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ORIGINAL ARTICLE

Primary oral leiomyosarcoma: a retrospective clinical analysis of 20 cases

B Yan[1], Y Li[1], J Pan[2], H Xia[2], L-J Li[1,2]

[1]*State Key Laboratory of Oral Disease, Sichuan University, Chengdu 610041, China;* [2]*Department of Head and Neck Oncology, West China College of Stomatology, Sichuan University, Chengdu 610041, China*

PURPOSE: As a review and clinical analysis of primary oral leiomyosarcoma (LMS) cases in West China stomatology Hospital in the past 37 years, this study provides demographic, therapeutic and prognostic information of this rare tumor.

PATIENTS AND METHODS: In our study, 20 cases of primary oral LMS treated between 1972 and 2008 in West China Stomatology Hospital were analyzed retrospectively. A thorough review of clinical records was carried out and potential indicators of survival were analyzed.

RESULTS: The most common symptom of oral LMS presented as a painless mass. The median age of patients was 37 years, and the peak incidence age of this tumor was in the 2nd and 5th decades. There was no predilection of gender, and the male-to-female ratio was 11:9. The most frequently occurring site of oral leiomysarcoma was the jawbones. The prognosis of this tumor was poor as a result of the high local recurrence and the estimated 2 year survival was 17.6%. The bony involvement and method of therapy was observed to have an influence on the prognosis and survival of this tumor ($P < 0.05$).

CONCLUSION: There was a predilection site of jawbones for oral LMS, and bony involvement was a potential indicator suggesting a poorer prognosis. The recommended method of therapy on this tumor was aggressive, radical surgical resection; however, adjuvant radiotherapy and chemotherapy may also have a beneficial effect.
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Keywords: primary oral leiomyosarcoma; therapy; prognosis

Correspondence: Li Long-Jiang, State Key Laboratory of Oral Disease, Sichuan University; Department of Head and Neck Oncology, West China College of Stamotology, Sichuan University, Chengdu 610041, China. Tel: 86 28 85503389, Fax: 86 28 85582167, E-mail: muzili63@163.com
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Leiomyosarcoma (LMS) is a malignant neoplasm of smooth muscle origin that most frequently occurs in the uterus, gastrointestinal tract and retroperitoneum. Although the LMS accounts for about 7% of all soft-tissue sarcomas (Fernandez Sanroman *et al*, 1992), the occurrence in the oral and maxillofacial region is considered exceptionally rare, probably because of the paucity of smooth muscle tissue. The prognosis of LMS in the oral and maxillofacial region is usually poor with a high percentage of recurrence and metastasis (Sumida *et al*, 2001; Nikitakis *et al*, 2002), and the estimated 5 year survival for the primary oral LMS is 55% (Ethunandan *et al*, 2007). As a result of the rare occurrence and poor prognosis of LMS, we lack sufficient systematic data and the criteria of therapy are scarce. The purpose of this article is to supply information about this rare tumor to help treatment and improve prognosis by reporting and analyzing 20 cases over the last 37 years, which were retrieved from the files of West China Stomatology Hospital of Sichuan University.

Patients and methods

The study covers all these hospitalized cases of primary LMS in the oral and maxillofacial region at West China Stomatology hospital of Sichuan University between 1972 and 2008, and there were in total 20 cases of primary LMS during the last 37 years. The clinical diagnosis was made following an incisional biopsy in 18 cases and definitive resection in two cases. All these cases were diagnosed by light-microscopic evidence and immunohistochemical demonstration. A thorough review of clinical records was carried. Patient age, gender, clinical manifestation, primary tumor site, size, the status of bony involvement, and regional lymph node, treatment, presence of recurrence and metastasis, follow-up and status of the patient at the last examination were recorded. Through the SPSS 13.0 software-package (SPSS Company, Chicago, IL, USA), an analysis of actuarial survival condition of these cases was considered by using Kaplan–Meier survival estimate and

potential indicators of survival was estimated by Cox's regression analysis to calculate the influence of various demographic and clinical variables on the patient's survival. This study has been approved by our Institutional Review Board. The Declaration of Helsinki protocols were followed during the whole study.

