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HEAD AND NECK

Comparison of prognostic implications between the 7th and 8th edition of AJCC TNM staging system for head and neck soft-tissue sarcoma in adult patients

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Abstract

Purpose This study aimed to investigate the prognostic factors for head and neck soft-tissue sarcoma (HNSTS) in adults, with the comparisons between the 7th and 8th edition of AJCC TNM staging system.

Methods From a cancer registry of a single, tertiary referral medical center, the medical records of 67 patients treated from February 2005 to December 2017 were reviewed.

Results T1b stage by AJCC 7th edition showed most diverse stage migration by AJCC 8th edition, and T1a or T2b stage by 7th edition remained in T1–3 or T3–4 by 8th edition. T2 stage by 7th edition showed a significantly higher death rate than the T1 stage, with fair discrimination in overall survival. Higher histologic grade and angiosarcoma were significant prognostic factors for recurrence as well as overall survival. Also, nodal and distant metastasis worsen overall survival.

Conclusions In our series of patients with HNSTS, higher histologic grade, angiosarcoma, N1, and M1 stage significantly increased the risk of recurrence and worse overall survival, which was not evident in revised T stage by AJCC 8th edition.

Keywords Head and neck · Sarcoma · Prognostic factor · Staging

Introduction

Head and neck soft-tissue sarcoma (HNSTS) is a rare malignant tumor of mesenchymal origin. It consists of heterogeneous groups of histology and sites of origin [1–5]. The incidence of HNSTS accounts for around 10% of all soft-tissue sarcomas, and within the head and neck area, it occurs more frequently than those arising from the bone. Although it is uncommon among head and neck neoplasms, the oncologic outcome of HNSTS is known to be worse than that for sarcomas from other body regions, because some histological forms of HNSTS show locally invasive and highly metastatic features. Also, even in the early staged tumor, the complex anatomy and adjacent vital structures in the head and neck sometimes makes it challenging to obtain enough surgical margins.

In case of a resectable tumor, the treatment of choice is surgical removal of tumors with adequate resection margins, followed by adjuvant therapy, such as external irradiation and chemotherapy according to histological types and grade [3, 6]. For unresectable tumors or tumors with systemic metastasis, non-surgical treatment modalities are preferred [7, 8]. Radiation therapy (RT) is known to improve the overall prognosis of HNSTS and is especially useful in patients with a high histologic grade and positive resection margin [9]. As the role of chemotherapy in head and neck sarcoma is underestimated, it can be beneficial in limited cases; however, the risk of toxicity and the oncologic benefit must be weighed [7, 10].

Previous studies regarding HNSTS have been mostly small case series of this rare tumor, and they lacked consistency in the tumor stratification and treatment of patients. In some reports, HNSTS and bone sarcoma were analyzed together, which data can be misleading [4, 6, 11]. A few studies have addressed the survival and prognostic indicators of HNSTS, based on the clinical analysis of a relatively

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larger number of patients [10, 12, 13]. As a result, it has been well accepted that the prognostic factors for head and neck sarcoma include the anatomic site, tumor size, resection margin, histological grade, TNM stage, and treatment method. However, there is no study which adopts (1) the recently revised 8th edition of the TNM staging manual by the American Joint Committee on Cancer (AJCC), as well as (2) the most-recommended histological grading system proposed by the FNCLCC (Fédération Nationale des Centres de Lutte Contre Le Cancer) for tumor stratification, the survival outcomes and prognostic factor analysis of HNSTS [14, 15].

This study aimed to assess the tumor stage change between the 7th edition and 8th edition of the TNM staging system and to investigate the oncologic outcomes (locoregional control, LRC, disease-free survival, DFS, and overall survival, OS) and relevant significant prognosticators of HNSTS in adult patients.

