

ORIGINAL ARTICLE Reconstructive

Utility of Multimodal Treatment Protocols in the Management of Scalp Cutaneous Angiosarcoma

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Background: Cutaneous angiosarcomas are rare, aggressive tumors with high recurrence and poor prognosis. We share our experiences with the challenging surgical management of these lesions, focusing on both ablative and reconstructive outcomes.

Methods: Retrospective cross-sectional chart review was conducted of patients diagnosed with scalp cutaneous angiosarcoma between 2005 and 2021. Resectability, defect reconstruction, and survival outcomes were analyzed.

Results: Thirty patients were included, 27 (90%) men and three (10%) women, with a mean age at diagnosis of 71.77 ± 7.3 years, and mean follow-up of 429.43 ± 305.6 days. Only 12 patients completed their regular follow-up, while the remaining patients died. There was a median survival of 443.50 days (range, 42–1283) and median time to recurrence of 21 days (range, 30–1690). Multimodal therapy compared with surgery alone had a significantly better overall median survival (468 days versus 71 days; P = 0.021). Defect coverage was obtained in 24 cases (75%) through an anterolateral thigh flap, two patients (6%) with a local transposition flap, and one patient (3%) with a transverse rectus abdominis myocutaneous flap. The remaining three patients received a skin graft. All of the flaps survived, with one requiring a vein graft for venous congestion.

Conclusions: Timely multimodal therapy with a histologically safe margin, combined with adjuvant therapy, improves survival and delays recurrence and metastasis, in cutaneous angiosarcoma patients. An anterolateral thigh flap facilitates the coverage of wide defects. Further investigations of advanced treatment modalities such as immunotherapy and/or gene therapy are required to deal with this highly aggressive tumor. (*Plast Reconstr Surg Glob Open 2023; 11:e4827; doi: 10.1097/GOX.00000000004827; Published online 3 March 2023.*)

INTRODUCTION

Angiosarcomas are rare and highly aggressive tumors that account for less than 1% of all sarcomas.¹ These neoplastic cells exhibit endothelial differentiation with vascular or lymphatic origin.² Angiosarcomas may arise in any part of the body, but are more common in soft tissue than in the bone. The peak age of incidence appears to be the seventh decade, and men are more affected. The head and neck area is probably the most common

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Copyright © 2023 The Authors. Published by Wolters Kluwer Health, Inc. on behalf of The American Society of Plastic Surgeons. This is an open-access article distributed under the terms of the Creative Commons Attribution-Non Commercial-No Derivatives License 4.0 (CCBY-NC-ND), where it is permissible to download and share the work provided it is properly cited. The work cannot be changed in any way or used commercially without permission from the journal. DOI: 10.1097/GOX.00000000004827 site of diagnosis, whereas the breast is the most common site of radiation-induced angiosarcoma development.¹ Clinically, angiosarcoma presents asymptomatically as a hematoma-like lesion with associated nodules, papules, plaques, or exophytic lesions that develop on the surface of the progressing tumor.³ The relative survival rates for cutaneous angiosarcoma of the scalp and neck areas range between 34%-39% and 14%-17% at 5 and 10 years, respectively.^{4,5} Current treatments are mainly surgical resection for localized disease, although with a challenging margin control due to the extensive infiltration of these tumors, particularly in the head and neck area. Additionally, strong evidence supports the utility of a combined approach in which systemic chemotherapy and radiation therapy in addition to surgical resection have been statistically found to improve the overall

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survival outcome.^{6–10} Furthermore, histopathological clear surgical margins have been reported to correspond with a better prognosis.¹¹ We have previously reported our experience with 15 cases of cutaneous angiosarcoma and the positive impact of multimodal therapy on overall survival.¹²

In this present study, we extend the description of our experiences with this disease through a further retrospective analysis of survival and reconstructive outcomes in patients with cutaneous angiosarcoma of the scalp.

METHODOLOGY

A retrospective chart review was conducted on patients managed at Asan Medical Center with a histologically confirmed diagnosis of cutaneous angiosarcoma of the scalp. We selected cases that had been amenable for surgical resection and were treated between 2005 and 2021 with tumor located primarily in the scalp. Patients with a distant metastasis at presentation or regional lymph node metastasis were included if surgical resection and reconstruction had been performed for palliative purposes. Patients with nonscalp tumor location, who refused surgical excision, or who were managed after the inclusion period were excluded from the analysis. We received institutional review board approval for this study.

