Angiosarcoma as a Potential Consequence of Autologous Lymph Node Transplantation for Lymphoedema

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With high interest have we read the article by Vignes et al., recording complications (lymphoedema, lymphocele, hydrocoele and donor-site pain) in patients undergoing autologous lymph node transplantation (ALNT) for limb lymphoedema. In addition, we recently witnessed a fatal case of angiosarcoma in a 21-year-old woman. We were amazed by the unusual presentation of this aggressive tumour in such a young otherwise healthy woman, located in the right groin exactly at the site of ALNT performed 14 and 7 years before.

This patient had a painful lump on the right thigh that had increased in size over a year (Fig. 1). She had had primary lymphoedema of the right part of body since she was 3 months old. She had had ALNT in the right groin at the age of 7 years repeated at the age of 15 years. The histological diagnosis of this lump was a cutaneous epithelioid angiosarcoma. The patient died from pleural metastases 3 months later.

Occurrence of angiosarcoma in a primary lymphoedema remains a rare event. The mechanisms for development of lymphoedema-associated angiosarcoma may involve local disturbance of angiogenesis. Lymphatic impairment may contribute to excessive production of pro-angiogenic cytokines from the Vascular Endothelial Growth Factor (VEGF) family, and VEGFs are overexpressed in most angiosarcomas.

ALNT could have enhanced local production of angiogenic growth factors and thus development of the angiosarcoma. There is increased expression of VEGF at the recipient site after such surgery, and in a study of patients with ALNT, VEGF-C mRNA expression in human lymph nodes was found to be higher than in other tissues.

As a conclusion, ALNT also raises concerns regarding the potential risk of neoplasia. The safety of these procedures cannot be presumed before long term observational cohort studies have taken place.

CONFLICTS OF INTEREST

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REFERENCES