Materials and methods

Patients

This single institutional, retrospective study was approved by the institutional review board of Samsung medical center. From a cancer registry of institutional head and neck cancer center, 121 patients were diagnosed and treated with HNSTS from February 2005 to December 2017. Among them, patients who were referred from other medical centers for the management of recurrent or incompletely treated tumor, or patients younger than 18 years old were excluded in the analyses. As a result, a total of 67 patients were enrolled for further analysis, and their demographics, tumor locations, staging, pathologic characteristics, treatment methods, and outcomes were assessed. All subjects were diagnosed by biopsy, and clinical staging was determined based on computed tomography, magnetic resonance imaging, and positron emission tomography-computed tomography before treatment. When regional metastasis was suspected, ultrasonography-guided fine-needle aspiration or core-needle biopsy was performed for accurate cancer staging. In patients who had an operation, resection margin status was evaluated and reported as R0 (negative), R1 (microscopic resection margin positive), or R2 (macroscopic resection margin positive).

Cancer staging was determined according to the AJCC 7th edition and 8th edition, and WHO classification method [16–18]. For histological grading, the three-grading system proposed by French Federation Nationale des Centres des Lutte Contre le Cancer (FNCLCC) was used [19]. All patients underwent periodic surveillance through physical and endoscopic examination and radiologic images. The periodic screening was done every 1 to 3 months in the first year, every 3 to 6 months for the next two years, and every six months during the fourth and fifth years. Recurrence was confirmed by imaging and/or histologic confirmation.

Statistical analysis

We compared the AJCC 7th and 8th T staging with Cohen's kappa coefficient using inter-rater agreement analysis. The maximum kappa coefficient is 1.00 which indicates perfect agreement, and zero indicates no agreement (poor agreement < 0.40, good agreement is 0.40 to 0.75, and excellent agreement > 0.76) [20, 21].

The univariate and multivariate analyses using the Cox proportional hazard model were performed to investigate risk factors for recurrence-free survival (RFS) and overall survival (OS). Several key factors of HNSTS were analyzed for associations to RFS and OS: age, TNM stage, histological grade (G1, G2, or G3), histologic subtypes (rhabdomyosarcoma, angiosarcoma, and others), resection margin (R0, R1), and treatment methods, which were categorized as operation only, operation with adjuvant therapy, and non-surgical treatments (RT, chemotherapy, and concurrent CRT). The significant statistical differences were considered to be those with $p < 0.05$. Multivariate analyses were done with clinical factors that had $p < 0.1$ on univariate analysis. All statistical analyses were done using IBM SPSS software version 20.0 (SPSS, Inc., Chicago, IL, USA).

Results

Clinical characteristics of study patients

Clinical characteristics are described in Table 1. Mean age was 51.9 ± 17.8 years, and gender distribution was male ($n = 41$, 61.2%) and female ($n = 26$, 38.8%). Tumor sites were most common in the scalp, face, and neck skin ($n = 25$, 37.3%), followed by the paranasal sinus/ nasal cavity ($n = 22$, 32.8%), the oral cavity ($n = 5$, 7.5%), the salivary gland ($n = 5$, 7.5%), the larynx ($n = 4$, 6.0%), the nasopharynx ($n = 2$, 3.0%), the oropharynx ($n = 2$, 3.0%), and the hypopharynx ($n = 2$, 3.0%).

By AJCC 8th edition, T stages were distributed to T1 ($n = 9$, 13.4%), T2 ($n = 11$, 16.4%), T3 ($n = 10$, 14.9%), T4a ($n = 31$, 46.3%), and T4b ($n = 6$, 9.0%). N staging consisted of N0 ($n = 48$, 71.6%) and N1 ($n = 19$, 28.4%). M staging consisted of M0 ($n = 57$, 85.1%) and M1 ($n = 10$, 14.9%). Histologic grade 1 was most common ($n = 31$, 46.3%), followed by grade 2 ($n = 19$, 28.4%), and grade 3 ($n = 17$, 25.4%). Histologic subtypes consisted of rhabdomyosarcoma ($n = 23$, 34.3%), undifferentiated pleomorphic sarcoma ($n = 9$, 13.4%), angiosarcoma ($n = 8$, 11.9%) and others.

