

# Leiomyosarcoma of the Base of the Tongue Treated with Radiotherapy: a Case Report

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**Soft tissue sarcomas of the oral cavity are uncommon malignancies; those of smooth-muscle origin are extremely rare. The world literature reveals about 80 leiomyomas and 26 cases of leiomyosarcoma. Only 4 of the cases with leiomyosarcoma occurred in the tongue. This report of leiomyosarcoma of the base and dorsum of the tongue adds 1 case which was primarily treated with irradiation and achieved an excellent response. A 1.5-year follow-up after radiation-therapy is included and a review of the literature is presented.**

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## INTRODUCTION

SARCOMAS ARE malignant tumours derived from mesodermal tissue. Soft tissue sarcomas comprise approximately 0.7% of all malignant neoplasms and of these only 2-8% are leiomyosarcomas. Leiomyosarcomas arise most commonly in the gastrointestinal and female genital tract [14, 16]. Other origins such as viscera, major arteries, veins and the extremities are less frequent sites of origin. Generally, soft tissue sarcomas aggressively invade the surrounding tissues and metastasise hematogenously, most frequently to the lung. Principally treatment is by an excision with sufficient tumour free borders and postoperative irradiation when necessary [16, 19]. The leiomyosarcoma is accepted to have a higher propensity to metastasise than fibrosarcoma or liposarcoma. The location of a leiomyosarcoma in the oral cavity is an extremely rare event.

## CASE REPORT

A 70-year-old man, who had progressive dysphagia for a few months, loss of weight and nasal speech was seen at the Department of Otorhinolaryngology. He could only swallow fluids and was only able to sleep sitting up. On physical examination a huge exophytic tumour was seen on the back portion of the mobile tongue, which was filling the lumen of oropharynx. The tumour was also involving the base of the tongue up to the level of Vallecula epiglottica. On CT-scan, an infiltration of the body of tongue (M. genioglossus) was found as well as a lymph node metastasis on the right side of the neck (Figs 1, 2). The patient used to smoke about 15 cigarettes daily and consumed 2 litres of beer daily over 30 years. The major laboratory parameters including full blood count, differential, electrophoresis of serum proteins, sedimentation rate and

tumour markers SCC and CEA were normal. X-ray of the lung showed no abnormality. A barium study of oesophagus, stomach and duodenum was normal.

### *Microscopic appearance*

Two biopsies of about 1 × 0.5 × 0.3 cm were taken from the posterior portion of the tongue. Histopathologically a solid tumour was seen which was rich in cells and mitoses and composed of atypical and mainly spindle-shaped cells and leiomyocytes which had moderate pleomorphic and hyperchromatic nuclei (Fig. 1a). The immunohistochemical studies gave the same result; the antibody reaction for cytoskeletal vimentin (V9, Dakopatts, Hamburg, Germany) and for actin (HHF-35, Dakopatts) were positive (Fig. 1b,c). The antibody reactions for cytoskeletal keratin (KL1, Dianova, Hamburg, Germany), for vessel endothelial antigen BMA 120 (Behring-Werke, Marburg, Germany) and for S-100 antigen (Dakopatts) were negative. At the surface of the tumour there was a superficial ulcer and a leucocyte-containing fibrin layer.

### *Radiological appearance*

CT-scan before treatment (Fig. 1a,b) showed a large, exophytic tumour of about 4 cm diameter on the dorsum of the tongue, which had more density (70 HU) than the musculature on contrast scan. One lymph node was seen at the level of the jugular vein junction, about 1.5 cm in diameter on the right side of the neck. The involved lymph node was homogenous (unlike squamous carcinoma) and had the same density in Hounsfield units as the main tumour. The tumour was staged T4-N1-M0, and was considered unresectable, especially because of the age and poor general condition of the patient.

