LONG SURVIVAL IN SOLITARY PLASMOCYTOMA OF BONE

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Although many examples of solitary plasmocytoma or myeloma of bone have been reported, in few has the diagnosis been clearly substantiated. Follow-up has often been too short because multiple myeloma, a closely related condition, may first manifest itself as a solitary growth identical with solitary plasmocytoma, and on occasion several years may elapse before other tumours appear. Sometimes the diagnosis of a solitary lesion has been made at necropsy, but even allowing for the difficulty in proving this, further tumours might still have appeared had the patient lived. Opinion has consequently hardened against the existence of true solitary plasmocytoma of bone, and prolonged survival after excision of the tumour, with failure to develop into multiple myeloma, seems to be the only reliable confirmatory criterion. The lack of such reports in the literature and their obvious importance in establishing the disease as an entity merits the recording of the following two examples. One is new: the other was reported from this department by Stewart and Taylor in 1932, but the after-history to date adds considerable support to the diagnosis.

Case 1—Man aged thirty-three. In December 1936 this patient, a labourer, slipped, suffering pain behind the right knee. The pain persisted for about three weeks, but no swelling was evident. He went back to work, but in February 1937 he again complained of pain behind the knee. It was aching in character, intermittent and worse on working, but there was still no swelling. The condition was treated as “rheumatism” until June, when he fell from a tar-spraying wagon and fractured his right femur. He was admitted to the Leeds General Infirmary under the care of Mr Broomhead. Radiographic examination showed a transradiant tumour in the middle third of the right femur, with a pathological fracture a little above the centre of the growth (Fig. 1). A biopsy specimen taken in August was reported by the late Professor Stewart to show a “typical plasmocytoma of great cellularity.” The right humerus, radius and ulna, and the skull were radiographed but found to be normal. The tumour was treated by radiotherapy (3,025 r over thirty-seven days). A further radiograph a month later showed that the transradiant expansion of the shaft was more noticeable, but not extreme. The tumour then occupied six inches of the length of the shaft, and opaque trabeculae

Fig. 1

Case 1—Radiograph of mid-shaft of the right femur, showing expansive osteolytic growth with multicystic trabecular pattern.
ran irregularly through the transradiant area. Shortly afterwards, in November 1937, the leg was amputated by disarticulation through the hip.

Pathological examination—In the middle of the shaft of the femur there was a large new growth composed of compact greyish tissue which had been freely ploughed up by old and recent haemorrhages. The tumour had destroyed much of the cortex of the bone throughout the whole of the affected segment of the femur, but did not appear to be invading the adjacent muscles. Some evidence of regeneration of bone was present in places.

Microscopically, sections of the tumour stained with haematoxylin and eosin, and with Unna Pappenheim stain for plasma cells, showed a typical plasmocytoma (Fig. 2). The section contained closely packed well differentiated plasma cells. The cells were notably uniform, with moderate pleomorphism, and occasional immature forms; larger cells with two or three nuclei were evident. Mitotic figures were encountered, but only in small numbers. Parts of the tumour were unduly vascular, with a network of channels formed merely of endothelial lining cells. There were large areas of necrosis and haemorrhage. The tumour was separated from muscle by dense collagenous fibrous tissue in which slender spicules of bone were arranged at intervals along the margin. Inguinal lymph glands were found to be normal.

Subsequent progress—Since the operation the patient has shown no sign of recurrence, and when seen in June 1959, nearly twenty-two years later, he was in good health apart from persistent stomach symptoms. Partial gastrectomy was performed for gastric ulcer in 1955. Plasma protein estimations carried out recently on several occasions varied only between 5 and 6 grammes per cent. The blood picture remained normal.

