PIGMENTED VILLONODULAR SYNOVITIS OF THE WRIST WITH PENETRATION INTO BONE

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Invasion and erosion of bone in nodular synovitis (histiocytic xanthomatous granuloma, xanthoma, giant-cell tumour, benign synovioma) are found fairly frequently in circumscribed lesions of the tendon sheaths of hands and feet. Fletcher and Horn (1951) in particular called attention to this. Bone involvement in diffuse forms, namely "pigmented villonodular synovitis, bursitis and tenosynovitis" (Jaffe, Lichtenstein and Sutro 1941) are much less common.

The clinical and pathological features of this condition are well known. Mandl (1928) reported a similar condition under the name of "haemorrhagic chronic villous synovitis": at a second operation abundant blood pigment and extensive xanthomatous areas were found. One of us (F. S.) reported another patient, also operated upon by Mandl, in 1937, under the same denomination (Schajowicz 1937).

In 1963 Schajowicz and Slullitel presented before the Argentine Orthopaedic Society a case of pigmented villonodular synovitis affecting the shoulder with invasion of the humerus and scapula. Reviewing the literature and material filed with the Latin-American Register of Bone Pathology, they found fifty-two cases with invasion of one or more bones adjacent to the affected joint, including twelve cases studied in the laboratory of the Register. The hip, ankle and knee were the joints most commonly affected. On only two occasions did the condition affect the wrist (Lewis 1955) and carpus (Smith and Pugh 1962). Following our paper, Chung and Janes (1965) reported four cases of affection of the hip. Including the case mentioned in this report we have observed six new cases of bone invasion among twenty-seven cases of pigmented villonodular synovitis studied in our laboratory. For this reason we do not believe that this complication of pigmented villonodular synovitis is so rare as appears from the few cases published.

The case reported here is of special interest because of the severity of the invasion of the lower end of the radius and because of the size of the lesion. These features raised the possibility that the lesion might be a giant-cell tumour of bone with extensive invasion of the neighbouring soft tissues and bones and perhaps with malignant change.

CASE REPORT

A sixty-one-year-old man attended the Orthopaedic and Traumatologic Service of the Hospital Israelita of Buenos Aires in August 1964. There was no significant history of previous illness except for brucellosis which lasted for five months when he was thirty-eight years old. Twenty years previously a "tumour" had appeared on the ulnar side of the left wrist. It caused no pain and the patient thought nothing of it. It continued to grow slowly and painlessly until the patient came for treatment.

The wrist was occupied by a swelling the size of a large grapefruit (Fig. 1). The swelling was multilobulated and of firm consistency. The radial pulse could be felt normally on the palmar aspect of the wrist.

There was perfect function of all long tendons and of all intrinsic muscles of the hand. There was a line of demarcation between the forearm and the tumour on one side and between the tumour and the hand on the other. The tumorous infiltration ran all around that segment of the wrist.

FIG. 1
The wrist and hand, showing the large tumour.

FIG. 2
Antero-posterior and lateral radiographs showing the trabeculated osteolytic lesion of the lower end of the radius and of the proximal carpal bones.

FIG. 3

FIG. 4

FIG. 5

FIG. 6

Photomicrographs showing different histological patterns of the process. Figure 3—Area of villous aspect with abundant dark coloured blood pigment. ($\times 80$.) Figure 4—Solid area showing histiocytic proliferation and abundant xanthomatous cells. ($\times 80$.) Figure 5—At higher magnification, typical xanthomatous and some multinucleated giant cells can be observed. ($\times 320$.) Figure 6—Xanthomatous tissue infiltrating the marrow spaces. Note the trabeculae of cancellous bone. ($\times 80$.)

Radiographs showed an osteolytic lesion of the distal end of the radius with septa and without any apparent break in the cortex. The ulna was not affected but the bones of the ulnar side of the carpus showed small osteolytic lesions (Fig. 2).

Biopsy—Initially, a needle biopsy was done. Examination of this specimen showed

FIG. 7

The material removed during the first operation, showing the dark colour of the irregularly shaped fragments, partly solid or villonodular. (Scale in centimetres.)

FIG. 8

The villonodular aspect of some of the fragments removed during the second operation.

granulomatous tissue, rich in reticulo-histiocytic elements and deeply pigmented. The provisional diagnosis of pigmented villonodular synovitis was made, but open biopsy was advised in order to establish the diagnosis. Macroscopically, the tissue consisted of several brownish irregular fragments with some frankly yellowish areas. There were also some fragments of cortical bone, thinned down to a fine membrane, and there were pieces of cancellous bone partly infiltrated by a yellowish-brown tissue similar to the soft tissues.

Histological examination showed compact areas of granuloma-like tissue. These showed intense reticulo-histiocytic proliferation, many cells being filled with abundant haemosiderin or lipids and many being in giant-cell evolution (Figs. 3 to 6). A frankly villous or hypertrophic hyperaemic synovium was observed, showing in places a similar histological appearance. Other areas showed fibrosis and evident lympho-plasmocytic infiltration.

The bone fragments showed penetration of the pigmented granulomatous and xanthomatous tissue within the marrow spaces, and in some places the cortex was very thin and newly formed. A diagnosis of pigmented villonodular synovitis with extensive intraosseous penetration was made.