Table 1 Clinical characteristics of enrolled subjects with head and neck soft-tissue sarcoma ($N=67$)

Characteristics	No. (%)
Age, years (mean ± SD, range)	51.9 ± 17.8 (19–89)
Gender (M:F), *N* (%)	41:26 (61.2:38.8)
Tumor sites, *N* (%)	
Scalp, face, neck skin	25 (37.3)
Paranasal sinus/Nasal cavity	22 (32.8)
Oral cavity	5 (7.5)
Salivary gland	5 (7.5)
Larynx	4 (6.0)
Nasopharynx	2 (3.0)
Oropharynx	2 (3.0)
Hypopharynx	2 (3.0)
Tumor staging, *N* (%)[a]	
T1/T2/T3/T4	10/15/18/24 (14.9/22.4/26.9/35.9)
N0/N1, M0/M1	48/19 (71.6/28.4), 57/10 (85.1/14.9)
Histologic grade: G1/G2/G3[b]	31/19/17 (46.3/28.4/25.4)
Histologic subtypes, *N* (%)	
Rhabdomyosarcoma	23 (34.3)
Undifferentiated pleomorphic sarcoma	9 (13.4)
Angiosarcoma	8 (11.9)
Carcinosarcoma	5 (7.5)
Leiomyosarcoma	5 (7.5)
Fibrosarcoma	5 (7.5)
Liposarcoma	4 (6.0)
Synovial sarcoma	2 (3.0)
Epithelioid sarcoma	2 (3.0)
Alveolar sarcoma	1 (1.5)
Dermatofibromasarcoma tuberans	1 (1.5)
Spindle cell sarcoma	1 (1.5)
Histiocytic sarcoma	1 (1.5)
Treatment modalities, *N* (%)	
OP alone	15 (22.4)
OP OPRT	26 (38.8)
OP OPCT	2 (3.0)
OP OPCCRT	7 (10.4)
RT	1 (1.5)
CT	4 (6.0)
CCRT	12 (17.9)
Oncologic outcomes, *N* (%)	
Recurrence	27 (40.3)
Local	12 (17.9)
Regional	6 (9.0)
Distant	14 (20.9)
Death	30 (44.8)

OP operation, *RT* radiotherapy, *CCRT* concurrent chemoradiation

[a]8th edition of the TNM staging manual by the American Joint Committee on Cancer (AJCC)

[b]Histological grading system proposed by the FNCLCC (Fédération Nationale des Centres de Lutte Contre Le Cancer)

Treatment methods consisted of operation alone ($n=15$, 22.4%), operation with radiation ($n=26$, 38.8%), operation with chemotherapy ($n=2$, 3.0%), operation with CRT ($n=7$, 10.4%), radiation therapy ($n=1$, 1.5%), chemotherapy ($n=4$, 6.0%), and concurrent CRT ($n=12$, 17.9%). About 75% of the patients underwent upfront surgery with curative intent. They were treated by extensive resection and various types of the reconstruction as required; free flap or local rotational flap was commonly employed reconstructive option. In the free-flap group, the anterolateral thigh (ALT) fascio-cutaneous was the most frequently used. Of the 50 patients who underwent surgery, 33 patients received postoperative, adjuvant radiotherapy (median 59 Gy; range 20–70 Gy), with a single daily fraction of 1.8 or 2.0 Gy in 5 days per week for 5–8 weeks, with ($n=7$) or without ($n=6$) chemotherapy. Twelve patients received chemoradiotherapy (CRT), and four patients with distant metastasis at initial presentation underwent palliative chemotherapy, mostly using an anthracycline (Doxorubicin) or an alkylating agent (ifosfamide). As of the last follow-up, the overall recurrence rate was 40.3% (27/67), and death by disease developed in 44.8% of all patients (30/67).

Recurrence was observed in 27 patients (40.3%): local recurrence ($n=12$, 17.9%), regional recurrence ($n=6$, 9.0%), and distant metastasis ($n=14$, 20.9%). Disease-specific death was found in 30 patients (44.8%). In a median follow-up of 25 months, the 5-year loco-regional control and OS rates were 76, 60 and 61%, respectively.

