Sarcomas, or tumors derived from the connective-tissue elements, are rare among the malignancies of the lip. The only available statistics as to sarcoma of the lip are from the Göttingen Pathological Institute, where 2 cases were found among 1,248 sarcoma cases (0.16 per cent). No mention was made as to whether the lesions were on the vermillion border, and the type of sarcoma is not specified. Gurlt found two cases of lip sarcoma, one on the upper lip and one on the lower lip, among 848 sarcoma cases (0.24 per cent).

In the Stuyvesant Square Hospital 4 cases, all involving the vermillion border, were found among 111 sarcoma cases (3.5 per cent), and among 898 affections of the lip, of which 429 were malignant. One of these involved the upper lip and three the lower lip. Two of these cases were mentioned in an article by Dr. Robert H. Kennedy in a report on epithelioma of the lip (Ann. Surg. 91: 81, 1934).

In the available literature we have found 20 cases described, quoted, or mentioned. Unfortunately in all but 5 of these the records are incomplete, either from a clinical or pathological point of view. Two of the 20 cases involved the upper lip; the others all developed on the lower lip.

The patients were of all ages, but most of them were advanced in years. One case of spindle-cell sarcoma of the lower lip was described in a child of a year and a half, while 3 patients were over seventy. Women predominated in the reported cases, but in our series there were 3 men and only one woman. The duration of the disease before the patient came for treatment varied from a few weeks to a few months. Many of the tumors arose spontaneously, but there was a history of injury (chemical) in one case.

Sarcoma of the lip appears as a new growth, varying in size from a few millimeters to several centimeters in diameter, rarely involving the entire lip. Its color is reddish blue or red. The surface is smooth, more or less lobulated, presenting very soon after its appearance an excoriation, bleeding easily. This becomes ulcerated and punched out in the center, with hard, indurated, infiltrated, ill-defined borders. The surrounding tissues are firm, and the lesion is not painful on palpation.

In several cases hyperplasia of regional lymph nodes was observed; this was due probably to the ulceration, but node invasion by new growth has been described in a case of round-cell sarcoma and in one of spindle-cell sarcoma. In these two cases the regional lymph nodes
were invaded before general metastasis occurred. In our cases microscopical examination of the lymph nodes showed hyperplasia.

The clinical diagnosis of epithelioma of the lip was made in every case after a negative serum reaction for syphilis. The true nature of the tumor was determined by pathological examination. Biopsies were done in only a few advanced cases, as it seems to have been assumed that this procedure would favor dissemination of the disease.

The types of sarcoma most frequently reported are round-cell and spindle-cell sarcoma, but mixed-cell sarcoma, lymphangiosarcoma, and melanosarcoma have been described. Our cases include only tumors developing on the vermilion border of the lip. The spindle-cell type predominated.

Sarcomas metastasize chiefly by way of the blood current, but that they are also disseminated by way of the lymphatics cannot be denied. Some observers believe, especially in the case of neurogenic sarcoma, that a recurrence in the diseased area may be considered as new tumor formation from the nerve endings.

Treatment has consisted in extensive excision of the primary lesion (regional node dissection is not mentioned). Irradiation has proved less effective than surgery. Irradiation and Coley's toxins have been employed for recurrent growths where operation appeared inadvisable. Most of the reports fail to give exact information as to end-results. In two cases in which end-results were recorded death occurred within a few months, from generalized sarcomatosis. Dr. W. B. Coley, in a personal communication, reported a patient with round-cell sarcoma, treated by extensive local excision and mixed Coley toxins, as alive and well after thirty-two years.

In the four cases to be reported, from the Stuyvesant Square Hospital, the patients were free from symptoms following wide local excision or electrosurgical removal of the original lesion with regional lymph-node dissection, after periods of from six months to four years.

**Case Reports**

**Case I** (Hist. No. 19566. Path. No. 6096–6151. Service of Dr. H. H. Lyle): A woman of twenty-one years gave a history of gastritis four years previously, and of irregular and painful menstruation. She had been operated on four years earlier for lupus erythematosus, then of four years' duration, involving both sides of the nose. This reappeared a year after operation in butterfly form over the nose and cheeks. There was a lesion on the upper lip of six months' duration.

