Lymphangiosarcoma

Report of A Case

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In 1948, Stewart and Treves[1] described a new type of neoplasm beginning in edematous extremities following radical breast surgery. Since the initial case report, 75 patients have been described.[2] Of this number, those having interscapulo thoracic amputation had the best results. Of 25 cases, 5 were living 1 to 11 years postoperative. Only 2 of 18 who had radiotherapy are alive, 1, 2 years after treatment and 1, 12 years. All 13 who had wide excision and irradiation expired. Ten had amputation of the arm and of these 1 was alive 51 months following surgery. All 10 who had no specific treatment were dead within 1 year.

The following case concerns a patient who had interscapulo thoracic amputation and various other modalities of therapy including one unique approach.

CASE REPORT

This white female secretary, aged 48, was initially seen in January 1955, because of low back pain. It was felt that the patient's symptoms were those of a ruptured disc and surgery for this was performed with a very satisfactory result. At this time she gave a history of having a radical left breast amputation performed in 1952, which was followed by x-ray therapy. Within 1 month following breast surgery, she developed edema of the left arm.

She did well until 1957, when she began having episodes of cellulitis of the left arm. This was characterized by sudden temperature elevation, chilliness, redness and increased edema of the extremity. The process frequently involved the chest wall. It usually responded promptly to antibiotic therapy. The patient had 3 such episodes from 1957 to 1960.

She was seen in May 1962, because of the sudden appearance of a bluish area in the left arm which she thought had resulted from bowling. The patient was treated for about 2 weeks as if this was a thrombosis and when no improvement occurred, a biopsy of the lesion was made and the diagnosis of lymphangiosarcoma was established. The patient had a forequarter amputation done on June 9, 1962, which was less than 6 weeks from the time the lesion was noted (Fig. 1).

At the time of surgery, the patient received 15 mg. Thio-TEPA and this was repeated on the first 2 postoperative days. The patient appeared to do well until November 1962, when some telangiectatic areas were noted on the chest wall. A biopsy of this was taken and found to be negative for malignancy. In February 1963, the patient developed a bluish area at the site of previous surgery and a biopsy taken revealed recurrent lymphangiosarcoma (Fig. 2).

It was felt the treatment of choice at this time was to give the patient both superficial and deep x-ray therapy. This was performed to maximum tolerance. Following this therapy the patient's lesions completely disappeared.

In April 1963 (2 months later) the patient suddenly had an explosive recurrence of metastatic lesions in the area where she had previously received irradiation; 3 lesions appeared at the same time.

It was decided to give the patient a course of intravenous nitrogen mustard (30 mg.). There seemed to be marked improvement of the lesion following this therapy with complete disappearance for nearly 1 month. At this time x-ray of the chest still showed no evidence of malignancy.

The patient was hospitalized in May because of leukopenia, nausea, vomiting and some dyspnea. It was felt that these symptoms were produced by the nitrogen mustard therapy. Chest x-ray showed some patchy infiltration in the lung fields which was thought to be due to x-ray therapy. She improved in a short period of time on symptomatic therapy. The patient's lesions began recurring in June and became progressively worse.

In July 1963, her chest x-ray revealed infiltration in the left lung field which was interpreted as neoplasm. It was the feeling of the attending physician that this situation was now becoming rapidly hopeless. It was thought at this time to try a form of therapy never previously employed as far as can be ascertained in medical therapeutics. Blood was taken from the placenta and umbilical cord immediately after the delivery, typed and cross-matched with the patient's blood and given to the patient as soon as feasible (Table I).

During and following this therapy, the patient appeared to feel considerably better. There was no change in the growth appearance of the lesions on the chest wall. The patient left the hospital and appeared to do well for several weeks when she began to go downhill again.

The patient was tried on Chlorambucil which had no effect on the lesions. In addition she received steroid therapy with no apparent benefit. She was readmitted to the hospital February 3, 1964 and required several thoracenteses. Thio-TEPA was inserted into the chest cage which had little effect on the course of events. The patient went downhill rapidly and expired on March 30, 1964.