Table 2 Comparison of T staging between American Joint Committee on Cancer (AJCC) 7th and 8th edition for head and neck soft-tissue sarcoma

	AJCC 7th edition				
	T1a	T1b	T2a	T2b	Total
AJCC 8th edition					
T1	5 (7.5)	5 (7.5)	0 (0)	0 (0)	10 (14.9)
T2	3 (4.5)	12 (17.9)	0 (0)	0 (0)	15 (22.4)
T3	1 (1.5)	4 (6.0)	1 (1.5)	12 (17.9)	18 (26.9)
T4	0 (0)	11 (16.3)	0 (0)	13 (19.4)	24 (35.9)
Total	9 (13.4)	32 (47.8)	1 (1.5)	25 (37.3)	67 (100)

Inter-rater agreement of AJCC 7th and 8th T staging: kappa coefficient = 0.269, $p<0.001$ (reference [20, 21])

Stage migration and clinical implications of T stage by AJCC 7th and 8th edition

Comparing the AJCC 7th and 8th edition of T staging of HNSTS, T1b stage by AJCC 7th edition showed the most diverse stage migration by AJCC 8th edition (Table 2). Specifically, in 9 T1a cases by AJCC 7th edition, 5 cases remained as T1, and other 4 cases were upstaged as 3 T2 and 1 T3 stage by AJC 8th edition. On the contrary, 31 T1b by AJCC 7th edition was evenly re-distributed into 17 T1–2 stages and 15 T3–4 stages by AJCC 8th edition. While one T2a case by AJCC 7th edition was up-staged as T3 by AJCC 8th edition, 25 T2b cases by AJCC 7th edition was mostly up-staged as 12 T3 and 13 T4a-b stages by AJCC 8th edition.

Kappa coefficient of inter-rater agreement was 0.269 with statistical significance ($p<0.001$), which indicates poor agreement between AJCC 7th and 8th T staging system.

When the association between T staging and oncologic outcomes were evaluated, local/overall recurrence rate and death by disease were not different in early T stage and advanced T stage classified either, AJCC 7th edition or 8th edition (Table 3). However, advanced T stage by AJCC 7th edition was associated with increased death rate than early T stage (T1, 13/41 vs. T2, 17/26, $p=0.007$), which was not evident in AJCC 8th edition (T1–2, 9/25 *vs.* T3–4, 21/42, $p=0.265$). In similar, when Kaplan–Meier estimate was compared in study patients staged according to T stage by AJCC 7th and AJCC 8th editions (Fig. 1), it was demonstrated that there was a fair discrimination between OS with a statistical significance only in the AJCC 7th edition (7th edition, T1 *vs.* T2, log-rank $p=0.015$, 8th edition, T1–2 vs. T3–4, log-rank $p=0.026$).

Prognosticators for recurrence-free and overall survival

In univariate analysis of recurrence-free survival, histologic grade 2 (hazard ratio 31.245; 95% confidence interval 3.813–256.042) and 3 (hazard ratio 41.648; 95% confidence interval 5.470–317.126) were significant risk

Table 3 Comparison of incidence of local recurrence, recurrence, and disease specific among T staging of AJCC 7th and 8th edition

	Local recurrence	*p*-value	Recurrence	*p*-value	Death	*p*-value
AJCC 7th staging						
T1, *n* (%)	5/41 (12.2)	0.191	14/41 (24.1)	0.197	13/41 (31.7)	**0.007**
T2, *n* (%)	7/26 (26.9)		13/26 (50.0)		17/26 (65.4)	
AJCC 8th edition						
T1–2, *n* (%)	5/25 (20.0)	0.751	12/25 (48.0)	0.321	9/25 (36.0)	0.265
T3–4, *n* (%)	7/42 (16.7)		15/42 (35.7)		21/42 (50.0)	