A detailed, comprehensive history was taken from all patients, who also underwent thorough physical examinations, blood testing, and radiological imaging. The diagnosis of cutaneous angiosarcoma in each case was confirmed through a histopathological examination for the following markers: cluster of differentiation (CD) 31, CD34, factor VIII-related antigen, von Willebrand factor II, podoplanin (D2–40), and nuclear protein marker of proliferation (MKi-67 or KI-67). The data collected for each patient included age at the time of diagnosis, gender, history of smoking, comorbid conditions, extent of lymph node involvement, recurrence and metastasis, treatment modality and reconstructive method applied, surgical margin, postoperative complications (if any), information on last follow-up, and time of death.

Kaplan-Meier curves were used to analyze overall survival, and any differences between patient groups were tested using multivariate analysis and the log rank test to evaluate different factors that may impact locoregional recurrence, metastasis, and overall survival. These variables included tumor size, resection size, satellite lesions at presentation, nodal disease, lymph node dissection, surgical margin status, and mode of therapy (single or combined). Overall survival was defined as the time from diagnosis to death, regardless of the cause. Recurrence was defined as the time from surgical treatment until either local or regional recurrence was documented.

PATIENT EVALUATION AND SURGICAL MANAGEMENT

Preoperative workups included histological confirmation, magnetic resonance imaging of the brain, chest radiography, computed tomography of the neck, positron emission tomography, and bone scans of the whole

Takeaways

Question: What is the best treatment modality of scalp cutaneous angiosarcoma?

Findings: Timely initiated multimodal therapy with surgical resection combined with adjuvant therapy improves survival and delays recurrence and metastasis.

Meaning: Surgical resection with margin control often yields huge defects, for which flap reconstruction assured timely wound healing even in the advanced age group (in this series; anterolateral thigh free flap). This was shown to have its efficacy in optimizing treatment course and allowing for promptly initiated multimodal therapy with chemoradiation.

body to detect any evidence of metastasis. In the operating theater, the boundaries of the primary lesion were marked, and the lateral resection margins were determined. Thereafter, a wide excision was performed while ensuring sufficient safety margins in both the lateral and the deep margins when possible. An intraoperative frozen section biopsy was performed to establish free surgical margins after removing the primary lesion, a cervical lymph node dissection was performed in patients with evidence or suspicion of a regional lymph node metastasis, based on preoperative imaging scans. In terms of the reconstructive plan, defect coverage was achieved with either a skin graft or free flap. In the skin grafted patients, the utility of skull outer table burring was applied in some cases when the defect was denuded of a periosteum during the surgical resection. For patients who received a flap reconstruction, surgery was conducted via a twoteam-approach. Flap options were tailored to the extent of the defect to be covered. Additionally, a cranioplasty was applied when a bone resection was needed during tumor bed control. After discharge, patients were followed regularly in the outpatient clinic with frequent imaging work-ups used to screen for possible recurrences or metastases.

RESULTS

A total of 30 patients were included in the analysis; 27 (90%) were men, and three (10%) were women. The mean age at diagnosis was 71.77 ± 7.3 years, and the mean follow-up period was 429.43 ± 305.6 days.

Further analysis of survivability showed that 12 patients (40%) have survived following the cancer ablation process and continued on routine screening. With regard to the survival outcomes, the median survival was 443.50 days (range, 42–1283) from the time of diagnosis in the study period. The median times to develop recurrence and metastasis were 21 days (range, 30–1690) and 19 days (range, 1–832), respectively. With regards to tumor ablation characteristics, gross tumor size had a mean value of 78.58 ± 161.47 cm³ ranging from 0.36 to 826 cm³. Negative surgical margins were achieved in 20 patients, whereas the remaining 10 patients had

microscopic positive surgical margin on final pathology report with mean resection margin of $4.3\pm2.6\,\mathrm{cm}$. In terms of the treatment modality, eight (26.7%) patients underwent surgery alone and 22 (73.3%) patients had surgery combined with adjuvant therapy. With regard to the chemotherapy protocols in the study cohort, a paclitaxel-based treatment regimen was the main approach. Furthermore, with regard to radiation therapy, a mean dose of 46.87 ± 14.20 Gy was given in fractions. These data are summarized in Table 1.

