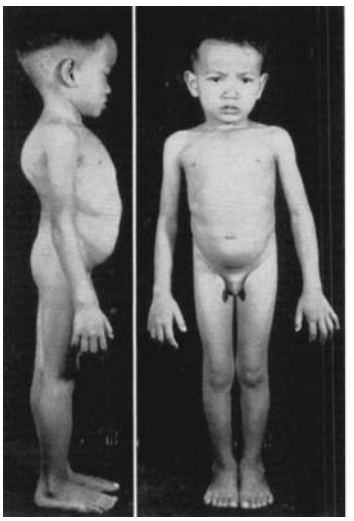

Fig. 19.
Patient's condition when last seen 3 years and 4 months after operation.

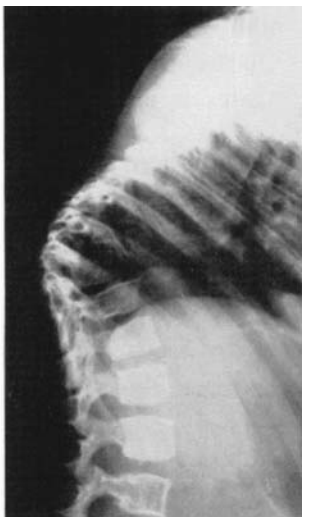

Fig. 20 a.

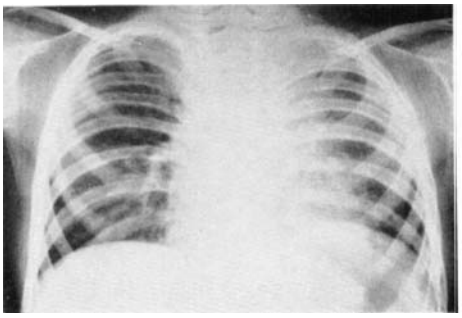

Fig. 20 b.

Fig. 20 c.

Fig. 20 d.

Fig. 20 e.

Figs. 21 a–e.
Pott's disease healed with good recalcification. No lesions detected in ribs, skull, forearms of tibia and fibulae.

part of right fibula is expanded by multiple cystic lesions surrounded by sclerosis.

7. Left fibula: Expansion of the midpart of the bone. In the centre is a cystic lesion surrounded by sclerosis.

3.6.1955. *Curettage* of right ulna.

Biopsy: positive for tuberculosis.

Postoperatively treated with streptomycin, PAS and INH for 6 months plus immobilisation in box and on frame for 10 months.

Last seen 3 years and 4 months after operation. Fit and well (Fig. 19). Full movements in elbows, hips and knees. No sinuses. The X-Ray examination of skeleton showed a well healed Pott's disease D.7—8–9. The rest of the skeleton was normal (Figs. 20 a–e).

SUMMARY

Two types of cystic appearance of bone tuberculosis are described, in all 6 cases. Three of them are single metaphyseal or metaphyseal-epiphyseal cysts well defined and clinically impossible to differentiate from other cystic lesions. The remaining 3 are thought to belong to the type of tuberculous skeletal manifestation called "Multiple Pseudo-Cystic Tuberculosis of Bone".

RESUME

Deux types d'apparence kystique de tuberculose osseuse sont décrits, en tout 6 cas. Trois d'entre eux sont de simples kystes métaphysaires ou métaphysaires-épiphysaires bien définis et qu'il est impossible cliniquement de différencier d'autres lésions kystiques. On pense que les trois autres appartiennent au type de la manifestation squelettique appelée « tuberculose osseuse pseudokystique multiple ».

ZUSAMMENFASSUNG

Zwei Typen der zystischen Form der Knochentuberkulose, insgesamt 6 Fälle, werden beschrieben. Drei davon sind einzelne, gut abgegrenzte metaphysäre oder meta-epiphysäre Zysten, die klinisch unmöglich von anderen zystischen Schädigungen unterschieden werden können. Die übrigen drei scheinen zu jener Type von Skelettuberkulose zu gehören, die als „Multiple pseudozystische Knochentuberkulose" bezeichnet wird.

REFERENCES

1. *Alexander, G. H. & Mansuy, M. M.:* Diseminated Bone Tuberculosis (So-Called Multiple Cystic Tuberculosis). Radiology, *55*: 839, 1950.
2. *Cholmeley, J. A.:* The Distribution and Treatment of Extra-Articular Foci In Tuberculous Arthritis of the Hip-Joint. British Journal of Surgery, *27*: 224, 1939.

3. *Clinton-Thomas, C. L. & Young, W. B.:* Multiple Pseudo-Cystic Tuberculosis of Bone. Journal of Bone & Joint Surgery. *37B*: 624, 1955.
4. *Girdlestone, G. R. & Sommerville, E. W.:* Tuberculosis of Bone and Joint. Oxford Medical Publication, London. 1952.
5. *Jungling, O.:* Ostitis Tuberculosa Multiplex Cystica. Fortschritte auf dem Gebiete der Roentgenstrahlen. 27: 375, 1919–21.
6. *Komins, C.:* Multiple Cystic Tuberculosis: a Review and a Revised Nomenclature. British Journal of Radiology. 25: 1, 1952.
7. *Murray, R. O.:* Observations on Cystic Tuberculosis of Bone, with a Report on Two Cases. Proceedings of the Royal Society of Medicine (section of Orthopaedics). 47, 133, 1954.
8. *Ratliff, H. C.:* Tuberculosis of the Femoral Neck in Childhood. The Journal of Bone and Joint Surgery. *39A*: 1365, 1957.
9. *Wallgren, A.:* The Time-Table of Tuberculosis. Tubercle, 29: 245, 1948.