ANGIOMA OF A MALE BREAST WITH MALIGNANT CHARACTERISTICS

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This case is presented because of its great rarity and the scant attention the condition receives in the literature. Only casual references to angioma of the breast are made in some standard works,[1, 2] whilst others do not mention it at all. This fact is rather surprising, since Ewing[3] described it in his work in 1919. The first reference to it was made by Bormann[4] in 1907. In all 18 such cases have been described previously. These were all in females. There are thus sufficient cases on record for it to be regarded as a clinical entity, and therefore to put on their guard clinicians and pathologists alike in dealing with a similar case, rare as it may be.

CASE HISTORY

The patient was a young African male aged 17 years, in good general health, mentally and physically. He gave a history that 'ever since he can remember' the chest wall had been slightly more prominent on the left side than the right. The left breast began to increase in size more noticeably in the last 2 years, i.e. since about puberty.

It had reached the size of a cricket ball when, 2 weeks before admission, the left breast began to swell rapidly and caused pain, until it reached the size shown in Figs. 1 and 2. There is no known history of trauma. At this juncture a doctor was consulted. A diagnosis of breast abscess was made and the swelling was incised. The result was alarming. A brisk haemorrhage ensued, which was controlled with the assistance of a fibrin foam preparation and gauze dressings, but oozing persisted for a few days.

During this interval the patient was transferred to hospital, 70 miles away. On admission his condition was very good and, apart from the discomfort of the tumour, he was quite happy and comfortable. Temperature, pulse and blood pressures, etc., were normal and no other abnormality could be found in any of the systems.

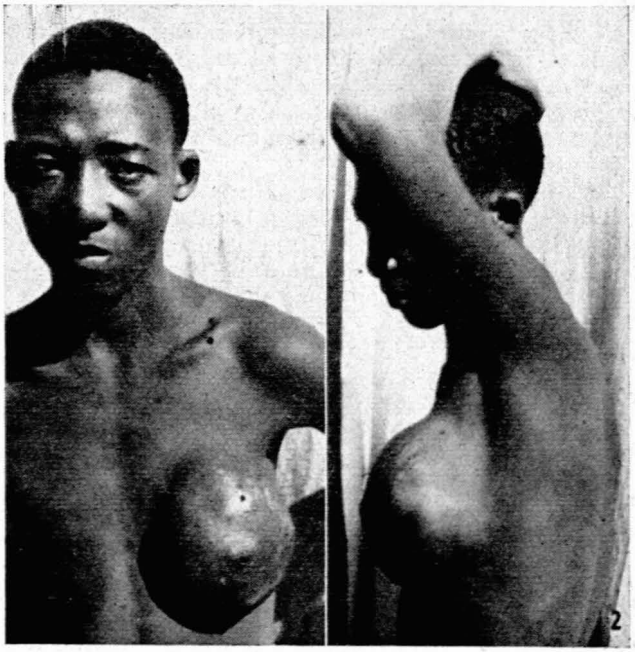
The tumour was almost circular and measured 8 inches across its diameter. It closely resembled a female breast, as may be seen in Fig. 1, except for a rather small nipple. The mass was soft and fluctuant, but tense in some areas and softer in others. It was well demarcated from the surrounding chest wall but was not mobile, being fixed to the underlying pectoral muscle. It was non-tender, although apparently it had been tender before incision, and the surface temperature was higher than that of the surrounding skin. The overlying skin was normal over the firmer areas, but thin and shiny where the tumour was softer. Redness cannot be observed clearly in such dark-skinned individuals, but the skin over the tumour was decidely darker than the rest of the body. The wound of the first incision, about ½ inch long, was closed and dry, and filled with black clotted blood.

A tender axillary gland, about the size of an almond, was palpable on the left side. It was fairly freely mobile and rubbery-firm. No other adenopathy was present.

A haemic bruit, rather faint and muffled, could be heard on auscultation of the tumour.

Radiology of the skeletal system showed no abnormality; nor did similar investigation of the chest and abdomen.