Univariate analysis of different cancer prognostic outcomes was also evaluated. The effect of tumor size was assessed in which lesions larger than 5 cm (T2 lesions) were compared with those equal to or smaller than this (T1 lesions), and indicated a median recurrence of 97 days versus 205 days (P = 0.393), metastasis of 97 days versus 183 days (P = 0.694), and an overall survival of 310 days versus 419 days (P = 0.641), respectively, although with no statistical significance. In addition, the presence of satellite lesions, or not, showed a median recurrence of 97 versus 105 days (P = 0.64), metastasis of 97 versus 105 days (P = 0.168), and overall survival of 310 versus

 Table 1. Demographics and Tumor Characteristics of the Included Patients

Characteristic	n (%)
Age	
Mean	71.77±7.3 y
Range	59–85 y
Gender	
Men	27 (90%)
Women	3 (10%)
Follow-up	
Mean	429.43 ± 305.6 c
Range	42–1283 d
Tumor size	
T1: ≤5 cm	13 (43.3%)
T2: >5 cm	17 (56.7%)
Satellites at presentation	
-ve	18 (60%)
+ve	12 (40%)
Nodal disease	
-ve	25 (83.3%)
+ve	5 (16.7%)
Mode of therapy	
Surgery alone	8 (26.7%)
Surgery combined with adjuvant therapy	22 (73.3%)
Safety margin status	
Positive	10 (33.3%)
Negative	20 (66.6%)
Resection margin, mean	4.3 ± 2.6 cm
Survival	
Median	443.5 d
Range	42–1283 d
Time to develop recurrence	
Median	21 d
Range	30–1690 d
Time to develop metastasis	
Median	19 d
Range	1-832

468 days (P = 0.086), respectively, again with no statistical significance. Further to this, the presence of a free margin (no compared to yes) was associated with a median recurrence of 55 versus 205 days (P = 0.02), metastasis of 77 versus 244 days (P = 0.012), and overall survival of 71 versus 468 days (P = 0.03), which were significant differences. These data are shown in Figure 1.

Moreover, the absence of any positive lymph nodes was compared with the presence of histopathological positive nodes and was found to be related to a median recurrence of 105 versus 73 days (P = 0.283), metastasis of 105 versus 87 days (P = 0.846), and overall survival of 468 versus 310 days (P = 0.394), respectively, again without significance. Patients with lymph node dissections of any type (eg, sentinel, selective, or modified radical lymph node dissections) compared with cases with no node dissections showed a median recurrence of 205 versus 97 days (P = 0.369), metastasis of 87 versus 244 days (P = 0.291), and overall survival of 310 versus 468 days (P = 0.989), but again without significance. Finally, the treatment modality was compared in which surgery alone was compared with surgery combined with adjuvant therapy and was associated with delayed median time to recurrence of 67 versus 117 days (P = 0.025), delayed median time to metastasis of 75 versus 147 days (P = 0.018), and better median overall survival of 71 versus 468 days, respectively (P = 0.021), all of which were statistically significant differences. These data are presented in Figure 2.

Twenty-seven of the current study (90%) patients underwent reconstruction with flaps, whereas the remaining three (10%) patients had their defects covered with split-thickness skin grafts. With regard to the flaps utilized, a reconstructive plan based on an anterolateral thigh (ALT) free flap was used in 24 (75%) of the patients, to cover defects with a mean surface area of 915.1±463.8 cm². Other flap options included a transverse rectus abdominis myocutaneous free flap in one (3%) patient, and local scalp transposition flap in two (6%) patients. Furthermore, when reviewing the reconstructive approaches applied in our present series, three patients who received skin grafts underwent burring of the bone to prepare the surgical site for grafting. One patient had a craniectomy to treat an erosive bone lesion that mandated a skull reconstruction with a titanium implant. With regard to the micro-anastomosis applied in flap reconstruction cases, all vascular anastomoses were performed utilizing the ipsilateral superficial temporal artery and its concomitant vein or facial artery and vein. In one case, a vein graft was required during surgical revision due to a venous congestion.

DISCUSSION

In our present series of 30 patients with a cutaneous angiosarcoma affecting the head and neck region, the scalp was the main tumor location. The median age at diagnosis in these cases was 73 years, and 27 of 30 of our subjects were men. These findings are comparable to those in earlier investigations of cutaneous angiosarcoma



Fig. 1. Kaplan-Meier plots of the association between the safety margin control status (yes, negative margin; no, positive margin) and local recurrence (A), metastasis (B), and overall survival (C).



