Liposarcoma of the floor of the mouth:
A case report

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Liposarcoma of the floor of the mouth is an exceptionally rare disease entity; only 5 cases have been described in the literature. We herein report a 35-year-old-woman with diagnosis of myxoid type liposarcoma of floor of the mouth. Following initial surgery, the patient developed a massive local recurrence within six months, for which palliative radiotherapy was attempted. [Turk J Cancer 2002;32(2):69-74]

Key words: Liposarcoma, floor of the mouth

Liposarcoma was first described in 1857 by Rudolph Virchow (1). It is one of the most common soft tissue sarcomas of the adulthood. It constitutes 15% of all soft tissue sarcomas (2). Although all body regions may be involved, the vast majority of liposarcomas are found in the lower extremities and retroperitoneum. Despite being one of the most common soft tissue sarcoma, liposarcoma is rarely found in the head and neck region, where less than 100 cases have been reported in the literature (3). The soft tissues of the neck and orbit are the most common primary sites for the liposarcoma of the head and neck region (4-5).

Case Report

A 35-year-old-woman presented with complaints of swelling in the mouth, excessive salivation, difficulty in eating and speaking of 3 months' duration. On clinical examination, a 4x5 cm, non-tender, lobulated swelling was found in the left half of the floor of the mouth. The swelling was also extending to the under surface of the oral tongue and pushing it to the opposite side due to mass effect (Figure 1). Extensive induration around the lesion was also noted. Multiple small, mobile, non-tender neck nodes from level I to III were present on the left side. Her systemic examination did not reveal abnormal clinical findings. Routine investigations, including chest X-ray, complete blood counts & biochemical profile were normal. Orthopantomogram (OPG) did not show involvement of the mandible radiologically. She was started on Ryle’s tube feeding. A punch biopsy of the lesion revealed features of myxoid type liposarcoma (Figure 2A,B,C).
Fig 1. Lesion in the floor of mouth at initial diagnosis

Fig 2. (A): Scanner view showing liposarcoma along with normal tissue of undersurface of the tongue (H&E, x4), (B): Low power view showing typical vascular and myxoid pattern (H&E, x10), (C): High power view showing myxoid liposarcoma, consisting of lipoblasts in varying stages of differentiation and an abundance of myxoid material between tumor cells (H&E, x100)

Immunohistochemistry revealed that tumor cells were positive for Vimentin & S-100 protein but negative for Cytokeratin (CK), AE1 and epithelial membrane antigen (EMA). Based on the microscopic examination and supported by
immunohistochemical markers, a final diagnosis of myxoid type liposarcoma was established. Surgical exploration was performed. Wide excision of the floor of the mouth, left hemiglossectomy and hemimandibulectomy along with left sided radical neck dissection were performed. Tracheostomy was also done. Macroscopic examination of the resected specimen showed a large submucosal growth in the floor of the mouth having an irregular white surface and firm in consistency. The growth was 4.5x3.5x2.0 cm in size. It was also involving muscles of under surface of the tongue. The growth was extending up to the mandible but not infiltrating the bone. Submandibular salivary gland was also normal. Base of the resection was infiltrated by the growth. Lateral, anterior, posterior, and medial mucosal margins of the resection were 1.0, 1.0, 1.0, and 2.0 cm away, and free from the involvement by malignant cells. Focal areas of hemorrhage, necrosis and chronic inflammation were evident. No lymphatic or vascular permeation was noted. All the resected nodes were negative for metastatic disease. The patient was advised post-operative radiotherapy, but she refused, and subsequently she was lost to follow-up. Six months later, she was again reported with painful swelling at the operated area and difficulty in taking food orally. A massive lesion at the operated site was noted (Figure 3).

![Figure 3. Massive recurrence on left side of face](image)

On CT scan examination, a big, mixed density, complex mass was reported at the primary site which was also extending and involving oropharynx, maxilla, and infratemporal fossa (Figure 4). The mass was also close to the blood vessels in the neck region. Areas of hemorrhages & necrosis were also evident. Her metastatic work-up was negative. In view of her locally advanced and inoperable disease, a course of palliative external radiotherapy was started on tele-Cobalt machine. A total dose of 4500 CGy/ 15 fractions/ 3 weeks was
delivered. Following radiotherapy, there was subjective improvement in her symptoms but only partial regression of the lesion was noted. The patient did not come for further follow-up.