Physical examination showed a fairly well developed and well nourished young woman. The entire fleshy part of the nose had been removed, leaving much scarring and some secondary infection. On the upper lip was a fungating tumor, which was removed by electrosurgical methods. The patient was in good condition six months later, when she left the United States and was lost track of.

The pathological diagnosis was round-cell sarcoma. The first specimen removed from the tumor on the upper lip measured 1 cm. in diameter. The rest of the growth was removed four weeks later, the excised tissue measuring 2.2 x 2.8 cm. Sections show a diffuse growth of moderate-sized cells of lymphoid type invading the muscle. There are many mitoses and small blood vessels in the growth. Because of the absence of white blood changes it seems possible to rule out leukemia and the invasive actively growing process corresponds most closely to a malignant growth of the sarcoma group.
Case II (Hist. No. 22–306. Path. No. 6995–7189. Service of Dr. C. A. McWilliams): The patient was a male of fifty-two years, and the final diagnosis was sarcoma of the left lower lip and prickle-cell epithelioma of the right side of the face, with hyperplastic lymph nodes.

Twenty months before the patient was seen, hydrochloric acid had been splashed over his face and lip on the left side. A small ulcerated area remained on the lip, which had never entirely healed. A physician treated the condition with local applications for two weeks, and the patient was then referred to the Skin and Cancer Hospital for x-ray treatments, receiving about 27 of these over a period of three months. The lesion, however, gradually increased in size, and a surgeon advised operation for epithelioma of the lip.

The lip presented a hard, lobulated mass involving the left side to within 1 cm. of the outer margin, and extending 0.5 cm. to the right side. There were palpable nodes under the jaw, larger on the left side than on the right. The diagnosis was epithelioma, and the entire lower lip was removed and the Dieffenbach operation performed, with bilateral neck dissection.

The specimen was a wedge of lower lip 5 cm. broad along the vermillion border, where there was an elevated ulcerated growth measuring 3 x 3 cm. with a depth of 5 to 7 mm. A block taken at the left edge showed lymph-nodes embedded in a mass of fat and fibrous tissue, together with submaxillary glands, the largest measuring 2 x 2 x 1 cm. Sections show a spindle-cell growth with surface necrosis or ulceration. The tumor cells are oval or fusiform, arranged in interlacing bundles. They invade the deeper tissues and surround the mucous glands. There are hyperchromatism, anaplasia, and mitosis. No suggestion of nests of epithelial cells appears in the sections. There are some nerve bundles in the tissue and also mucous degeneration, so that the growth resembles the other tumors of this series.

In this case there was an associated prickle-cell epithelioma on the surface of the lip, with pearls. The nodes showed only hyperplasia. Four months later operation for a growth in the parotid region showed a deep-seated carcinoma in the muscle, with a superficial prickle-cell epithelioma close to the parotid gland. It would seem, therefore, that we are here dealing with two distinct types of new growth, a spindle-cell sarcoma.
FIG. 2. CASE II: SPINDLE-CELL SARCOMA OF LOWER LIP, SHOWING INTERLACING BUNDLES OF LARGE HYPERCHROMATIC CELLS OF FIBROBLAST TYPE

FIG. 3. CASE III: SPINDLE-CELL SARCOMA OF LOWER LIP; SAME TYPE OF GROWTH AS IN CASE II, BUT WITH SMALLER CELL
of the neurogenic type and an epithelial tumor. The question of compression of epithelial cells to resemble spindle cells can apparently be ruled out, as there was no evidence of compression in the sections.

The patient was well six months after operation. No further information could be obtained.

Case III (Hist. No. 29–404. Path. No. 12569. Service of Dr. H. H. Lyle): The patient was a male of sixty-two years, and the final diagnosis was sarcoma of the right lower lip with hyperplastic (right) cervical nodes.

A few weeks before the patient was seen, a small "cold sore" appeared on his lower lip on the right side. This grew larger and for a month had been discharging pus. The lesion was ulcerated, with a punched-out, infected center and indurated pearly border, typical of squamous-cell epithelioma. There were palpable nodes in the right submaxillary space and in the carotid region, but none on the left side. The diagnosis at this time was epithelioma of the lower lip.

The tumor was excised, with cervical node dissection on the same side. Four years later there was no evidence of local or regional recurrence.

