SQUAMOUS-CELL EPITHELIOMA ORIGINATING IN CHRONIC OSTEOMYELITIC CAVITIES

REPORT OF TWO CASES

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Squamous-cell epithelioma originating in chronic osteomyelitic cavities is of special interest, first because of its rarity, and secondly because of the association of malignancy with a chronic inflammatory process.

REPORT OF TWO CASES

CASE No. 1 (File No. 177640): F. W., a white male, aged sixty-nine years, was admitted to the Buffalo City Hospital on June 2, 1928. His chief complaint was of a discharging sinus in the upper third of the right tibia, and continuous pain in the right leg.

At the age of ten years, or fifty-nine years before, the patient had sustained a severe contusion of the right foot. High fever, pain, and swelling of the leg ensued in a few days, and a large abscess in the anterior upper third of the tibia was incised and drained. A persistently discharging sinus, leading from the bone, resulted. A slight amount of pain in the leg had been present almost continuously, but it was not sufficiently severe to incapacitate the patient for work. Occasionally small pieces of bone would be expelled through the sinus. In 1923 the sinus was explored and pieces of bone were removed. In February 1928 there was a very profuse hemorrhage from the sinus, followed by three less severe hemorrhages within the course of a few months. Hospital care was sought on account of the general debilitated condition, and fear of future hemorrhages.

Although the patient was not acutely ill on admission, yet he had a cachectic appearance. The skin and mucous membrane were very pale. The chest was emphysematous, and the supraclavicular spaces were depressed. The chest expansion was particularly limited on the right side. Resonance was impaired over the left lung from the apex to the level of the third rib, both anteriorly and posteriorly, and over the whole right lung, posteriorly. There was bronchial breathing over the right and left lungs anteriorly, over the right lung posteriorly, and over the apices. The vesicular breathing was suppressed at the bases. No râles were heard. The systolic blood pressure was 102 and the diastolic 64. The heart sounds were distant and feeble. All the peripheral arteries showed marked arteriosclerosis. On the inner flat surface of the upper third of the right tibia was a deep sinus exuding a very foul-smelling, purulent discharge. The tissues around the sinus were edematous. Both legs showed varicose veins.

The laboratory findings were as follows: hemoglobin, 40 per cent; red blood cells, 2,490,000; white blood cells, 11,110; polymorphonuclear leukocytes, 67 per cent; large mononuclear cells, 3 per cent; small lymphocytes, 22 per cent; eosinophils, 6 per cent; transitional cells, 1 per cent; myelocytes, 1 per cent; color index, 0.83. The Wassermann reaction in the blood serum was negative. The blood urea was
23.1 mgm. and the blood sugar was 133 mgm. The urine was acid and the specific gravity was 1.018. There was no albumen nor sugar present. There were a few pus cells, red blood cells, and hyaline casts.

Radiographic films showed changes characteristic of chronic osteomyelitis of the upper half of the right tibia. There were several areas of rarefaction scattered throughout the affected part of the bone.

The preoperative diagnoses were: chronic osteomyelitis of the right tibia; severe secondary anemia; chronic pulmonary tuberculosis and peripheral arteriosclerosis.

On August 24, 1928, the patient received a blood transfusion and, on the following day, a disarticulation of the leg at the knee was done. Complete anesthesia was obtained by blocking the sciatic and the anterior crural nerves with 0.5 per cent novocaine solution. The postoperative convalescence was without incident and the amputation stump healed slowly. On December 13, 1928, death occurred from lobar pneumonia.

Pathologic studies were made by Dr. William F. Jacobs. The gross specimen consisted of the amputated leg. The mouth of the sinus was located on the inner surface of the upper third of the tibia, about 5 cm. from the articulating surface. It measured $3 \times 2$ cm. A sagittal section (Fig. 1) of the tibia, through its upper third, revealed a cavity $14 \times 2$ cm. lined with gray, necrotic, nodular granulation tissue, which was bathed by a purulent exudate. This was very foul-smelling and contained a few loose pieces of bone. The cavity extended above to the articulating surface of the tibia; below, it tapered into a narrow sinus tract.

Microscopic sections (Fig. 2) made from the granulation tissue in the sinus tract and in the bone cavity revealed the presence of a squamous-cell epithelioma invading bone tissue as well as normal skin. Pearl formation was frequently noted, although most of the cells showed a very slight degree of differentiation.

In addition to the above findings, the autopsy revealed chronic bronchitis, chronic fibrous pleurisy, pulmonary emphysema, marked pneumoconiosis, pulmonary fibrosis, healed pulmonary tuberculosis, general arteriosclerosis, benign hypertrophy of the prostate, hypertrophy of the urinary bladder with trabeculations, ulcerative enterocolitis, and lobar pneumonia with fibropurulent pleurisy. There was no amyloid degeneration present in any of the organs, nor was there any evidence of metastasis.

Case No. 2. (File No. 179154): E. H. K., a white male, aged fifty years, was admitted to the Buffalo City Hospital on June 30, 1928. His chief complaint on admission was pain in the right hip joint. Twenty-five years before, he fell while skiing and struck the ground with the right hip. A few days later the hip was incised and pus was evacuated. A discharging sinus leading from the bone resulted. However, at intervals of a few months, the sinus would heal completely. There was considerable pain in the hip at all times, although it was not severe enough to incapacitate the patient until December 1927, when he had to quit work.