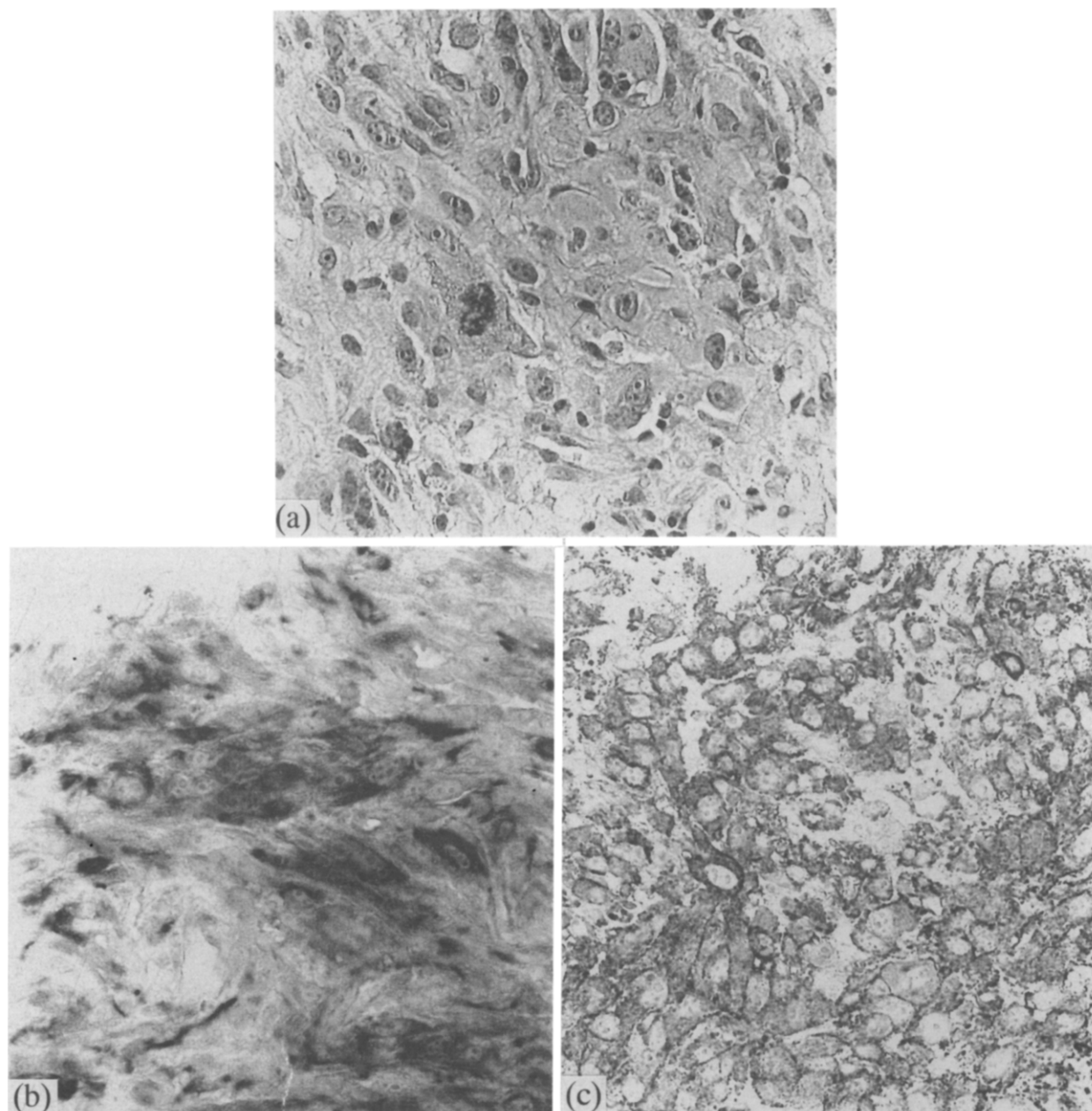
### *Treatment*

A dose of 50 Gy was delivered with 18 MV photons, using lateral-opposing portals. In the first week, radiation treatment was applied in a sitting position, because the patient was not