The diagnosis of a vascular tumour was fairly obvious. In view of the rapid increase in size, and the pain, it was thought that malignant change was present, and angiosarcoma was diagnosed. The treatment therefore decided upon was removal of the breast, which was carried out by radical mastectomy. The wound was closed completely and it healed, somewhat surprisingly, uneventfully.



Figs. 1 *and* 2.

There was no disability and no untoward sequelae occurred in the subsequent 6 months. Regrettably, the patient was not seen again—the usual follow-up difficulty with such patients.

Macroscopically the tumour extended to muscle, and consisted largely of numerous blood-filled spaces of varying sizes and shapes, the more superficial being the larger. Some areas of the tumour were covered by skin and fascia, others only by skin, which was adherent. Scattered areas of fat and fibrous tissue intermingled with these 'blood-lakes' which were not delimited by any form of capsule.

Histologically the vascular spaces were lined by endothelium, which was poor in parts and tended to be absent where covered by skin only. The mesenchymal tissue was richly vascular with capillaries and veins and vessels with varying development of muscle coats. Some irregular perivascular cells and scattered leucocytes were evident. There was no evidence of infiltration, mitosis or other sign of malignancy.

Section of the gland showed inflammatory change only.

DISCUSSION

The first reaction to be expected from this case history is that the tumour, after all, was not malignant, as there was no evidence of infiltration, no glandular involvement, and no histological evidence of malignancy.

It would be appropriate at this juncture to point out the variety of nomenclature that has been attached to this tumour, as is so often the case with any little understood condition. It has been called angiosarcoma, haemangioblastoma, malignant angioma or haemangio-endothelioma, etc., and even benign metastasizing angioma, which, of course, is a classic contradiction of terms, but illustrates the difficulties in histological diagnosis.

Let it be said at once that errors of microscopic diagnosis are very frequent in this condition. McClanachen and Hogg,[5] in their review of 6 cases, state that the disarmingly innocent histological appearance of angiosarcoma (haemangioblastoma) is most misleading, and that the tumour is in fact malignant. This view is similarly held by Patrick, Jarvie and Miln,[6] who recently described 3 more cases. The experience of most of the writers[7-10] who have recorded cases has been similar.

Several aspects of the present case closely resemble those features which are common to all the malignant vascular tumours of the breast that have been described. The patients are nearly always young, in their 2nd or 3rd decade, and the history is recent, being a matter of months. Trauma appears to have played some part.

In all cases the primary growth is massive, and has attained its size in a few months. There is invariably overlying skin discolouration. As in this case, the tumour consists of a large friable haemorrhagic mass varying in appearance in different parts, both macroscopically and microscopically. There is never any involvement of axillary nodes, and no evidence of encapsulation.

Further important negative points are the absence of discharge from the nipple, retraction of the skin or oedema.

McClanachen and Hogg[5] hint that benign angiomas are never of bulky size but are always small and discrete, and the walls of their vessels are thicker. One readily admits that histologically angiosarcoma in some areas is of this nature, but one feels that serial sections will almost always reveal the features of malignancy.

Histologically there was no confirmation of malignancy in the present case. Patrick, Jarvie and Miln[6] emphasize the difficulty in distinguishing malignant cells in these tumours, and quote several instances where they were in fact not found, but nevertheless the patients succumbed from metastases. McClanachen and Hogg[5] confirm this by their assertions that the histology is deceptively benign to those unfamiliar with the condition. In no fewer than half the cases recorded malignant characteristics were not found histologically, but all the cases died from secondary spread in an average of just over 2 years; hence the erroneous term benign metastasing angioma.

The tumour in question seemed obviously a cavernous haemangioma of developmental origin arising in the left breast. The malformation increased in size *pari passu* with the host until, probably owing to inadvertant minor trauma and inflammatory change, it rapidly became larger. It was situated in an exposed region, which in a young African male is particularly prone to repeated minor trauma. It would be feasible that this and low-grade infection should give rise to a rapid increase in size, even if the benign nature of the mass were accepted without question. This explanation would be even more acceptable concerning the events in a malignant mass in which trauma, causing interstitial haemorrhage in a friable mass, added to the size of an already rapidly growing tumour.