Fig. 2. Kaplan-Meier plots of the association between treatment modality (surgery alone compared with surgery combined with adjuvant therapy) and local recurrence (A), metastasis (B), and overall survival (C).

that described a significant male predominance and that the majority of patients were older than 60 years.^{4,6,9,13–15} Although ultraviolet irradiation has been regarded as a risk factor for these lesions, the causes underlying a predisposition to head and neck cutaneous angiosarcoma remain uncertain.^{16,17} Angiosarcomas are classified within the broad category of vascular tumors, and although a clinical history can contribute to their identification, an expert histological examination is still required for a definitive diagnosis.¹⁸

The diagnosis and management of cutaneous angiosarcomas are hampered by an initially benign clinical manifestation of these tumors in which they resemble a bruise on the head, which can cause delays in seeking medical help. Hence, by the time an angiosarcoma is diagnosed in many instances, there may have been widespread and diffuse infiltration of the afflicted area, as well as metastasis. These lesions can grow so quickly if untreated that they can develop into several highly raised nodular lesions with hemorrhagic regions.^{3,19} When reviewing our current patients' manifestations, symptoms (including itching, bleeding, and discomfort) were present, with perhaps the most prevalent symptom being the appearance of a hematoma-like lesion associated with an expanding nodular mass. (See figure 1, Supplemental Digital Content 1, which displays the preoperative image of a representative scalp angiosarcoma

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patient in the study cohort with a nodular lesion and overlying crush as the main presenting symptom (A and B). This patient underwent lesion resection and reconstruction with an ALT free flap (C). Postoperative follow-up findings are shown in D–F. http://links.lww. com/PRSGO/C417.)

Our current series further confirms the poor clinical outcomes for patients with a cutaneous angiosarcoma of the head, as previously reported in many studies.^{4,5,8,13,15,20,21} Twelve patients only (40%) have survived following the cancer ablation process and continued on routine screening. Moreover, the majority of mortality in our series had occurred within two years of the diagnosis. A scalp location of a cutaneous angiosarcoma is generally related to poor overall survival when compared with other sites, and this is most likely owing to the high rate of positive surgical margins for the tumors in this area.²² Our surgical resection protocol aimed at radical resection obtaining a safe margin of at least 5 cm. This was similar to other reports that recommended any margin macroscopically more than 3-5 cm should be considered, as in handling of soft tissue sarcoma.^{15,23} This was also supported by other reports that showed limited resection margin, with less than 1 cm, was associated with poor survival outcomes.²⁴ For that, the optimal safety margin of cutaneous angiosarcoma is difficult to define because it involves the skin extensively.

With a multifocal lesion with satellite lesions or difficult tumor location, surgical resection with sufficient margin is difficult to achieve.³ Additionally, due to disease rareness and limited number of large-cohort studies, optimal amount of resection margin remains controversial. In this series, we managed to obtain a mean resection margin of 4.3 ± 2.6 cm on final biopsy specimen due to the difficulty associated with tumor relation to functional facial structures like the eye. We tried to evaluate the efficacy of margin size on tumor recurrence, time to metastasis and overall survival, and we failed to show any true significant results, largely due to insufficient sample size match (Table 2).

Moreover, obtaining a histopathological negative margin did have a positive impact on overall survival, together with a delay in recurrence and metastasis, in our series. However, we found a relatively high chance of positive resection margin on final pathology assessment involving 33% of the subjects, although we implemented the principle of wide radical resection. Findings similar to these have been reported previously. Andrew et al found that one of the factors associated with a poorer overall survival was the presence of a positive resection margin.²² Clifford et al reported that achieving negative surgical margins was associated with the longest survival outcome for patients with angiosarcoma of the head and neck.²⁵ Therese et al demonstrated that an initial tumor excision with a complete resection produced significantly reduced mortality rates.²⁶ In addition to these reports, the findings of other investigative efforts have supported the contention that a positive surgical margin negatively impacts the overall prognosis.²⁷ However, some authors reported in their series of 29 patients that an initial diagnosis of T1 lesion turned out to be T2 after final pathologies.⁶ Others suggest that wide-margin surgery may not deliver favorable reports, reporting no difference of clinical outcomes between positive and negative resection margin.^{26,28,29} In our series, we did not perform further resection in case of margin involvement at final pathology, rather we preferred to have a timely initiation of adjuvant treatment with radiation and chemotherapies without any delay. Although further resection may achieve clear resection