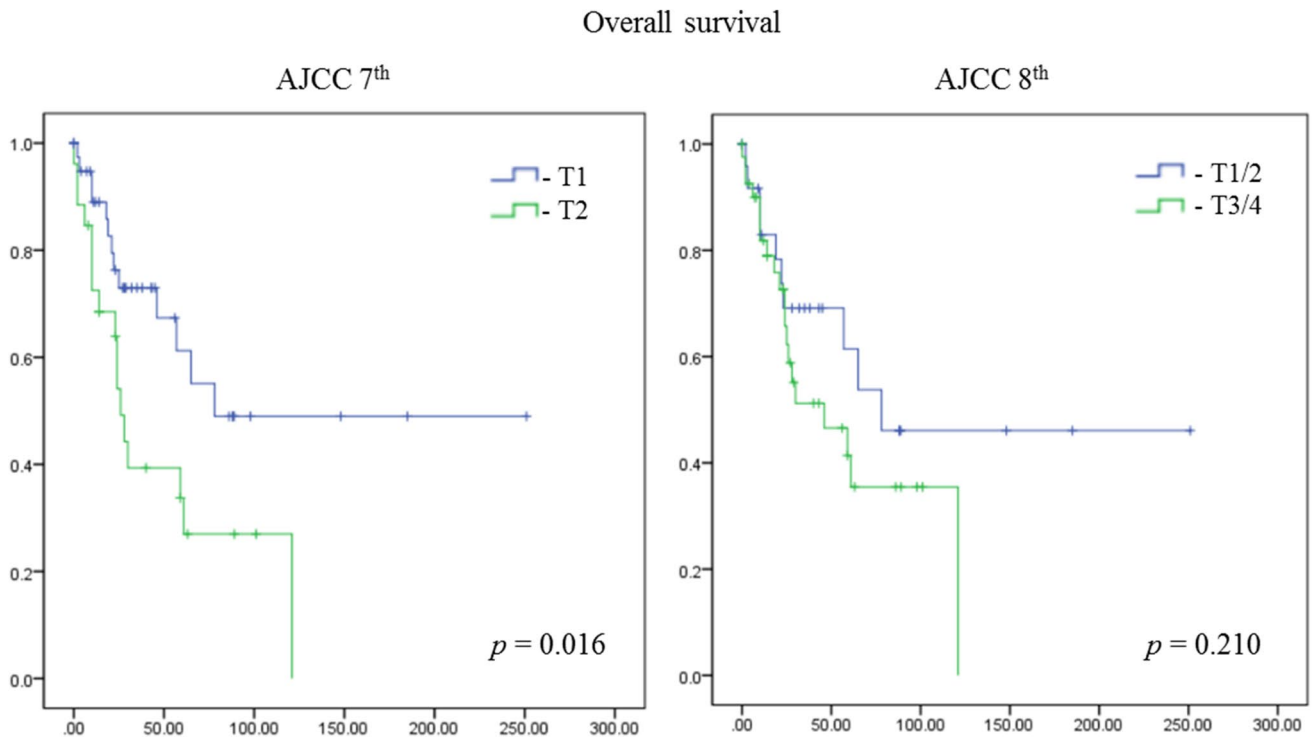

Fig. 1 Kaplan–Meier curves showing the comparisons of overall survival according to T classification by AJCC 7th edition (T1 vs T2) and 8th edition (T1–2 vs T3–4). P value by Log-rank test

factors for recurrence compared to grade 1 (Table 4). Angiosarcoma (hazard ratio, 6.500; 95% confidence interval, 2.328–18.146) had statistical significance ($p < 0.001$) compared to other subtypes; otherwise, rhabdomyosarcoma was not a significant risk factor ($p = 0.648$). R1 resection status was not a significant risk factor ($p = 0.094$) compared to R0 resection. In multivariate analysis, histologic grade 2 (hazard ratio 49.377;

Table 4 Univariate and multivariate analysis using Cox proportional hazard model for recurrence-free survival in patients with soft-tissue sarcoma of head and neck ($N = 67$)

	Univariate analysis			Multivariate analysis		
	HR	95%CI	*p*-value	HR	95%CI	*p*-value
Age (1 year increased)	1.008	0.985–1.032	0.498			
Tumor staging						
T3–4 (Ref: T1–2)	0.715	0.335–1.523	0.384			
N1 (Ref: N0)	1.388	0.603–3.192	0.440			
M1 (Ref: M0)	2.024	0.737–5.557	0.171			
Histologic grade						
G1	Ref			Ref		
G2	31.245	3.813–256.042	0.001	49.377	5.630–433.030	< 0.001
G3	41.648	5.470–317.126	< 0.001	84.893	10.204–706.258	< 0.001
Histologic subtypes						
Others	Ref			Ref		
Rhabdomyosarcoma	0.814	0.337–7.967	0.648	0.426	0.163–1.094	0.076
Angiosarcoma	6.500	2.328–18.146	< 0.001	17.488	4.043–75.642	< 0.001
Resection margin						
R0	Ref					
R1	2.062	0.884–4.811	0.094			

HR hazard ratio, *CI* confidence interval

95% confidence interval 5.630–433.030, $p < 0.001$) and 3 (hazard ratio 84.893; 95% confidence interval 10.204–706.258, $p < 0.001$) were significant risk factors for recurrence for histologic grade 1. Angiosarcoma (hazard ratio 17.488; 95% confidence interval 4.043–75.642, $p < 0.001$) also had statistical significance compared to subtypes.