The patient was emaciated and lay in bed with the right hip flexed. The temperature on admission was 102.6 degrees and the pulse rate 110 per minute. The systolic blood pressure was 182 and the diastolic 85. All the teeth were missing and there was a moderate degree of pharyngitis. The peripheral arteries showed arteriosclerosis. The heart, lungs, and abdomen were normal. Rectal examination revealed normal findings. The right thigh was held flexed and the motions at the hip were attended with pain. Over the region of the great trochanter there was a sinus, $2^{1/2}$ cm. long, 1 cm. wide, and $1^{1/2}$ cm. deep, exuding foul-smelling, purulent material. The soft tissues around the sinus were red and indurated.

The laboratory findings were as follows: hemoglobin, 80 per cent; leukocytes, 6950; red blood cells, 4,200,000. The differential count was normal. The Wassermann reaction in the blood serum was negative. The urine was acid in reac-
FIG. 1. CASE NO. 1: SAGITTAL SECTION THROUGH THE TIBIA, SHOWING THE OSSEOMYE-
LITIC CAVITY AND THE NODULAR APPEARANCE OF THE TUMOR MASS

FIG. 2. CASE NO. 1: SQUAMOUS-CELL CARCINOMA IN CHRONIC OSSEOMYELOITIC SINUS
TRACT. X 150
tion; the specific gravity was 1.022 and there was no sugar nor albumen. The sediment showed an occasional hyaline cast. Culture made from the exudate in the sinus showed hemolytic Staphylococcus aureus and Gram negative bacilli.

Radiographic studies made by Dr. Orr showed an extensive, destructive process involving the head and neck of the femur, the greater trochanter, and the intertrochanteric space. There was very little bony proliferation. The pelvis, as well as the remainder of the femur, showed nothing abnormal.

On August 8, 1928, the sinus was curetted. The material consisted of very friable granulation tissue mixed with blood. Microscopic examination showed marked atypical proliferation of the stratified epidermis and chronic granulation tissue containing numerous foreign-body giant cells. Although in many instances the epidermis was invaginated, yet the proliferation was so regular that it was considered an inflammatory process rather than a malignant one. Staining of the sections failed to show the presence of tubercle bacilli.

On Oct. 15, 1928, the patient had a sudden severe pain in the right hip while moving in bed. There was both clinical and x-ray evidence of a fracture in the hip. Due to the severe pain, which did not respond to the usual treatment of fractures in this location, a disarticulation at the hip was advised. On Oct. 18, the right hip was disarticulated. The patient did not rally after the operation and died of shock on the following day.

Pathologic examination of the specimen (Fig. 3) showed an epithelioma infiltrating the neck of the femur and a pathologic fracture in that area. An autopsy was not performed. Histologic examination of the specimen showed a low grade squamous-cell epithelioma with pearl formation (Fig. 4).

**DISCUSSION**

It is interesting to note that in these two cases no evidence of metastasis was found, despite the fact that in Case No. 1 the tumor cells showed very slight degree of differentiation. White and Weidman (1) have reported eight cases of malignant degeneration of cutaneous ulcers. They divided these cases into three groups upon the basis of the histologic appearance of the cells and the extent to which the cells invade the subcutaneous tissue. None of their cases showed metastasis. For the above reasons they have chosen to call this type "pseudo-epitheliomatous hyperplasia." Knox (2) also reports epitheliomata in varicose ulcers. She remarks that these tumors run a slower course than other squamous cell epitheliomata and believes that they may lead eventually to metastasis.

Another interesting feature in this series was the presence of persistent draining sinuses with foul-smelling discharge. Milgram (3) has called attention to epithelial lining of osteomyelitic cavities as a cause of persistent drainage. None of his cases, however, showed epitheliomatous changes in the sinus tract.

Brunschwig (4) reports three cases of chronic osteomyelitis with draining sinuses of long standing. The sinuses were lined with epithelium and, in some instances, "pearl" formation was observed. In his discussion he remarks that skin carcinoma not
Fig. 3. Case No. 2: Saggital Section through the Head and Neck of the Femur
The gray areas represent tumor masses in the bone tissue

Fig. 4. Case No. 2: Squamous-cell Carcinoma in Chronic Osteomyelitis of Femur.
× 80

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infrequently develops on chronic discharging sinuses. He believes that the constant inflammatory process in these cavities affords the stimulus to malignant degeneration of the epithelial cells. Kaufmann (5) states that the epithelial lining of bone fistulas may undergo malignant changes. He reports a case in a patient fifty-four years of age, with malignant degeneration in an osteomyelitic cavity of thirty-four years standing; there was a sequestrum corresponding to the location of the sinus tract. Kaufmann concludes that the sequestrum was the exciting cause of the malignant degeneration.

Conclusions

1. The two cases reported definitely show the presence of squamous-cell epithelioma in the lining of osteomyelitic cavities. This type of epithelioma apparently does not give rise to metastasis.

2. It would appear that the epithelial lining of osteomyelitic cavities is the cause of persistent drainage. Brunschwig's theory that the chronic inflammatory process constitutes a stimulus to malignant degeneration of epithelial cells is also accepted.

3. It is highly desirable, in view of the above findings, that chronic osteomyelitic cavities should be curetted and the tissue carefully studied.

References