Table 2. Univariate Analysis of the Effects of Different Factors on Overall Survival

		Р
Variable	n (%)	(Log Rank Test)
Tumor size	≤5 cm: 13 (43.3%) >5 cm: 17 (56.7%)	<i>P</i> = 0.641
Satellites at presentation	-ve: 18 (60%) +ve: 12 (40%)	<i>P</i> =0.086
Nodal disease	-ve: 25 (83.3%) +ve: 5 (16.7%)	<i>P</i> =0.394
Lymph node dissection	-ve: 19 (63.3%) +ve: 11 (36.7%)	<i>P</i> =0.989
Mode of therapy	Surgery alone: 8 (26.7%) Surgery combined with adjuvant therapy: 22 (73.3%)	*P=0.021
Safety margin status	-ve: 20 patients (66.6%) +ve: 10 patients (33.3%)	*P=0.03

margin, this may be associated with increased surgical burden and delay the timing of adjuvant treatment. We recommended the adjuvant treatment should be initiated within 3 weeks after surgical resection. However, this requires additional assessment and evaluation, especially with the adoption of further imaging modalities to evaluate margin status to improve survival outcomes.

The effect of tumor size on survival outcomes (T1 lesions with tumor size $\leq 5 \text{ cm}$ versus T2 lesions with tumor size >5 cm) was also investigated in our current series, but no correlation was found. Other series have also found no association between tumor size and survival,^{4,14} but some reports have indicated that patients with tumors greater than 5 cm have a poorer survival rate.^{6,7,25,30} Moreover, a prior meta-analysis and systematic review of angiosarcoma of the scalp and face reported that patients with tumors smaller than 5 cm had a greater 5-year survival rate.³¹ The inconsistencies between these different reports are likely due to the insidious nature of cutaneous angiosarcoma of the head and neck and the small sample sizes that are available as a result. The nature of this cancer also makes it difficult to estimate the real tumor itself and/or the disease extent, and this may underlie why these variables showed no prognostic value in our current series. For that, at whatever time point attainable, mapping biopsies are done to superiorly appreciate the tumor extent.

Other prognostic factors investigated in our present analyses included the presence of satellite lesions at presentation, which was found not to have any statistically significant impact on the prognosis of our included subjects. Another study on this issue by Pawlik et al did report a statistically significant value for this detection in terms of a longer median disease-free survival in patients who presented with a single lesion.⁶ Oashi et al have found that the presence of multifocal lesions in cutaneous angiosarcoma cases was linked to a poorer prognosis.³² Guadagnolo et al have also reported that the existence of satellite cutaneous lesions was one of the tumor features linked to a poor prognosis,⁷ whereas the results of other studies have indicated no impact of this on survival.⁹ (See figure 2, Supplemental Digital Content 2, which displays the preoperative image of a representative scalp angiosarcoma patient in the study cohort showing satellite lesions at presentation (A, white arrows). This patient underwent wide resection of the lesion followed by scalp reconstruction with an ALT free flap (B, C). Postoperative followup findings are shown in (D-F). http://links.lww.com/ **PRSGO/C418.**)

We investigated the effects of histopathological lymph node positivity in our current analyses and whether a lymph node dissection was performed, but found no association of this with a delay in local recurrence or metastasis, or on any overall survival impact. Prior reports have also demonstrated that resections of soft tissue and cutaneous angiosarcomas did not correlate with overall survival outcomes.²² Moreover, another previous study, in which the majority of patients had head and neck cutaneous angiosarcoma, reported that patients who had a lymph node dissection survived for a median of 34 months but that this was not a statistically significant improvement.²⁵ Furthermore, and interestingly, a previous study reported that regional lymph node metastasis was not substantially associated with overall survival among cutaneous angiosarcoma cases, based on univariate analyses.³²