Case 2—This case was recorded by Stewart and Taylor (their Case 1) in 1932 as a solitary plasmocytoma of the humerus, the diagnosis being supported by an eight-year survival after amputation. The patient, a farmer aged thirty-four, had a diffuse swelling of the upper part of the right arm. A radiograph had shown an endosteal growth at the upper end of the humerus, with expansion and thinning of the bone and a pathological fracture of its uppermost and middle thirds. A forequarter amputation was performed in 1924 and the humerus, which is now in the museum of the Institute of Pathology at Leeds, showed an irregularly lobulated tumour arising in, and largely destroying, the uppermost third of the shaft of the humerus (Fig. 3). The tumour was still contained within a thinned shell of bone except on the medial aspect where it had grown out into the adjacent soft parts. A full account of the histological
structure was given in the original case report, but the sections have been reviewed and the diagnosis of plasmocytoma has been confirmed. The tumour showed more pleomorphism than in Case 1, and there was invasion of muscle.

The original report stated that eight years had elapsed since the operation and that the patient was in perfect health. Jaffe (1958) cited a personal communication from Stewart informing him that this was still true eighteen years after the report. In July 1959 the writer visited the patient and found him perfectly fit apart from some osteoarthritis, and still farming, thirty-five years after the forequarter amputation.

**DISCUSSION**

Plasmocytoma may be classified into three groups: 1) multiple myeloma or myelomatosis; 2) solitary myeloma or plasmocytoma of bone; 3) primary extramedullary myeloma or plasmocytoma. Multiple myeloma is uniformly fatal and usually of short duration, but occasionally there may be a relatively long survival. Kenny and Moloney (1956) reported survival in one case for nine years and in two cases for eleven years from onset of the disease in a series of fifty-seven cases. Other instances of even longer survival are on record. Multiple myeloma not infrequently presents initially as a single tumour, though the existence of a truly solitary form has been in some doubt. In a series of ninety cases of plasma cell myeloma, Carson, Ackerman and Maltby (1955) reported sixty patients in the multiple group, eighteen with an apparently solitary bone lesion, and twelve with primary extramedullary tumours. These authors believed that if followed for a long time the solitary bone lesion will eventually develop into diffuse myeloma. Of the cases in their series, one patient died within three months, seven survived from six to twenty-four months, and seven from two to six and a half years. Only three remained alive—after three years nine months, seven years four months, and nine years ten months. When a single tumour is the first manifestation of multiple myeloma, further tumours tend to appear rapidly, though on occasion there may be a delay of several years. In a patient originally described by Rogers (1930), later follow-up by Yentis (1956) revealed an interval of eight years from time of amputation. Yentis (1956) also reported, in his Case 3, an interval of ten years. No unequivocal case of solitary plasmocytoma of bone could be found by either Snapper, Turner and Moscovitz (1953) in their series of ninety-seven cases or Bayrd and Heck (1947) in eighty-three cases.

Although a number of examples of solitary plasmocytoma of bone have been recorded, and later accepted by other authors, there has been considerable uncertainty in recent years whether the tumour is in fact an entity, and, if so, what criteria should appertain for its
acceptability. The solitary growth is said to present as a large destructive lesion of bone, usually with pathological fracture when a long bone is affected. According to Lumb and Prossor (1948) solitary tumours tend to be larger than any individual mass of the multiple disease, when the tumours are usually small and do not expand the bones. Snapper et al. (1953), in ninety-seven cases of multiple myeloma, found palpable masses in only twenty-four. Raven and Willis suggested in 1949 that the tumour could be accepted as truly solitary only if no further tumours appeared and if careful radiographic examination of the rest of the skeleton remained negative for “two or three years or longer.” Alternatively, they considered that adequate necropsy examination might prove the solitary nature of the growth. These authors accepted only eighteen cases, several of which were based on necropsy findings. As they pointed out, however, there could be no certainty in the latter instances that if the patient had survived other tumours would not have developed. Yentis (1956) emphasised that “the validity of a true solitary tumour would still pivot on meticulous and prolonged follow-up.”