The combination of the above evidence, one feels, must be accepted as strong argument in favour of malignancy in this case.

No close relationship with the pectoral muscle was made out in the way of bundles of muscle fibres between the endothelial lined spaces. This discounts the possibility of haemangioma of muscle, also a rarity, but perhaps more generally recognized.

The ultimate outcome in this case is not known, and it is indeed most unfortunate that adequate follow-up was not possible. Had it been, the diagnosis of malignancy would doubtless have been confirmed.

A common cause of death is haemorrhage from a secondary deposit. Haemoptyses are frequent. The average interval between diagnosis and death is just over 2 years, often less than 1 year.

Aetiology

A brief mention of aetiology may be made. Other reported cases give no indication of the previous existence of a congenital malformation. In the female the normal contours of the breast would easily obscure this; a small unnoticed mass could rapidly enlarge and become prominent, and perhaps even resemble a subcutaneous haematoma. The occurrence of this growth in a male breast provided a means of closer observation, and the existence from birth of a mass seems fairly certain.

An interesting suggestion is the hypothesis of multifocal angiomatous malformations, simulating metastases. There is little evidence to support this, apart from the constant post-mortem findings. Local recurrence and pulmonary metastases are the most frequent. Other sites of spread are to bone and viscera.

Differential Diagnosis

The diagnosis should present less difficulty once a case has been seen or the possibility of the condition realized. It should be differentiated from other forms of sarcoma, adenocarcinoma, giant fibro-adenoma, cystosarcoma phyllodes, haematoma, lipoma, unilateral hypertrophy, infected cysts, and tumours of the chest wall.

Treatment

A satisfactory method of treatment is not known. This may be due, in part, to the delay in diagnosis and in recognition of the malignancy of the tumour. In the cases reviewed, all known forms and combinations of treatment were carried out with apparently little difference in the end-result. The methods used included both local and radical excision of the breast, with or without radiotherapy. One case was treated with pre-operative radiotherapy followed by radical excision.

Radical mastectomy was performed in the present case, but the outcome is not known. As in the treatment of malignant disease of the breast in general, the question of local or radical operation is still debated. The constant absence of axillary glandular involvement would favour local removal. However, unless one can be sure of removing the mass from the pectoral fascia and muscle, it is probably best to remove this structure. By and large, the treatment of choice at present is probably simple mastectomy followed by radiotherapy. The use of roentgen rays should always follow either surgical procedure. The response of secondary deposits to radiotherapy is disappointing.

SUMMARY

A case of angioma of the breast (in a male) is presented.

This rare tumour and its characteristics are described, and discussed.

A case is made for the malignancy of the case presented.

The importance of serial section in revealing malignant cells is stressed.

Brief mention is made of the aetiology, differential diagnosis and treatment.

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REFERENCES

1. Willis, R. A. (1948): *Pathology of Tumours*. London: Butterworth.
2. Boyd, W. (1955): *Pathology for the Surgeon*. Philadelphia and London: Saunders.
3. Ewing, J. (1919): *Text-book on Tumours*. Philadelphia and London: Saunders
4. Bormann, R. (1907): Beitr. path. Anat., **40**, 372.
5. McClanachen, B. J. and Hogg, L. Jn. (1954): Cancer, **7**, 586.
6. Patrick, R. S., Jarvie, J. and Miln, D. C. (1957): Brit. J. Surg, **45**, 188.
7. Robinson, J. M. and Castleman, B. (1936): Ann. Surg., **104**, 453.
8. Adair, F. E. and Herrman, J. B. (1946): Surgery, **19**, 55.
9. Tibbs, D. (1953): Brit. J. Surg., **40**, 465.
10. Mallory, T. B. *et al.* (1949): New Engl. J. Med., **241**, 241.