The pathological specimen consisted of a wedge of tissue 4 cm. long and measuring 3 cm. along the vermilion border, where there was a somewhat elevated, ulcerated growth 2 cm. in diameter. This approached close to the line of excision on either side. The growth was firm and had a depth of 6 mm.

The tissue obtained by lymph-node block dissection included the submaxillary gland and some small nodes, some of them with softened centers.

Sections show surface ulceration and a growth of interlacing bundles of fibroblasts, invading the lip downwards and laterally, with no suggestion of epithelial new growth at its edges. The appearance is similar to that of the other spindle-cell growths of the lower lip here reported, but there are few mitoses and less anaplasia. Lymph-nodes showed hyperplasia only.

The pathological diagnosis was spindle-cell sarcoma and hyperplasia of nodes.

Case IV (Hosp. No. 33–2334. Path. No. 18707. Service of Dr. A. B. Morrow): The diagnosis in this case was neurogenic sarcoma of the lower lip, grade 2, with hyperplasia of left anterior cervical nodes.

The patient, a man of sixty-two, complained of a sore on the left side of his lower lip, which started as a "cold sore" four weeks earlier. This never healed, but did not bleed, and was not painful. It had received no previous treatment.
Examination revealed, to the left of the midline of the lower lip, a bluish red, oval, ulcerated lesion which measured upon the surface about 1 cm. in diameter. On palpation the lesion was found to extend for at least 1 cm. into the underlying tissue. This region felt stony hard. No palpable nodes were felt on either side of the neck. A blood Wassermann test was negative. A diagnosis was made of prickle-cell epithelioma of the lower lip, and a wedge-shaped excision was done with a good margin of healthy tissue.

The tissue from the lip consisted of a wedge 2.5 cm. broad along the vermilion border, where there was an ulcer 8 mm. in diameter. Beneath this was a firm growth extending downwards 1 cm. The lymph nodes from the left side of the neck were also removed in block dissection, together with the submaxillary gland. Sections show surface ulceration, with new growth extending laterally under the epidermis and downward into the muscle. The tumor cells are of the fibroblast type, oval and fusiform, with an irregular interlacing arrangement and abundant chromatin in their nuclei. Scattered through the growth are small bundles of nerve fibers. The picture is that of a neurogenic sarcoma. The lymph nodes from the left side showed no neoplasm, though enlarged. Bielschowsky's stain showed connective-tissue fibers running between the tumor cells.

The pathological diagnosis was neurogenic sarcoma, Grade 2, and hyperplasia of cervical lymph nodes.

The patient is in good condition and symptom-free after eight months.

**Neurogenic Sarcoma**

Neurofibrosarcoma and neurosarcoma are terms applied to spindle-cell growths which vary in the number and size of cells and the amount of anaplasia present, so that we have both the hard fibrous types and the softer more cellular forms which may show myxomatous changes. A palisade arrangement of the cells is often visible. Interlacing whorls of cells are characteristic of the active form of this tumor.

In the 3 tumors of the lower lip here reported the irregular diffuse character and fascicular arrangement of the growth point to a neurogenic type of sarcoma. Bielschowsky's stain in Case IV shows the intimate relation of cells to stroma in contrast to epithelial tumors.

While neurogenic tumors are uncommon in the lip, their occurrence in the body is so widespread that there is no reason why the deep tissues here should not show them. Many of these tumors are associated with other features of von Recklinghausen's disease, but in the lip cases described here we do not appear to have that association.

Many pathologists consider that the fundamental cell of these tumors is the Schwann cell. Stewart and Copeland believe that in many of the cases definite nerve connections cannot be demonstrated. The three lower lip tumors described above showing the spindle-cell growth, resemble one another so closely that while the first two (Cases II and III) were diagnosed as spindle-cell sarcoma it seems fair to question whether we may not now group them with the fourth case, of neurogenic sarcoma, and consider them all as of neurogenic origin.

**Summary**

1. In the available literature 20 cases of lip sarcoma were found, in only 5 of which complete records were given.
2. Four additional cases are reported: a round-cell sarcoma of the upper lip, two spindle-cell sarcomas and one neurogenic sarcoma of the lower lip, all of them on the vermilion border.

3. Three of the cases reported were treated by wide local excision followed by regional node dissection. The tumor on the upper lip was removed by electrocoagulation. The patients were all symptom-free after periods of six months (Cases I and II), eight months (Case IV), and four years (Case III).

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