Primary Angiosarcoma of Chest Wall and Cutaneous Metastasis: A Systematic Review and Pooled Analysis of Published Reports

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Abstract

Angiosarcomas represent a family of malignant vascular tumors arising from the endothelial lining of blood vessels. Vascular tumors arising in the chest wall are rare, and primary chest wall angiosarcomas (PACW) are even rarer. They have high propensity to metastasize to distant organs, but metastatic colonization of the skin by primary chest wall tumors has never been reported. Presented is a systematic review of the literature to identify cases of PACW reporting skin metastasis. A systematic review was conducted in accordance with PRISMA guidelines. A PubMed, Scopus, Web of Science, and manual search through references of relevant publications was used to identify all published case reports of PACW. Data extracted from each case included age, sex, symptomatology, immunohistochemistry markers, metastasis, management, follow-up, and outcome. The systematic review identified 9 publications reporting 11 cases of PACW. Mean age at presentation was 35 years (range, 8-84 years). Treatment strategies included surgical excision alone (n = 5), surgical excision and adjuvant radiotherapy and chemotherapy (n = 3), and radiotherapy and chemotherapy alone (n = 2). Death was the final outcome in 44.44% of patients. There was no report of cutaneous metastasis in any of the included cases; however, nonskin metastasis was reported in 2 cases. Based on this systematic review, there was no case found of PACW metastasizing to the skin.

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Introduction

> Skepticism is a healthy response to diagnosis of any tumor as angiosarcoma.
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> —Lane, 1952

Angiosarcoma (*angio*-blood or lymph; *sarcoma*-flesh or connective tissue) is an aggressive malignant vascular neoplasm of high metastatic potential harboring dismal prognosis.[1,2] It is characterized morphologically by anastomosing vascular channels lined by rapidly proliferating, extensively infiltrating anaplastic cells derived from endothelium of blood vessels.[3] Angiosarcomas (AS) are subclassified into cutaneous, lymphedema-associated, radiation-induced, primary breast, and soft tissue.[4] The clinical presentation varies depending on the anatomic site involved. They may occur in any region of the body but are more frequent in skin and soft tissue. They also can originate in liver, breast, spleen, bone, or heart.[5] Angiosarcoma has a high rate of metastasis involving lungs (most commonly), liver, regional lymph nodes, bone, and other sites.[6]

A general review of literature revealed that thoracic angiosarcomas, like those arising from pleura,[7] heart,[8] and aorta,[9] have been reported to metastasize to skin. But we did not come across any report of primary chest wall angiosarcomas (PACWs) with skin metastasis.

It often poses a diagnostic and therapeutic challenge for many clinicians who are unfamiliar with the condition. The present study includes a systematic review of literature on the subject with a summary of the clinical and demographic parameters of all reported cases.

Methods

The aim of this systematic review was to identify cases of primary angiosarcoma of chest wall metastasizing to the skin. To facilitate this, an exhaustive review of the literature was carried out first to pool in cases of PACW. This was then followed by their evaluation for cutaneous metastasis.

With search terms "Primary Chest Wall Angiosarcoma" OR "Primary Chest Wall Haemangiosarcoma" OR "Primary Chest Wall Haemangioendothelioma," we performed a systematic