FIG. 9

Radiograph of the hip of a 46-year-old woman showing an extensive multiloculated osteolytic lesion of the trochanteric region extending into the femoral neck. A similar lesion with sclerotic borders is seen in the acetabulum.

Operation—Two operations were required. The first operation was done in October 1964. A tourniquet was applied and a longitudinal incision was made on the ulnar aspect of the dorsum of the lowest third of the forearm and of the wrist and hand. A large mass was found and an attempt was made to dissect this out. Although the extensor tendons were unaffected and could be freed the dissection proved very difficult.

Another mass, the size of a duck's egg, was found on the radial side and over the palmar surface of the hand and wrist. This was removed through a second incision on the dorsal and radial aspect of the forearm and wrist. The tourniquet was then released and haemostasis was secured.

The ulnar aspect of the tumour was then approached again, but it was found impossible to remove the tumour completely because of its extension into the palm. This part was left for removal at a second operation. Haemostasis was secured and the wounds were closed. The course after operation was uneventful.

The appearance of the tumour is shown in Figure 7. Histological study confirmed the initial diagnosis.

Second operation—Three weeks later the second operation was done. A tourniquet was applied and an incision was made on the anterior aspect of the forearm and wrist. The ulnar nerve was dissected free from the tumour mass. The mass was then opened and the flexor tendons, included in it but not affected, were freed. The median nerve was dissected free and the incision was extended distally. Much of the mass was excised but some fragments remained adherent (Fig. 8). The lower end of the radius was curetted out and the cavity was filled with chips of cancellous bone. The tourniquet was then released and haemostasis secured. A plaster was applied. The patient was given antibiotics.

The course after operation was uneventful, and two months later the patient had full movements of the wrist and fingers. Sixteen months after operation he reported that he was doing his normal work on the land without pain or any other disability.

DISCUSSION

Bone involvement in pigmented villonodular synovitis appears to occur more often than is to be inferred from the literature (Smith and Pugh 1962, Chung and Janes 1965). In our experience the joints most often affected are the hip (six cases) and the ankle (five cases).

There was involvement of bone in only three cases of affection of the knee. The contrast with the high incidence of villonodular synovitis in the knee is possibly because increased intra-articular pressure caused by profuse synovial hypertrophy is more manifest in joints with a narrow lumen, in which penetration of the bone by proliferating villi is easier, especially in the area of the chondro-osseous junction. Invasion of bone as severe as that shown in the present case has only been seen by us in a case of affection of the hip (Fig. 9). One other similar case was described by Smith and Pugh (1962). At first we thought we were dealing with a bone tumour, but the long period of evolution and the good general health of the patient made us discard the idea of a malignant process. The possibility still existed of sarcomatous change in a benign giant-cell tumour.

Examination of the material removed during the two operations confirmed the diagnosis of villonodular synovitis, probably originating in the tendon sheaths of the wrist rather than in the synovial membrane of the radiocarpal joint, although with so advanced a lesion the latter origin could not be discounted. Wide excision of all the affected tissue, with curettage of the cystic lesions of the carpus and radius and filling of the cavities with bone chips, gave an excellent functional result.

SUMMARY

A case of unusually extensive pigmented villonodular synovitis of the wrist with involvement of bone, particularly of the distal end of the radius, is reported. The clinical and radiographic evidence suggested a diagnosis of primary bone tumour, possibly a giant-cell tumour with sarcomatous transformation.

REFERENCES

- CHUNG, S. M. K., and JAMES, J. M. (1965): Diffuse Pigmented Villonodular Synovitis of the Hip Joint. Review of the Literature and Report of Four Cases. *Journal of Bone and Joint Surgery*, **47-A**, 293.
- FLETCHER, A. G., and HORN, R. C., Jun. (1951): Giant Cell Tumors of Tendon Sheath Origin. A Consideration of Bone Involvement and Report of Two Cases with Extensive Bone Destruction. *Annals of Surgery*, **133**, 374.
- JAFFE, H. L., LICHTENSTEIN, L., and SUTRO, C. J. (1941): Pigmented Villonodular Synovitis, Bursitis and Tenosynovitis. *Archives of Pathology*, **31**, 731.
- LEWIS, R. W. (1947): Roentgen Diagnosis of Pigmented Villonodular Synovitis and Synovial Sarcoma of the Knee Joint. *Radiology*, **49**, 26.
- LEWIS, R. W. (1955): *The Joints of the Extremities. A Radiographic Study*. Springfield, Illinois: Charles C. Thomas.
- MANDL, F. (1928): Chronische Arthritis villosa hämorrhagica des Kniegelenkes. *Zentralblatt für Chirurgie*, **55**, 597.
- SCHAJOWICZ, F. (1937): Über chronische Arthritis villosa hämorrhagica des Kniegelenkes. *Zentralblatt für Chirurgie*, **64**, 1649.
- SCHAJOWICZ, F., and SLULLITEL, I. (1963): Sinovitis vellosa-nodular pigmentada (xantomatosis articular) del hombro con invasión ósea. *Boletín y trabajos de la Sociedad argentina de ortopedia y traumatología*, **18**, 118.
- SMITH, J. H., and PUGH, D. G. (1962): Roentgenographic Aspects of Articular Pigmented Villonodular Synovitis. *American Journal of Roentgenology, Radium Therapy and Nuclear Medicine*, **87**, 1146.