Results

General information

There were 20 cases of primary LMS without distant metastasis treated in our hospital during the last 37 years between 1972 and 2008 (Table 1). The data show that the age range of the patients was from 6 to 63 years with a median age of 37 and a peak incidence in the 2nd and 5th decades that accounts for 55%. The patients consisted of 11 men and nine women and the male-to-female ratio was 11:9. The most frequently involved sites were mandibule, maxilla and cheek, and the floor of mouth accounts for only 5%. The most common clinical presentation was that of non-ulcerated painless mass and the size of the lesion ranged from 1 to 10 cm. Most of tumors presented as firm and non-tender to palpation, and the ones invading bones were not mobile. With the utilization of computed tomography (CT), which was used only in the cases after 1990, and X-ray examination to detect the bony involvement, 16 patients were found to have bony involvement. The radiologic feature presented as a soft-tissue mass occupation usually with an extensive, ill-defined radiolucent destruction in involved jawbones. The clinical palpable lymph node and CT examination were considered as the criteria of identification of enlarged regional lymph node. In these cases, we got eight patients with enlarged lymph nodes, accounting for 40% (Table 2).

Treatment

All the patients received surgical treatment in the form of extensive resection except one patient who refused treatment. And all the margins of resection were found non-infiltrative by pathological manifestation. There were 14 patients with tumors arising in jawbones and five in soft tissues. In the eight patients with tumors in the mandibles, according to the size of lesion, five of them received segmental mandibulectomy, while the other three received hemimandibulectomy. In the other six patients with tumors in maxillas, partial maxillectomy, submaxillectomy, or extensive maxillectomy were performed according to the sites and sizes of lesions. The five patients with tumors occurring in the oral soft tissues were treated by wide local excision (LWE; Table 1). Among the eight patients (40%) with enlarged regional lymph node, only in three of them combined neck dissection was performed with agreement and the others required the intensive supervision for the sake of the postoperative functional defect. In some patients' operative process and some specific anatomic locations, the difficult access to operative region, adjacent vital structures and patients' consideration for postoperative defect could not ensure an enough wide resection and may result in some microscopic residual diseases. So, to avoid the recurrence and metastasis in these patients, postoperative treatment was given. In accordance with this criterion, there were in total eight patients to receive combined postoperative radiotherapy (RT) and/or chemotherapy. The total dose of RT was confined approximately to a range of 40–50Gy through external-beam RT. Cisplatin, docetaxel and fluorouracil were used in the management of chemotherapy.

Follow-up

The time of follow-up covered a wide range from 3 to 53 months except for the three lost cases and the median was 15 months. One patient denied any treatment and died after 3-month follow-up. Only three (17.6%) patients were found to have no local recurrence and distant metastasis in the follow-up period, and their follow-up time ranged from 20 to 53 months. A total of 12 (70.6%) patients suffered local recurrence and 11 patients died of the disease, only one was alive with the disease at the last examination. Metastasis was rare in these cases and only one patient had metastasis to right submandibular lymph node, who died of the disease at the 6th month after first surgical operation.

Statistics

Through the Cox's regression analysis with $P < 0.05$ by the mean of SPSS 13.0, age, gender, size, site, enlarged regional lymph node, recurrence, and metastasis were not potential indicators of survival. However, the analysis suggested that survival was associated with bony involvement ($P = 0.033$) and treatment ($P = 0.004$; Table 3). The estimated 1 year and 2 year survival was 52.9% and 17.6% for the whole group, respectively (Figure 1). The Kaplan–Meier survival estimate revealed that the estimated mean survival times were 16.7 ± 3.9 months for the bony involvement group and 40.3 ± 6.3 months for the soft-tissues group, respectively (Figure 2). Treatment management also had an influence on the prognosis and survival of these cases. The Kaplan–Meirer analysis suggested that the estimated mean survival time of the combined treatment group (surgery combined with RT and/or chemotherapy) was 30.9 ± 6.4 months, which was distinguished from the 10.4 ± 1.7 months of the single treatment (only surgery; Figure 3).