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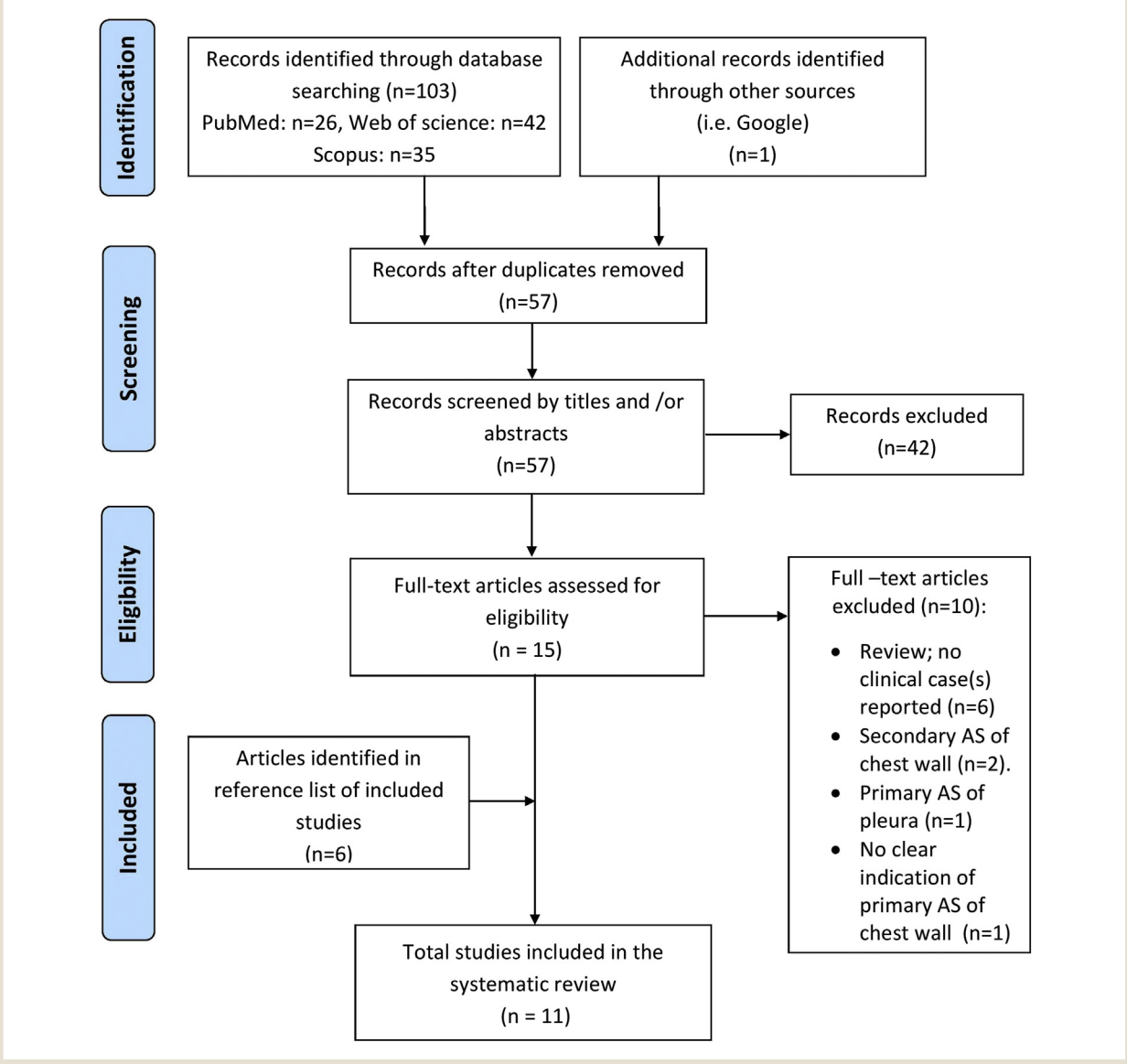
electronic search in the following databases through December 25, 2016: PubMed (1998 to present), Web of Science (1998 to present), and Scopus (1998 to present). The studies were restricted to humans and English language with no constraint on the type of article.

To find additional relevant records in the gray literature, Google was used. All studies were considered eligible for inclusion if they clearly reported PACW and were deemed potentially relevant if they reported cutaneous metastasis. Studies were excluded if a clear diagnosis of PACW could not be ascertained. Duplicate records and redundant publications were discarded. Articles were screened based on the titles and/or abstracts. All the studies fulfilling the inclusion criteria were retrieved and final selection was based on full-text review. The references of all the selected articles also were put to scrutiny to identify other potential studies. Each included article was reviewed by the author and, where possible, data extracted from each case included age, sex, symptomatology, immunohistochemistry markers, metastasis, management, follow-up, and outcome. The literature search and study selection process was in accordance with PRISMA guidelines (Figure 1).

Results

A total of 104 records were obtained through database searching and other sources. Forty-two records were excluded after screening of titles and/or abstracts. Fifteen full-text articles were evaluated and 10 were discarded, as they failed to meet the inclusion criteria. The reference list of the selected 5 articles was also reviewed and 6 relevant studies were added to the list.

Figure 1 PRISMA Flow Diagram of Literature Search and Study Selection Process for the Systematic Review



Abbreviation: AS = Angiosarcoma.