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Abstract

A report is made of a 52-year-old male whose main complaint was a painless tumor at the right side of the palate resulting in speech disturbance. He was diagnosed as a case of what Stout called benign mesenchymoma. Some discussion is also made of the tumor pathology in terms of genetic factors, predirective sites, age range, sex differences and therapy.

KEYWORDS: benign mechenchymoma

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A CASE OF MESENCHYMOMA IN THE ORAL CAVITY CLINICALLY RESEMBLING A LARGE PLEOMORPHIC ADENOMA

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Abstract. A report is made of a 52-year-old male whose main complaint was a painless tumor at the right side of the palate resulting in speech disturbance. He was diagnosed as a case of what Stout called benign mesenchymoma. Some discussion is also made of the tumor pathology in terms of genetic factors, predirective sites, age range, sex differences and therapy.

Key words: benign mechenchymoma

Various benign tumors develop in the palate; for example, papilloma, fibroma, hemangioma, pleomorphic adenoma, osteoma and lymphoma. Lipoma of the palate is said to be rare. Stout (1) reported 8 cases of malignant tumor derived from more than two elements of mesenchymal tissue and diagnosed each tumor as a mesenchymoma. This tumor can occur at any site, appear as a combination of various types, and develop irrespective of the sex or age of the patient. We encountered recently a case of benign mesencymoma that developed in the palate, and present it with some comments in relation to available literature.

A CASE REPORT

The patient was a 52-year-old male whose chief complaint was of a painless tumor mass at the right side of the palate causing speech disturbance. He had noticed a painless tumor of small finger-tip size at the right side of the palate for 13–14 years. The tumor had gradually increased in size without pain.

The patient was of average build and nourished. A lymph node was palpated in each submaxillary area. They were the size of 0.5×0.5 cm, not fixed and not tender. A pedicled tumor mass, about 3×3 cm in size, was seen on the right side of the hard and soft palate. It had a smooth surface, normal mucosal color, the consistency of rubber and was not painful or tender (Fig. 1). Clinical examination showed no other abnormalities (Table 1).

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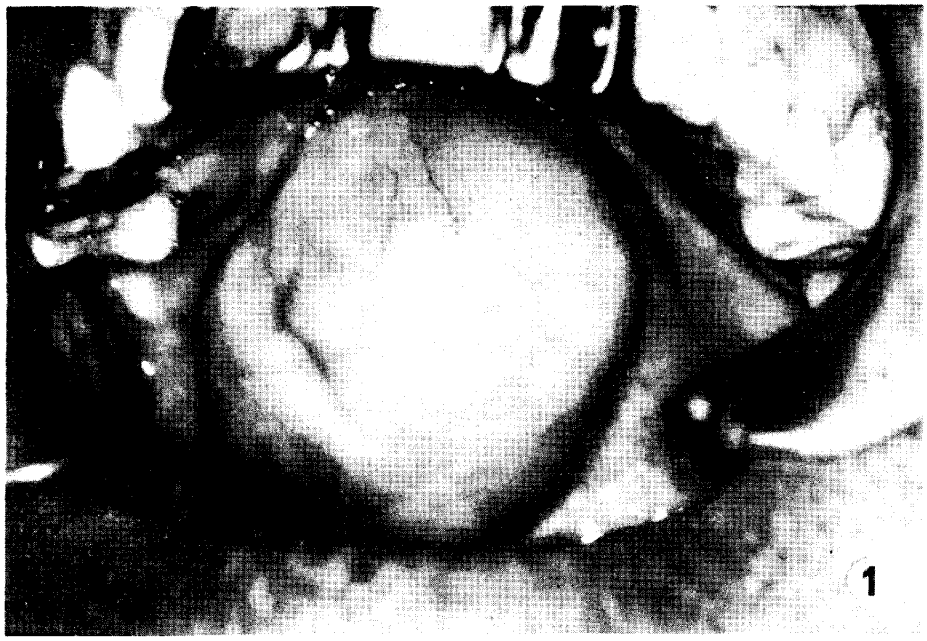

Fig. 1. General view of the oral cavity tumor

TABLE 1. RESULTS OF LABORATORY EXAMINATIONS

ESR	7mm/1h
	12mm/2h
RBC	$494 \times 10^4/\mu l$
Hb	15.6g/dl
Ht	45.0%
WBC	$6300/\mu l$
WaR	(−)
Serum protein	7.4g/dl
Electrolytes	
Na	138.9mEq/L
K	4.9mEq/L
Ca	10.0mg/dl
Cl	101mEq/L
Liver function	
GOT	24 U
GPT	19 U

On January 14, 1977 excision of the whole tumor was performed under general anesthesia. The tumor was well encapsulated and was excised as a mass using alternate and blunt dissection. There was neither infiltration of the tumor