Discussion

The LMS is a malignant tumor of mesenchymal origin exhibiting smooth muscle differentiation, which most frequently occurred in the uterine myometrium, the gastrointestinal tract, and the retroperitoneum. The occurrence of oral LMS is rare as a result of the paucity of smooth muscle tissue in this region and the source of the smooth muscle is derived from primitive mesenchyme and found mainly in the blood vessels, erector pile musculature of skin, circumvallate papilla, primitive mesenchyme and myoepithelial cells of salivary glands (Amarapala and Tilakaratne, 2006). The prognosis of LMS arising in the oral and maxillofacial region is poor and the standard criteria of therapy are not established,

Table 1 Summary of the clinical data collected from the hospitalized cases

No.	*Time*	*Gender*	*Age (years)*	*Site*	*Clinical manifestation*	*Size (cm^2)*	*Bone*	*Lymph node*	*Primary treatment*	*Recur*	*Metastasis*	*Follow-up (months)*	*Status*
1	2007	M	55	L soft palate	Non-ulcerated painless mass	4 × 5	No	Yes	WLE + L mandibular ramus ostectomy + L RND; postoperative chemotherapy	No	No	20	AND
2	2007	F	33	Hard palate	Non-ulcerated painless mass	1 × 1	Yes	No	WLE	Yes	No	7	DFD
3	2007	M	42	R maxilla	Ulcerated pain mass	2 × 2	Yes	Yes	R submaxillectomy	No	Yes	6	DFD
4	2006	F	11	R cheek	Non-ulcerated pain mass	4 × 4	No	No	WLE + partial maxillectomy and mandibulectomy; Postoperative chemotherapy	Yes	No	20	AWD
5	2005	M	40	L mandibule	Non-ulcerated pain mass	4 × 4	Yes	No	L segmental mandibulectomy and free rib graft	INA	INA	LOST	INA
6	2004	M	34	R mandibule	Non-ulcerated pain mass	4 × 2.5	Yes	No	WLE + R segmental mandibulectomy; postoperative chemotherapy and RT	No	No	53	AND
7	2004	F	21	R cheek	Non-ulcerated painless mass	2 × 1	No	No	WLE + R partial maxillectomy; postoperative chemotherapy and RT	No	No	48	AND
8	2003	M	13	L mandibule	Non-ulcerated painless mass	4 × 3.5	Yes	No	L segmental mandibulectomy; postoperative RT	Yes	No	11	DFD
9	1999	M	25	L maxilla	Non-ulcerated painless mass	7 × 4	Yes	Yes	L submaxillectomy and mandibular ramus ostectomy and zygomatic ostectomy	INA	INA	LOST	INA
10	1997	F	16	L cheek	Non-ulcerated painless mass	4 × 4	Yes	No	WLE + L mandibular ramus ostectomy and partial maxillectomy	INA	INA	LOST	INA
11	1996	F	46	R mandibule	Ulcerated painless mass	2 × 1	Yes	Yes	R segmental mandibulectomy; postoperative RT	Yes	No	21	DFD
12	1996	F	63	R maxilla	Non-ulcerated pain mass	6 × 4	Yes	No	R extensive maxillectomy	Yes	No	5	DFD
13	1993	M	48	R floor of mouth	Non-ulcerated painless mass	3 × 3	No	No	WLE + suprahyoid neck dissection	Yes	No	17	DFD
14	1992	F	12	R cheek	Non-ulcerated pain mass	5 × 5	Yes	No	WLE + R submaxillectomy; postoperative RT	Yes	No	8	DFD
15	1992	M	56	R mandibule	Non-ulcerated painless mass	6 × 5	Yes	No	L hemimandibulectomy	Yes	No	15	DFD
16	1984	F	6	L cheek	Non-ulcerated painless mass	10 × 5	Yes	Yes	Refuse treatment	No	No	3	DFD
17	1978	F	47	L mandibule	Ulcerated painless mass	3 × 6	Yes	Yes	L hemimandibulectomy	Yes	No	16	DFD
18	1974	M	19	L mandibule	Ulcerated pain mass	3 × 4	Yes	Yes	L hemimandibulectomy and RND; postoperative RT	Yes	No	35	DFD
19	1974	M	52	L mandibule	Non-ulcerated painless mass	3.5 × 2.5	Yes	No	L segmental mandibulectomy	Yes	No	11	DFD
20	1972	M	49	L maxilla	Non-ulcerated painless mass	6 × 6	Yes	Yes	L extensive maxillectomy	Yes	No	12	DFD

L, left; R, right; WLE, wide local excision; RND, radical neck dissection; RT, radiotherapy; AND, alive with no disease; AWD, alive with this disease; DFD, died of this disease; INA, information not available; LOST, lost during follow-up.