As for OS (Table 5) nodal metastasis (hazard ratio 3.056; 95% confidence interval 1.456–6.411) and distant metastasis (hazard ratio 3.932, 95% confidence interval 1.642–9.417) were significant risk factors ($p = 0.003$ and 0.002, respectively). Histologic grade 3 (hazard ratio 2.612; 95% confidence interval 156–5.900) was also a significant risk factor ($p = 0.021$) for worse overall survival than histologic grade 1. In terms of the histologic subtypes, angiosarcoma (hazard ratio 7.911; 95% confidence interval 2.652–23.596, $p < 0.001$) was a significant prognostic factor for worse overall survival compared the others. Otherwise, resection margin status and treatment modalities did not present significance as risk factors.

In multivariate analysis, N1 (hazard ratio 2.807; 95% confidence interval 1.185–6.651, $p = 0.019$), M1 (hazard ratio 3.328; 95% confidence interval 1.271–8.717, $p = 0.014$), and angiosarcoma (hazard ratio 7.238; 95% confidence interval 2.335–22.442, $p = 0.001$) remained as significant risk factors for worse overall survival.

Discussion

As AJCC has recently been revised and updated to its 8th edition for bone and soft-tissue sarcomas, tumors are described separately according to the primary sites, and for soft-tissue sarcomas, four specific tumor locations are described: trunk and extremity, retroperitoneum, head and neck, and visceral sites [22]. More importantly, the depth of the primary soft-tissue tumor (superficial or deep from the superficial fascia), which had been considered in the 7th edition, was eliminated in the 8th edition. Also, 2–4 cm size criteria, as well as the invasion of adjoining structures, are included for T stratification of HNSTS. In this retrospective analysis of 67 consecutive patients who had been treated in a single institution for HNSTS, new AJCC 8th edition of cancer staging manual was applied, and it was demonstrated that new edition up-staged previous T1b by the AJCC 7th edition into T2–4 stage, and T2b by the AJCC 7th edition into T3–4 stage. On the contrary, T1a by the AJCC 7th edition was distributed in T1–3 stage by the AJCC 8th edition. Inter-rater agreement analysis demonstrated poor agreement between AJCC 7th and 8th T staging system (κ coefficient = 0.269, $p < 0.001$).

When prognostic implications were assessed and compared between the 7th and 8th edition, T1 vs. T2 staging by the 7th edition showed more significance in regards to the OS. However, this finding must be validated in systematic review or meta-analysis due to the small number of patients in this study and the rarity of HNSTS.

Table 5 Univariate and multivariate analysis using Cox proportional hazard model for overall survival in patients with soft-tissue sarcoma (N = 67)

	Univariate analysis			Multivariate analysis		
	HR	95%CI	*p*-value	HR	95%CI	*p*-value
Age (1 year increased)	1.016	0.994–1.038	0.163			
Tumor staging						
T3–4 (Ref: T1–2)	1.623	0.751–3.506	0.218			
N1 (Ref: N0)	3.056	1.456–6.411	0.003	2.807	1.185–6.651	0.019
M1 (Ref: M0)	3.932	1.642–9.417	0.002	3.328	1.271–8.717	0.014
Histologic grade						
G1	Ref					
G2	1.685	0.514–5.530	0.389			
G3	2.612	1.156–5.900	0.021			
Histologic subtypes						
Others	Ref			Ref		
Rhabdomyosarcoma	1.870	0.832–4.202	0.130	0.878	0.331–2.330	0.795
Angiosarcoma	7.911	2.652–23.596	< 0.001	7.238	2.335–22.442	0.001
Resection margin						
R0	Ref					
R1	1.146	0.481–2.731	0.759			