With regard to the treatment modality, we here investigated the utility of surgery combined with adjuvant therapy and found that it appears to have a positive impact on survival. The findings in many other series have also indicated the significance and survival benefits of adjuvant treatments that incorporate radiation therapy, as well as chemotherapy to a lesser degree.⁶⁻¹⁰ In our present series, the univariate data indicated that multimodal therapy was associated with a significantly improved overall survival, and a delay in both recurrence and metastasis. The treatment of angiosarcoma thus demands aggressive and comprehensive therapy by a multidisciplinary team.⁶ Not surprisingly, the mainstay of cutaneous angiosarcoma treatment is still a combination of surgical intervention together with radiation.7 Notably however, there is no standard unified radiation regimen at this time for these lesions. With respect to the role of chemotherapy in the management of such tumors, studies are also inconclusive. While some reports have demonstrated that adjuvant chemotherapy improves overall survival, this has not been found in others. Chemotherapy is also commonly utilized in the treatment of metastatic cutaneous angiosarcoma and for lesions that cannot be completely removed.³ Doxorubicin-based regimens are regarded as the gold standard for treating soft tissue sarcomas. Taxanes (paclitaxel) have also been demonstrated to be more effective against angiosarcoma. Both anthracyclineor taxane-based regimens are the most often utilized first-line treatments for advanced angiosarcoma. The evidence is currently insufficient to indicate the superiority of either approach, and they are thus employed depending on the scenario being managed.¹¹ Modern therapeutic techniques, including immunotherapy and targeted agent therapy, may prove to be more effective, but this requires further investigations.¹¹ Our treatment algorithm includes surgical treatment combined with adjuvant radiotherapy for all localized cutaneous angiosarcoma. Chemotherapy is considered as an additional treatment after surgery with or without radiation in locally advanced disease. Paclitaxel was considered as the first-line option, followed by docetaxel, doxorubicin, ifosfamide, and gemcitabine. When patients are resistant to conventional chemoregimen, targeted agents and immunotherapies are considered, such as tyrosine kinase inhibitor and antivascular endothelial growth factor agents. Additionally, for patients with metastatic and unresectable tumors, or for those who refuse the surgery, concurrent chemoradiation is considered. The majority of our patients (22 patients; 73.3%) had followed this protocol with significant impact over survival outcomes when compared with those who did not follow such protocol due to adjuvant therapy refusal after surgical resection.

With regard to reconstructive outcome analysis in the present series, the most common reconstructive method after a resection of a cutaneous angiosarcoma of the head and neck was with the use of a free ALT flap. In our current cohort, 24 (75%) patients underwent reconstruction with an ALT flap, followed by a transverse rectus abdominis myocutaneous free flap that was used in one (3%) patient, and local scalp transposition flaps that were applied in two (6%) patients. The remaining cases received skin grafts for soft tissue coverage. All cases were managed in a two-team approach. Tumor ablation involved excising the full layer of skin envelope deep to the periosteum, and in case of tumor invasion to the subcutaneous layer, we perform extensive burring of the skull bone. Free flap coverage is the preferred choice, followed by skin graft. The purpose of flap reconstruction was to provide adequate tissue coverage to prevent the soft tissue problems caused by adjuvant radiation. When patients had additional significant morbidities making them unable to bear the surgical burden, or when poor vascular status prevented performing microvascular surgery, skin grafting was performed as an initial option. In this series, adjuvant radiation or chemotherapies could be initiated at least 3 weeks after operation, which was similar between flap and skin graft options. We often found that wound problems occurred relatively higher in the skin graft group, as there was sometimes loss of the graft, which required further time to be healed with subsequent delay in treatment course. The ALT flap was the preferred flap option, as it facilitated a two-team approach, coverage of large size defects, low donor site morbidity, ease of harvest with a decreased operative time, and a satisfactory aesthetic reconstructive outcome in the head and neck region. The use of an ALT free flap has gained popularity over other options such as a latissimus dorsi flap in providing coverage of a scalp defect, as demonstrated in several investigative reports.^{33–37} Chou et al have also revealed that the ALT flap is more frequently used for scalp reconstructions, particularly after an angiosarcoma excision, due to the above-mentioned advantages.¹³ When the free flap option was considered, superficial temporal vessels were the preferred recipient option, followed by facial vessels when the lesion involved the lateral aspect of the scalp with the potential need for vein grafting.

Last, this study is not void of limitations, and those include its retrospective nature with a relatively small number of participant, considering the rare nature of the disease. Therefore, future studies with a larger sample size and multi-institutional randomized trial together with unified treatment regimen are required to further clarify the best treatment approach.

CONCLUSIONS

Angiosarcoma is a rare malignant tumor with a poor prognosis. Surgery, combined with adjuvant therapy, yields a significant delay in recurrence and metastasis, and an improved overall survival in these cases. Verifying the presence of an adequate and histologically established safe margin is critical. New agents and treatment plans for the management of cutaneous angiosarcoma are needed and require further investigation.

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