Table 2 Characteristics of the 20 cases of primary oral leiomyosarcoma

Case information		*Patients(%)*
All patients		20 (100)
Age	Range from 6 to 63 years Median 37 year	
Gender	Female	9 (45)
	Male	11 (55)
Tumor Site	Maxilla/palate	6 (30)
	Cheek	5 (25)
	Mandible	8 (40)
	Floor of mouth	1 (5)
Clinical manifestation	Mass without pain	13 (65)
	Mass with pain	7 (35)
	Non-ulcerated	16 (80)
	Ulcerated	4 (20)
Bone involvement	Soft tissue only	4 (20)
	Bone involvement	16 (80)
Regional lymph node	Enlarged lymph node	8 (40)
	Without enlarged lymph node	12 (60)
Treatment	Surgery	11 (55)
	Surgery combined with chemotherapy and/or radiotherapy	8 (40)
	Untreated	1 (5)
Recurrence		12 (70.6)
Metastasis		1 (5.9)

Table 3 Potential indicators of survival (Cox's analysis)

Parameter	*RR (relative risk)*	P-*value*	*95% Confidence interval*
Bony involvement	11.33	0.033	1.22–105.43
Treatment	0.045	0.004	0.01–0.37

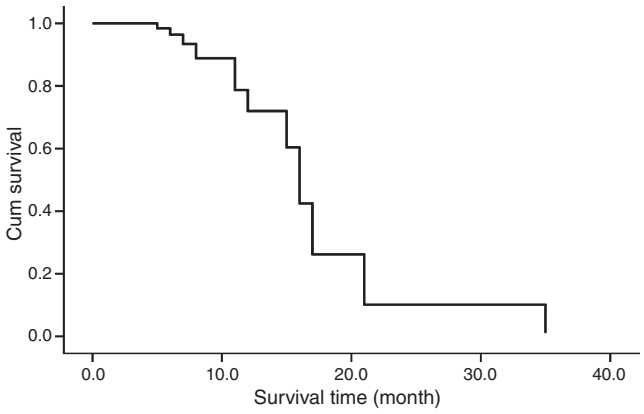

Figure 1 A survival curve by Cox's regression for the whole group from the time of treatment

although the initial complete excision with tumor-free margins is recommended (Fernandez Sanroman *et al*, 1992; Akcam *et al*, 2005; Amarapala and Tilakaratne,2006; Ries Centeno *et al*, 2006; Ethunandan *et al*, 2007). We provide demographic, clinical and therapeutic information of the LMS in this study to reflect the status in West China.

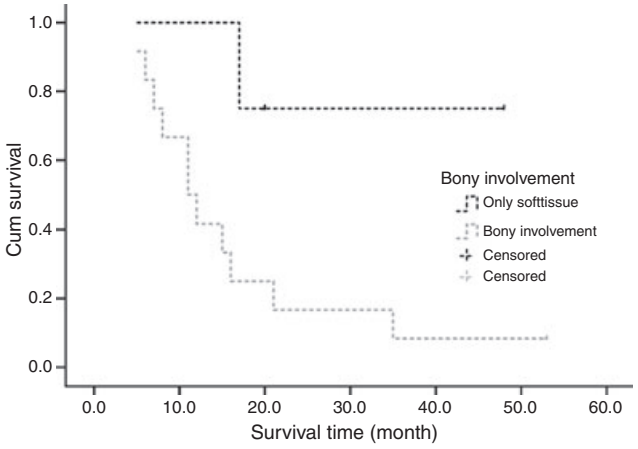

Figure 2 Kaplan–Meier estimated survival for bony involvement *vs* only soft-tissue involvement