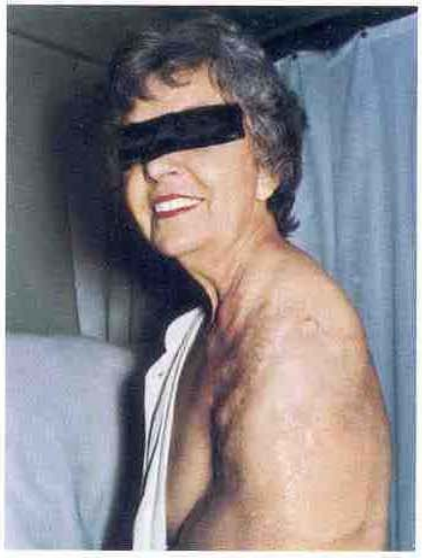

Fig. 1. Appearance following forequarter amputation.

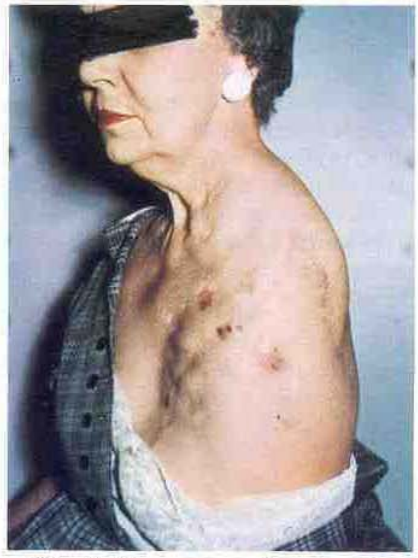

Fig. 2. Appearance of metastasis following surgery.

Postmortem examination was obtained and revealed lymphangiosarcoma invading the chest wall from the neck to the posterior crest of the iliac and from the sternum to the vertebrae. The skin displayed wart-like hardness. The left lung was completely collapsed and had been replaced by tumor. There was no evidence of spread to any other organs.

Table I

UMBILICAL BLOOD CHART

	cc
7/11/63	20
7/12/63	20
7/14/63	20
7/16/63	20
7/17/63	30
7/18/63	10
7/19/63	45
7/20/63	50
7/22/63	38
7/23/63	50
7/26/63	50
7/29/63	20
7/30/63	50
7/31/63	52
8/2/63	30
8/5/63	25
8/8/63	47
Total	577

DISCUSSION

The author felt that possibly, since malignancies are relatively rare in new-born children, that there might be some factors in their blood which prevent them from occurring. It is well known that the body contains growth-stimulating factors. Possibly there are factors that inhibit growth which have not yet been discovered. Based on this idea, the giving of umbilical blood might make some change in the course of a neoplasm.

It is recognized in the above case that this theory was not attempted until it was deemed hopeless. What effects such a procedure might have on individuals who are not as far advanced is yet to be evaluated.

Search of the literature revealed that in 1959, Dr. H. Th. Schreus[3] in Germany conceived of the idea of "youth factors" which he thought inhibited the appearance of melanosarcomas in pigmented nevii until after puberty.[4] He took blood from young children and gave it to adult patients with melanosarcoma with some seemingly good effect. He did not use umbilical blood. The patients treated in this series were also far advanced.

The giving of fetal blood required a considerable amount of cooperation and coordination

between the obstetrical department, laboratory department and attending physician. The object was to give the blood to the patient as quickly as possible following its collection in the delivery room under sterile conditions. Since this individual had only one arm, it made it even more difficult to bring this about.

It was the feeling that the therapy was of some temporary benefit other than might be expected from emotional factors.

SUMMARY

The seventy-seventh case of lymphangiosarcoma is recorded which was treated by forequarter amputation and many chemotherapeutic agents. In addition, this patient received blood obtained from the umbilical cord from which she seemed to derive some temporary benefit.

REFERENCES

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3. Schreus, H. Th.: Dtsch. Med. J., 9:319, 1958.
4. Hess, Paul and Hess, Dieter: Das Melanomproblem in never Sicht, Strahlentherapie, Vol. 119, December 1962.

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