In two of the earlier cases accepted by subsequent authors recent information by Yentis (1956) has shown that they did in fact develop into multiple myeloma. Shaw’s (1923) case was reported by Cutler, Buschke and Cantril (1936) to have survived for nine years, but Yentis (1956) was able to trace the later history and found that the patient had died from multiple myeloma with involvement of the spine and left femur. He also gave the after-history of the other case, in which the course proved to have been remarkably protracted. The patient had his right leg amputated in 1929 for a tumour described by Rogers (1930) as a solitary plasma-celled myeloma. All was well until 1937 when a further tumour was discovered in the left ischium, and a biopsy specimen showed a myeloma. In 1951 a destructive lesion was found in the left mandible and again a biopsy specimen revealed a myeloma. More masses appeared in the left temporal region and in the palate, and the patient died in 1953.

In contrast, additional follow-up information in two cases which similarly had been accepted lends support to the concept of a true solitary plasmocytoma of bone. Stewart and Taylor’s case, herein further reported, is one instance. The other, that of Chesterman (1936), received added confirmation from a second report (Chesterman 1948) in which he stated that the patient was still well after twelve years. In a personal communication Chesterman has informed the writer that the patient died nearly four years later, or nearly sixteen years after amputation, from a cerebellar astrocytoma. Necropsy failed to show any evidence of plasmocytoma.

Two more examples of long survival have been reported by Christopherson and Miller (1950). In their Case 1 the patient was living sixteen years after hemimandibulectomy for solitary plasmocytoma of the mandible, and in their Case 2 a survival of twelve years after curettage and radiotherapy for a tumour of the uppermost third of the tibia was recorded. Yentis (1956), from personal communication with Christopherson in 1955, quoted later survival figures of twenty-two and nineteen years.

Notwithstanding these two instances Yentis believed that solitary plasmocytoma of bone, given a sufficiently long time, would eventually develop into multiple myeloma. Carson et al. (1955) were of the same opinion. Lichtenstein (1959) also thought that this was the “great likelihood” but on the basis of certain recorded cases with necropsy he admitted the possibility of a genuine solitary tumour. Jaffe (1958) considered that there were a few patients who, many years later, did not show myelomatous involvement of other bones. He specifically quoted the case of Stewart and Taylor. Raven and Willis (1949) believed that a truly solitary plasmocytoma of bone was an entity distinct from myelomatosis and that it could be cured by adequate local treatment. That all cases of solitary plasmocytoma of bone do not develop into multiple myeloma seems reasonably proven by a small group of cases which have been followed up for a long period after histological confirmation (Table I). Stewart and Taylor’s case is outstanding in this respect, with a thirty-five year survival.
TABLE I
CASES OF SOLITARY PLASMACYTOMA OF BONE WITH LONG SURVIVAL

<table>
<thead>
<tr>
<th>Author</th>
<th>Age (years)</th>
<th>Sex</th>
<th>Site of tumour</th>
<th>Treatment</th>
<th>Survival after treatment (years)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Stewart and Taylor (1932)</td>
<td>34</td>
<td>M</td>
<td>Humerus: uppermost third</td>
<td>Amputation</td>
<td>35</td>
</tr>
<tr>
<td>Chesterman (1936)</td>
<td>39</td>
<td>M</td>
<td>Tibia: uppermost third</td>
<td>Amputation</td>
<td>16</td>
</tr>
<tr>
<td>Christopherson and Miller (1950)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Case 1</td>
<td>29</td>
<td>F</td>
<td>Mandible: left half</td>
<td>Hemimandibulectomy</td>
<td>22</td>
</tr>
<tr>
<td>Case 2</td>
<td>25</td>
<td>F</td>
<td>Tibia: uppermost third</td>
<td>Curettage and radiotherapy</td>
<td>19</td>
</tr>
<tr>
<td>Wright (1961)</td>
<td>33</td>
<td>M</td>
<td>Femur: mid-shaft</td>
<td>Amputation</td>
<td>22</td>
</tr>
</tbody>
</table>

SUMMARY

1. A case of solitary plasmacytoma of bone with survival for twenty-two years after amputation is described.
2. Recent follow-up of a previous tumour reported from this department by Stewart and Taylor in 1932 has shown survival for thirty-five years after amputation.
3. These two instances of long survival, together with three culled from the literature, substantiate the hitherto doubted concept of a true solitary plasmacytoma of bone.

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REFERENCES