HR hazard ratio, *CI* confidence interval

In AJCC the 8th edition, histologic grading system by FNCLCC is officially recommended, which classifies the soft-tissue sarcoma into three (grades 1, 2, and 3) according to the total of the scores of tissue type, the extent of necrosis and mitotic counts. In a recently published study with 122 HNSTS patients, it was found that tumor size (categorized by 5 cm and 10 cm according to the 7th edition) and nodal metastasis were independent prognostic factors for locoregional control, and that histologic grade (NCI grading system) was a significant variable for both disease-specific survival and OS [12]. For recurrence-free survival, it was identified that histologic grade 2 (hazard ratio 49.377) and 3 (hazard ratio 84.893) were significant risk factors for recurrence, as compared to grade 1, and this implication was not maintained for overall survival. This result is consistent with a previous report that demonstrated a hazard ratio of 1.73 in grade 3 for local recurrence in soft tissue sarcoma [23]. In the 8th edition for HNSTS, histologic grading was not notated and considered in prognostic stage grouping, which was not stipulated in the head and neck site tumor. As more data file up and support the prognostic implications of histologic grading in HNSTS, it is expected that future edition of AJCC staging manual might incorporate TNM factor as well as histologic grading into the staging system.

According to the clinical practice guideline by national comprehensive cancer network (NCCN), surgical wide resection is recommended as the first line of treatment in case of resectable tumor size with acceptable functional outcomes [24]. In localized cases, extensive resection with an appropriate resection margin is most effective. Head and neck surgeons should secure the safe resection margin without functional and esthetic complications. As the delicate reconstructive surgery should be accompanied by surgical resection at the same time, head and neck surgeons who are experienced in both oncologic and reconstructive surgery are essential for HNSTS treatment. Although the appropriate resection margin width has not been established, the primary tumor was resected with a 1.5-cm width of surrounding tissue in our cohort. Interestingly, however, unclear or incomplete surgical resection margin did not deteriorate RFS or OS in this group of patients. Also, the addition of adjuvant treatment modalities did not improve the oncologic outcomes, either. In contrast, the presence of lymph node metastasis (N) and distant metastasis (M) was significant prognosticator for OS.

Angiosarcoma and rhabdomyosarcoma are known to recur more than did other subtypes and had poor OS [1, 25]. Likewise, in our study, angiosarcoma was a significant risk factor for both RFS and OS, unlike the other subtypes. It was also found that rhabdomyosarcoma increased recurrence risk in our cohort; however, it did not influence OS.

Based on the findings of the present study, it was assumable that initial tumor presentation (N/M stage, histologic grade, and histologic subtype) may be significantly associated with recurrence and survival outcomes, rather than surgical margins or the combination of treatment modalities. Treatment of HNSTS has been changed consequently along with the development of expert opinions and each institution's experiences. Given that there is no randomized controlled trial of treatment of HNSTS, previous studies have discussed inconsistent prognostic factors for oncologic outcomes of this rare tumor. In several reports, histologic grade and surgical resection margins were the most frequently suggested prognostic factors [5, 26–28]. Besides, other clinical factors, such as histologic subtype and treatment methods, have also been emphasized [29, 30].

On the contrary, our results showed that resection margin, treatment modalities, and histologic grade had no significance for OS. In our cohort, 74.6% of patients who had had an operation as an initial treatment had resectable HNSTS without distant metastasis. As a consistent operation-based treatment protocol was applied in our cohort, prognostic implications of various clinicopathological factors must be validated in the more comprehensive and intensive analysis.

This study has limitations inherent to its retrospective design and a small number of patients enrolled for analysis. Besides, various histologic subtypes were identified in our subjects, which could affect oncologic outcome analysis.

Conclusion

The present study suggests that wide surgical resection of HNSTS, in combination with proper adjuvant treatment, offers acceptable oncologic outcomes of 5-year LRC, DFS, OS.

When making initial treatment planning of this rare tumor, several poor prognosticators such as higher histologic grade, nodal or distant metastasis, and angiosarcoma subtype must be considered to provide adequate treatment extents and guideline for surveillance.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval This article does not contain any studies with human participants performed by any of the authors.

Research involving human participants and/or animals This article does not contain any studies with human participants or animals performed by any of the authors.

Informed consent Consent is not required because of retrospective study. This study is granted exemption from IRB review.

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