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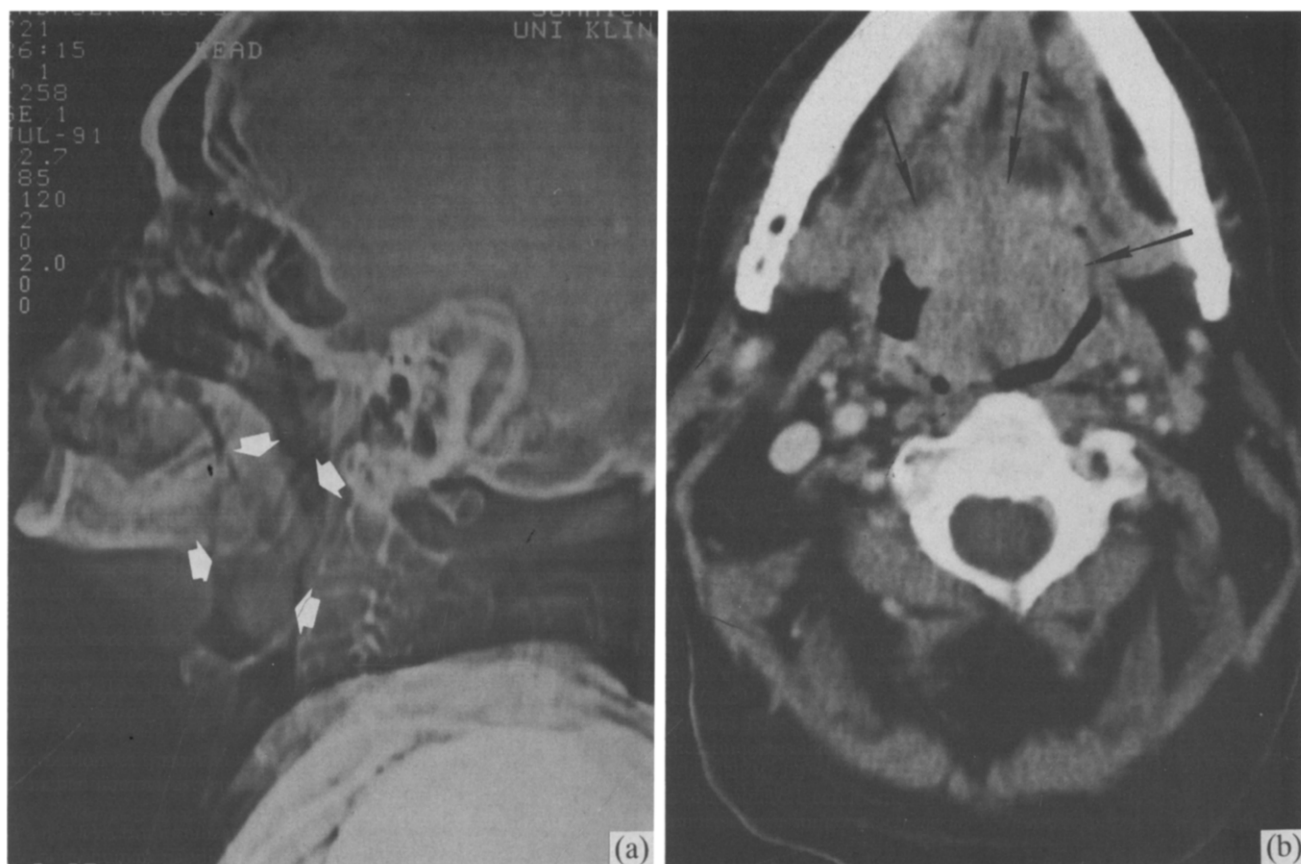


**Fig. 1. (a) Strongly pleomorphic tumour cells with polygonic borders of nuclei and obvious nucleoli. Haematoxylin and eosin, magnification 400 $\times$ . (b) Dark cells of leiomyosarcoma due to positive immunohistochemical reaction against vimentin antibody V9 APAAP, 400 $\times$ . (c) Cells of leiomyosarcoma which show a strong and uniform positive immunohistochemical reaction of their cytoplasm against aktin antibody. APAAP, 400 $\times$ .**