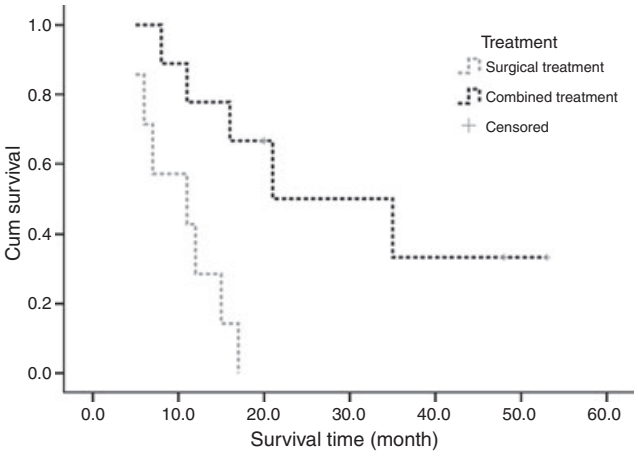

Figure 3 Kaplan–Meier estimated survival for surgical treatment *vs* combined treatment

The LMS is a rare tumor in the oral and maxillofacial region and generally has non-specific sign and symptoms, which usually presents as a non-ulcerated painless mass. As a result of the lack of distinguishing clinical features and the rarity of these lesions in the oral and maxillofacial region, some lesions of LMS are occasionally mistaken for the other common lesions affecting the oral cavity, and correct diagnosis is made only following definitive histologic examination (Amarapala and Tilakaratne, 2006; Ethunandan *et al*, 2007). In this study, a painless mass is the most frequently presented oral LMS, which accounts for 65% in the whole group and this findings is similar to those cases reported in the literature (Montgomery *et al*, 2002; Amarapala and Tilakaratne,2006; Ethunandan *et al*, 2007). The median age in our cases is 37 years and the range of age covers from 6 to 63 years with a peak incidence in both the 2nd and 5th decades which differs from what is reported in the literature (Schenberg *et al*, 1993; Izumi *et al*, 1995; Vilos *et al*, 2005; Ethunandan *et al*, 2007). Gender predilection was not found in our study and there are 11 males and nine females (M:F = 11:9). The size of the

primary lesion varied from 10 to 1 cm. The most frequent sites of the tumor in this study are mandibule and maxilla, all of which arose in the jawbones. The other sites are the cheek, soft palate and floor of mouth. These findings suggest that there is a site predilection in the jawbones for oral primary LMS and an aggressive property. Vilos *et al* (2005) reported that the jawbones appear to be the site of predilection for oral LMS and approximately 70% of these tumors arise in the maxilla or mandibule. Schenberg *et al* (1993) and Izumi *et al* (1995) also suggested that the jawbones were the most frequent site in oral LMS.

Bony involvement did seem to be related to survival and was considered as one of the potential indicators of survival because there was a statistical significant difference in this study through Cox's regression analysis ($P = 0.033 < 0.05$). There were 16 (80%) patients with bony involvement in our group and 10 of them suffered local recurrence which accounts for 83.3% in the local recurrence group. In the bony involvement group, only one patient suffered metastasis to the right submandibular lympha node but distant metastasis was not discovered during the follow-up period. In Kaplan–Meier survival estimate, the mean survival time of the group with bony involvement was approximately 14 months shorter than the group without bone involvement (Figure 2). These findings are similar to the report by Ethunandan *et al* (2007).

Among these patients, eight (40%) had enlarged regional lymph node and only one case (5%) was identified as metastasis to the right submandibular lymph node without distant metastasis. In the previous literatures, it was reported that metastasis to regional lymph nodes was relatively rare for oral LMS and the most common site suffering metastasis was the lungs (Weitzner, 1980; Nishi *et al*, 1987; Schenberg *et al*, 1993; Izumi *et al*, 1995; Yang *et al*, 2006). Among the eight patients with enlarged lymph node, three patients received radical neck dissection (RND) or suprahyoid neck dissection, of whom only one was found suffering regional lymph node metastasis while the histopathologic examinations of others showed reactive hyperplasia of lymph nodes. The ratio of lymphatic metastasis was only 12.5% and far lower than the local recurrence, but despite these results and for the sake of prognosis, we completely agree with the opinion that RND may be required if enlarged cervical lymph nodes are evident on clinical examination (Izumi *et al*, 1995; Crossman *et al*, 2008) because the success of the initial surgical management seemed to be an important prognostic factor (Amarapala and Tilakaratne, 2006).

