

**Table 1** Clinical data (Rec recurrence, Met metastasis, Pr primary, Tmf temporomandibular fossa, Smr submandibular region, S surgery, R radiation therapy, C chemotherapy, UR primary site of tumor unresectable, L regional lymph node metastasis, D distant metastasis, Ao alive without disease, Ac alive with disease, Dc died of disease)

Case no.	Diagnosis	Case	Sex	Age (years)	Site	Treatment	Rec	Met	Follow-up	Status
1	Osteosarcoma	Pr	M	25	Tmf	S+R+C	—	—	7 years 1 month	Ao
2	Osteosarcoma	Pr	F	34	Maxilla	S	—	—	9 years 6 months	Ao
3	Osteosarcoma	Pr	F	40	Mandible	S	+	—	6 years 4 months	Ao
4	Osteosarcoma	Pr	M	46	Mandible	S	—	D	5 years	Dc
5	Osteosarcoma	Pr	M	50	Mandible	S	+	D	13 years 7 months	Ac
6	Osteosarcoma	Pr	M	50	Maxilla	S+C	—	—	3 years 2 months	Ao
7	Osteosarcoma	Rec	M	56	Maxillary sinus	S+R+C	+	—	4 years 1 month	Dc
8	Osteosarcoma	Rec	F	63	Maxilla	S+R+C	—	D	3 years 3 months	Dc
9	Osteosarcoma	Rec	M	67	Mandible	S	—	—	9 years 3 months	Ao
10	MFH	Pr	M	29	Maxillary sinus	S+C	+	—	1 year 4 months	Dc
11	MFH	Pr	M	43	Mandible	S	+	L	2 years	Dc
12	MFH	Pr	M	45	Smr	S+R	—	—	8 years 1 month	Ao
13	MFH	Rec	M	48	Mandible	S	—	—	4 years 6 months	Ao
14	MFH	Pr	M	56	Maxilla	S+R+C	—	—	11 years 8 months	Ao
15	MFH	Pr	M	57	Maxillary sinus	S	+	L	9 months	Dc
16	MFH	Pr	M	63	Maxillary sinus	S+R+C	—	—	5 years	Ao
17	Rhabdomyosarcoma	Pr	M	11	Buccal mucosa	S	+	—	17 years 3 months	Ao
18	Rhabdomyosarcoma	Pr	F	27	Buccal mucosa	S+R+C	—	D	1 year 6 months	Dc
19	Rhabdomyosarcoma	Pr	M	55	Maxilla	S+C	—	D	2 years 3 months	Dc
20	Rhabdomyosarcoma	Pr	M	58	Maxilla	C+R	UR	L+D	4 months	Dc
21	Rhabdomyosarcoma	Pr	M	77	Mandible	S	—	—	2 years 9 months	Ao
22	Fibrosarcoma	Pr	F	5m	Mandible	S+C	—	—	9 years 9 months	Ao
23	Fibrosarcoma	Pr	F	10	Mandible	S	—	—	11 years 6 months	Ao
24	Fibrosarcoma	Pr	F	16	Mandible	S	—	—	19 years	Ao
25	Plasmacytoma	Pr	M	53	Mandible	S+R	—	—	14 years	Ao
26	Plasmacytoma	Pr	M	59	Maxillary sinus	S+R	—	—	5 years 1 month	Ao
27	Leiomyosarcoma	Rec	M	25	Maxilla	C+R	UR	L+D	2 months	Dc
28	Leiomyosarcoma	Pr	M	43	Mandible	S	—	D	8 years	Ac
29	Angiosarcoma	Pr	M	34	Tmf	C+R	UR	—	4 years	Dc
30	Angiosarcoma	Pr	M	53	Maxilla	S+R	+	L	8 months	Dc
31	Liposarcoma	Pr	F	33	Buccal mucosa	S	+	—	2 years 10 months	Dc
32	Ameloblastic fibrosarcoma	Rec	M	31	Mandible	S	+	—	5 months	Dc

The most common histological type was osteosarcoma ( $n=9$ ; 28%), followed by malignant fibrous histiocytoma (MFH; ( $n=7$ , 22%), rhabdomyosarcoma ( $n=5$ , 16%), fibrosarcoma ( $n=3$ , 9%), plasmacytoma ( $n=2$ , 6%), leiomyosarcoma ( $n=2$ , 6%), angiosarcoma ( $n=2$ , 6%), and liposarcoma ( $n=1$ , 3%), ameloblastic fibrosarcoma ( $n=1$ , 3%). The histological subtypes of osteosarcoma were chondroblastic ( $n=5$ ; Table 1, nos. 1, 2, 6, 8, 9), osteoblastic ( $n=3$ , nos. 3, 4, 7), and telangiectatic ( $n=1$ , no. 5). In rhabdomyosarcoma, the histological subtypes were alveolar ( $n=3$ , nos. 17, 18, 20) and pleomorphic ( $n=2$ , nos. 19, 21). Patients with malignant lymphoma were excluded from this study because malignant lymphoma is a systemic disease. Two cases of plasmacytoma were both of the solitary type, therefore these cases were included.

Surgical resection (S) alone was carried out in 15 patients. Triple modality of S, radiation therapy (R) and chemotherapy (C) was performed in 6, combined therapy of S/C in 4, and S/R was in 4. We judged that the primary tumors were unresectable in three cases; therefore combined therapy of R and C was given to these 3 patients. Radiation therapy was given preoperatively in 6 cases, postoperatively in 3 and both in 1. The doses used varied from 18 to 80 Gy, with an average of 48 Gy. All radiation therapy was external irradiation. The fraction sizes were 2.5 Gy, treating 4 days in a week. Chemotherapy was given preoperatively in 8 cases and postoperatively in 2. The chemotherapy regimens were varied.

Treatment was determined according to the experience of the surgeon. This resulted in a lack of unity in the treatment protocol. The common agents were cisplatin ( $n=5$ ), 5-fluorouracil ( $n=4$ ), and doxorubicin ( $n=4$ ). The regional intra-arterial infusion chemotherapy administered via the superficial temporal artery was given in 9 of 11 patients who had sarcoma of the maxilla, maxillary sinus, or

the temporomandibular fossa. The survival rate and survival curve were calculated by the Kaplan-Meier method using SPSS for Windows, version 10.

## Results

Fourteen patients of 32 were making satisfactory progress without local recurrence or metastasis at a mean follow-up period of 8.5 years, ranging from 2 years 3 months to 19 years. Local recurrence was found in 10 cases (31%) and metastasis in 11 (34%). Among 10 patients with local recurrence, 3 are alive after 6–17 years, and 7 died of their tumors after 5 months to 4 years. Among the 3 patients alive, one (no. 3) was treated by surgery and the other (no. 17) by surgery and postoperative radiation therapy (50 Gy) after local recurrence was found. These two patients are alive without evidence of disease over 3 years and 7 years after salvage treatment. Another patient (no. 5) was treated by preoperative radiation therapy (45 Gy) and surgery for local recurrence. The local recurrence was controlled, but this patient developed distant metastasis to the lung.

The 5-year survival rate of patient with local recurrence was 30% (Fig. 1). Among 11 patients with metastases 9 died