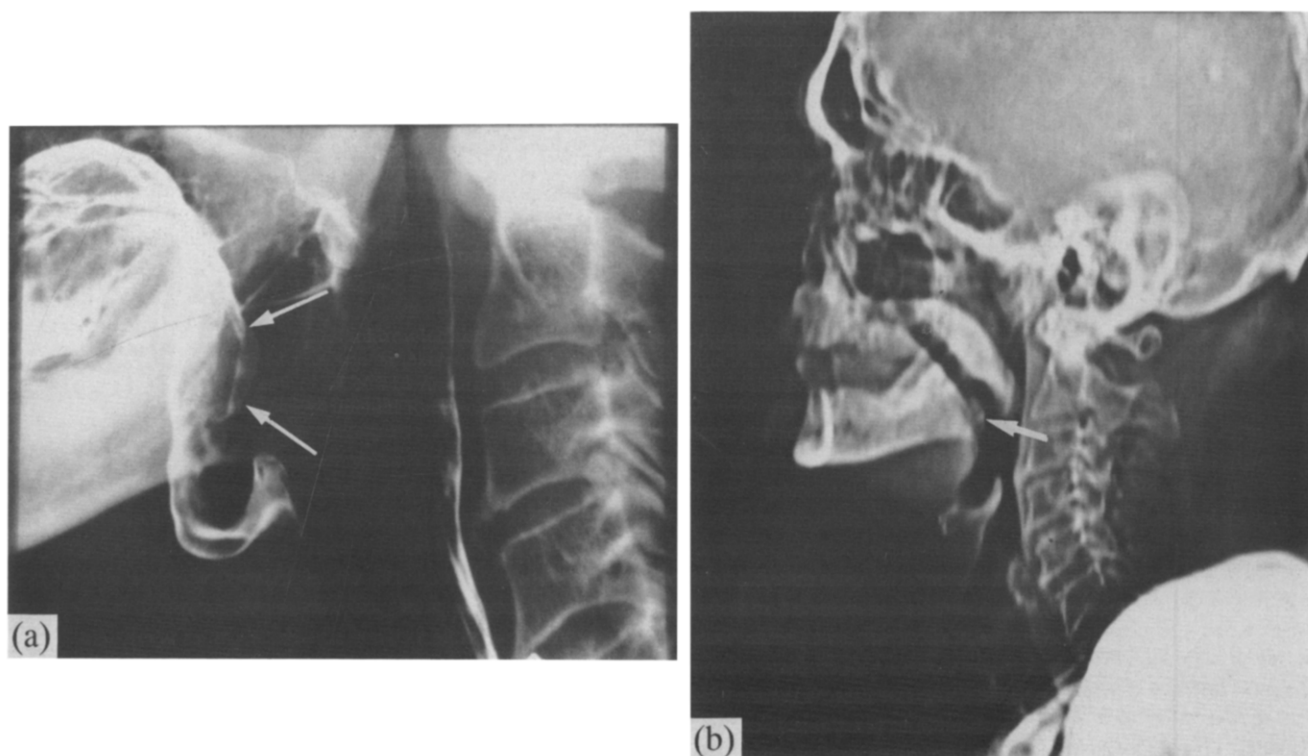
able to breathe sufficiently in a supine position. The tumour regressed excellently during this therapy (Fig. 2a,b) therefore the treatment was continued with a curative aim, after a reduction of the target volume up to 64 Gy, using 18 MV photons (Fig. 3a). Then after a further reduction of target volume the remaining tumour (about 1.5 cm in diameter) was irradiated up to a complete dose of 72 Gy in a rotational-beam technic, again with 18 MV photons on linear accelerator (Fig. 3b).

The radiotherapy was tolerated very well and the patient

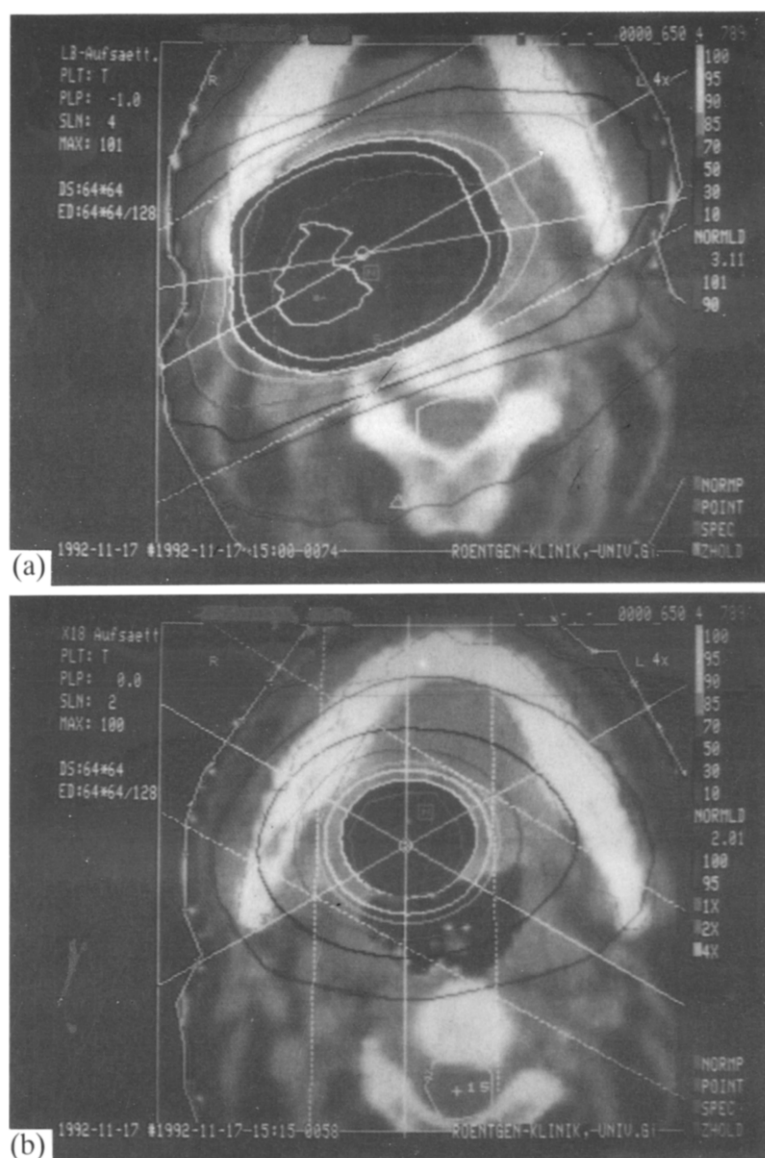
became able to breathe, eat, drink and speak normally. He gained 5 kg in weight during radiation therapy. There was only a very moderate radiation-induced mucositis. Eight weeks after the completion of the irradiation, biopsies were taken from the suspicious region at the base of former tumour site (a slightly indurated region, 1 cm in diameter was palpated base of tongue). They were free of tumour. Now, 1.5 years after finishing the treatment the patient is feeling well without evidence of local recurrence or distance metastasis. The CT-scan shows a complete remission of the tumour (Fig. 4).