According to the literature review, as to the best option of the principal treatment methods, LWE or complete surgical resection with tumor-free margins is recommended to control local recurrence (Goldschmidt *et al*, 1999; Montgomery *et al*, 2002; Akcam *et al*, 2005; Ries Centeno *et al*, 2006; Yang *et al*, 2006; Ethunandan *et al*, 2007; Rodini *et al*, 2007; Crossman *et al*, 2008; Misra *et al*, 2008), and adjuvant RT or chemotherapy is considered to have little beneficial effect on decreasing recurrence of LMS or increasing survival time (Schenberg *et al*, 1993; Ries Centeno *et al*, 2006; Yang *et al*, 2006; Ethunandan *et al*, 2007; Rodini *et al*, 2007). However, in some specific anatomic locations such as the vicinity of the infratemporal fossa, the maxillary sinus, the pterygoid plates and the mandibular condyle, it may be difficult and technically less feasible to perform an operation reaching the requirement of a LWE with tumor-free margins because of difficult access, adjacent major and vital vascular structures, and consideration of postoperative defect reconstruction, possibly resulting in residual microscopic disease leading to the local recurrence of the tumor and a poorer prognosis (Izumi *et al*, 1995; Rapidis *et al*, 2005; Vilos *et al*, 2005). Postoperative therapy is considered as an adjuvant method to improve recurrence and survival rates as a result of the beneficial effect on the treatment of microscopic focuses (Fernandez Sanroman *et al*, 1992; Schenberg *et al*, 1993). Akcam *et al* (2005) emphasize the role of adjuvant RT in reducing the risk of recurrence of surgically treated head and neck of LMS even without tumor positive margins and state that radiation therapy may be necessary after surgery. The value of adjuvant chemotherapy for the treatment of oral LMS is still ambiguous and chemotherapy is most often considered as a palliative modality for inoperable patients (Nishi *et al*, 1987; Schenberg *et al*, 1993; Dry *et al*, 2000). However, Mitsudo *et al* (2006) reported effective chemotherapy on the treatment of maxillary LMS with docetaxel and cisplatin using superselective intra-arterial infusion via superficial temporal artery. In our study, eight patients underwent surgery combined with RT and/or chemotherapy and the other 11 only underwent surgery. Four of the eight patients with combined therapy died of the disease during follow-up, and eight of the 11 patients with surgery alone died except the three lost patients, which accounted for 66.7% of the dead patients who received treatment. Although there was a significant difference of actuarial survival time between the two groups and the postoperative-treated group appeared to have a better outcome (Figure 3), it is still argued that RT and chemotherapy might be beneficial to enhance the effect of surgery of oral LMS and improve the survival time and quality of life because of the absence of sufficient cases and no standardized treatment. Recommendation with postoperative treatment such as RT and chemotherapy should be cautiously given because we need more cases and a further study to evaluate the treatment of oral LMS.

In conclusion, LMS is an exceedingly rare tumor in the oral and maxillofacial region and has a poor prognosis as a result of high local recurrence. We provide information of 20 cases to assist the treatment and prognosis of this tumor. There may be a predilection of site in the jawbones and bony involvement may indicate a poorer prognosis. Aggressive surgical treatment with LWE is necessary for radical cure, and the adjuvant RT and chemotherapy may also have a beneficial effect in terms of reduced recurrence, improved survival time and the possibility of less radical resection.

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Author contributions

Long-jiang Li is the guarantor of the integrity of the study and study design. Bing Yan and Yi Li contributed in clinical data acquisition, statistical analysis and manuscript preparation. Jian Pan and Hui Xia contributed in literature research and manuscript editing.

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