Fig 4. CT scan of the head (axial and coronal sections) showing massive recurrence at the operated site

Discussion

Intraoral liposarcomas are extremely rare. Gagari et al. (6) reviewed the literature and found that till 2000, only 45 cases had been reported in the literature. A search of the literature revealed that further four cases were reported as single case reports, thus making a total of 49 cases (7-9). Among the various sub-sites in the oral cavity, cheek is the most frequently reported site of involvement followed by oral tongue, floor of mouth, soft palate, mandible, gingiva, upper lip, etc. in decreasing order of frequency (Table 1).

The first case of liposarcoma of the floor of mouth was described by Enterline et al (10) in 1960, and until now, only five cases have been reported in the literature (9-13). The present patient is only 6th case of liposarcoma of the floor of the mouth in the literature and the first being reported from India.

Liposarcoma arises from fatty tissues. The relative lack of these tissues in the head and neck reflects the rarity of finding liposarcoma in these areas, while accounting for their relative abundance in the lower extremities and retroperitoneum. It should be noted that there is relative abundance of adipose tissue in those sites of the head & neck region (i.e., neck, orbit, and cheek, etc.) where majority of the head & neck liposarcomas have been reported.

Enzinger and Winslow (14) proposed the first classification of liposarcoma, currently used by the World Health Organization, grouping histologically into four types, e.g. myxoid, well differentiated, round cell, and pleomorphic. The well differentiated liposarcoma can be divided into several subtypes, including lipoma like, inflammatory, sclerosing and de-differentiated type. Myxoid liposarcoma is the most common type and accounts for approximately 40-50% of all liposarcomas (14,15). Majority of the head and neck liposarcomas are myxoid type. Of the five previously reported cases of liposarcoma of floor of
mouth, majority of the patients had this histologic sub-type. Our patient also had myxoid histology. Liposarcoma has no predilection for any particular race or geographic region. It is most often seen in middle aged patients with slightly higher incidence in men. Yueh et al. (3) reviewed the literature on reported cases of head and neck liposarcoma and observed a significant male predominance (67%), and seen in all age groups from infancy to 88 years, but most commonly found in 5th and 6th decades. Our patient was 35-year-old woman. Surprisingly, most of the liposarcomas involving the floor of the mouth region have been reported in women. Surgery remains the mainstay of the treatment. Many liposarcomas appear to be well circumscribed or encapsulated. However, they actually spread relentlessly into adjacent tissues and their satellite nodules may be missed and left behind at the time of excision. Despite aggressive surgery, 50 to 70% recur locally (15,16). Enzinger and Winslow (14) reported recurrence rates of 53% for well differentiated and myxoid liposarcoma, while 73% for pleomorphic and 85% for round cell type. Lymph node metastasis is rare, thus, lymphadenectomy is not indicated unless there is evidence of involvement of lymph nodes (17). Metastases are more commonly hematogenous (10). Several studies have shown that adjuvant radiotherapy reduces local recurrence after surgery (10,18,19). Adjuvant chemotherapy has not been proved to be of value (17). It is reported that highest 5 year survival rate was found in patients with well differentiated and myxoid varieties and the lowest in those with round cell type of liposarcoma (7,16). In conclusion, liposarcoma of the floor of mouth is an exceptionally rare malignant lesion with poor prognosis. However, in order to reduce the risk of local recurrence, adjuvant post-operative radiotherapy should be considered in initial management of these tumors.

Table 1
Intra-oral cases of liposarcoma classified according to location

<table>
<thead>
<tr>
<th>Intra-oral location</th>
<th>Number of cases</th>
</tr>
</thead>
<tbody>
<tr>
<td>Cheek</td>
<td>24</td>
</tr>
<tr>
<td>Tongue</td>
<td>12</td>
</tr>
<tr>
<td>Floor of Mouth</td>
<td>6</td>
</tr>
<tr>
<td>Palate</td>
<td>3</td>
</tr>
<tr>
<td>Mandible</td>
<td>2</td>
</tr>
<tr>
<td>Gingiva</td>
<td>2</td>
</tr>
<tr>
<td>Upper Lip</td>
<td>1</td>
</tr>
<tr>
<td>Total</td>
<td>50</td>
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References