**Fig. 2.** (a) CT-scan before treatment; on lateral scannogram a large soft tissue mass is seen on the rear portion of the tongue and base of the tongue (arrows). (b) CT-scan before treatment; exophytic tumour  $4 \times 5 \times 6$  cm, at the dorsum and base of the tongue (arrows). It has invaded the floor of the mouth and obliterates the lumen of mesopharynx. The tumour has in contrast scan a higher density (70 HU) than musculature (55 HU) due to good vascularisation.



**Fig. 3.** (a) Barium swallow X-ray film in lateral view shows an excellent response to radiotherapy after 40.0 Gy. There is a residual tumour in the base of the tongue, 2.5 cm in diameter (white arrows). (b) CT lateral scout view after 50.0 Gy shows only a small residual tumour of 1.5 cm diameter at the same location (white arrow).



**Fig. 4. (a) Reduced treatment volume after 50 Gy. A dose of  $7 \times 2$  Gy at 90% isodose (up to total 64 Gy) is applied with 18 MV photons on a three field technique (two opposing portals and an arc field from 240 to 80 degrees). Field size  $6 \times 6$  of all three fields. Dose calculation was made at levels of 0, -2 cm and +2 cm, but shown here only in the main level (middle). (b) The second shrinkage of the treatment volume. Now  $4 \times 2$  Gy in 95% isodose is given with 18 MV photons in arc technic with field size  $4 \times 4$  cm. A total dose of 72 Gy is herewith given to the root of tumour region. Notice the excellent protection of the skin, mandibula and salivary glands with this technique.**

## DISCUSSION

Since there is only striated musculature in the oral cavity, a smooth muscle tumour can only originate from the smooth muscles of the walls of vessels or the circumvallate papillae of the tongue [2]. In general, the treatment of choice for a localised adult soft tissue sarcoma is the operative resection and postoperative irradiation [16, 19]. The value of chemotherapy for treatment of leiomyosarcoma is not yet established [16–19]. The extent of the surgical approach depends on many factors, such as the site, stage, histopathological type and grading of the tumours as well as the host-specific factors such as the age and the general condition of the patient. The

advanced tumours of the head and neck, especially those with involvement of the base of the tongue, are principally unresectable irrespective of the histo-pathological type of the tumour. Local tumour control is the most important objective whether or not there is evidence of distant metastases. The common opinion of many oncologists, that sarcomas are principally “radiation-resistant” cannot be accepted. Undifferentiated sarcomas especially, as in this case, may prove to have an excellent radiosensitivity. When the tumour is inoperable, for a variety of reasons, local control is the main problem and radiation therapy is the treatment of choice.



**Fig. 5. CT-scan 15 months after completing the radiotherapy: a complete response is seen with no evidence of tumour or lymphatic involvement.**

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