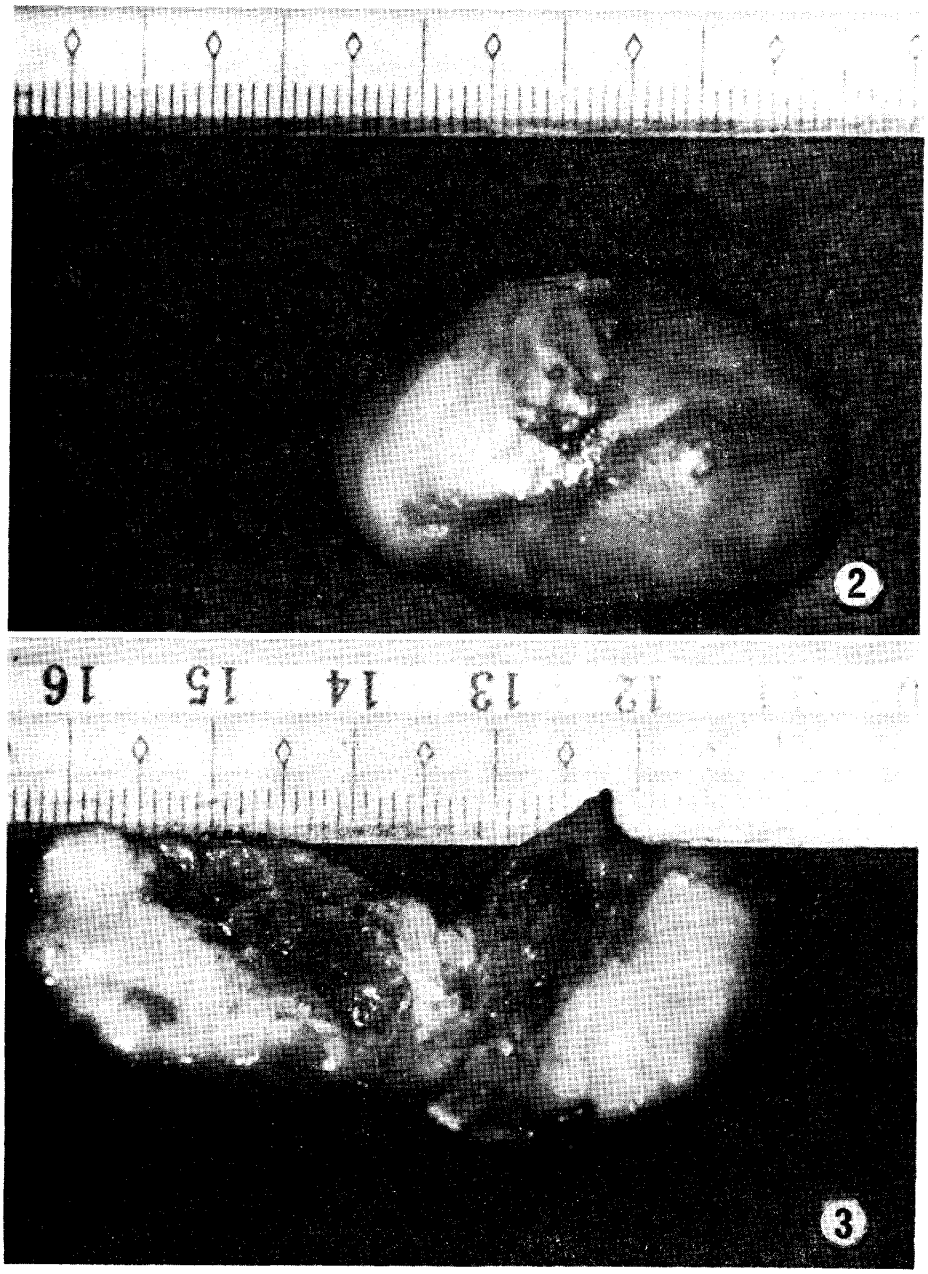

Fig. 2. Gross appearance of the tumor mass
Fig. 3. A cross-section

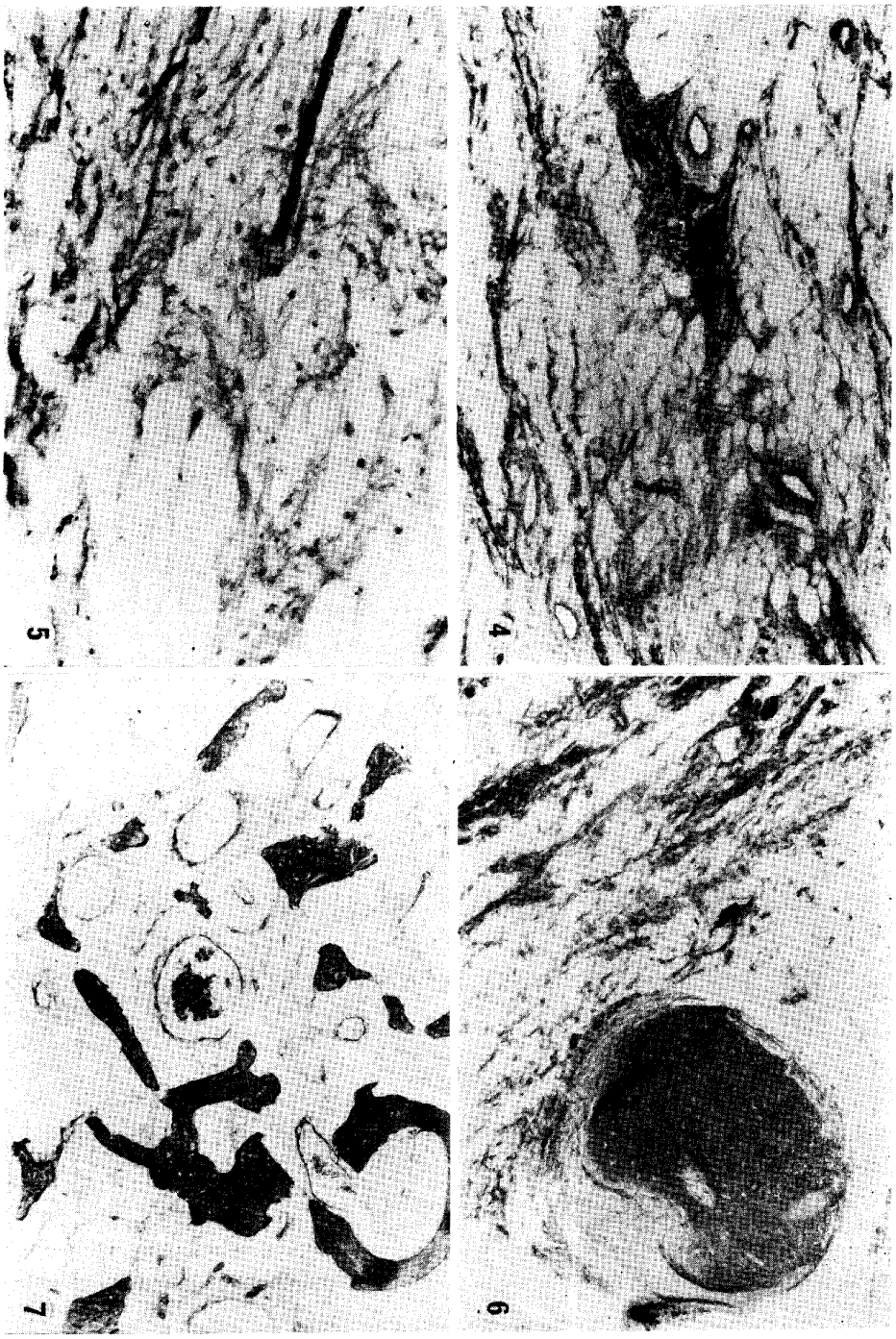
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TABLE 2. REPORTS OF MESENCHYMOMA IN JAPAN

Author	Year reported	Sex	Age	Site		Malignancy
Ishikawa *et al.* (4)	(1958)	F	20 years	Retroperitoneum		Malignant
Kuroda *et al.* (5)	(1966)	F	4 years 4 months	Multiple		Benign
Sano (6)	(1967)	M	26 years	Right neck		Malignant
Iwasa (7)	(1967)			Retroperitoneum		Malignant
Murakami *et al.* (8)	(1970)	F	11 months	Lower abdomen		Malignant
Samezima *et al.* (9)	(1970)	M	10 years	Liver		Malignant
Ichikawa (10)	(1971)	M	38 years	Retroperitoneum		Malignant
Akashi *et al.* (11)	(1972)	M	28 years	Retroperitoneum		Malignant
Maruyama *et al.* (12)	(1972)	M1 F7	18~73 years	Upper arm	1 case	Malignant
				Buttock	1 case	
				Inguinal region	1 case	
				Femur	1 case	
				Retroperitoneum	3 cases	
				Lower thigh	1 case	
Hirono *et al.* (13)	(1973)	M	5 years	Abdomen		Malignant
Yamada *et al.* (14)	(1973)	F	64 years	Left femur		Malignant
Kawayama *et al.* (15)	(1973)	F	51 years	Right breast		Malignant
Suga *et al.* (16)	(1973)	F	19 years	Right breast		Malignant
Naka *et al.* (17)	(1974)	F	2 years 7 months	Retroperitoneum		Malignant
Higashihara *et al.* (18)	(1975)	F	36 years	Right labia majora		Benign
Aoyama *et al.* (19)	(1975)	F	25 years	Parietal lobe		Malignant
Masuda *et al.* (20)	(1975)	M	76 years	Spermatic cord		Malignant
Kurosawa *et al.* (21)	(1976)	F	13 years	Retroperitoneum		Malignant

into surrounding tissues nor was there any communication with minor salivary glands. There was no bone absorption of the palate which was of normal hardness.

The surface of the removed tumor was smooth. There was a pedicle on the middle portion. The cut surface was white and solid with a hard substance in some areas which resembled bone tissue. The tumor measured $3.5 \times 2.5 \times 2.0$ cm and weighed 13 g (Figs. 2 and 3). Microscopically, the lobulation of mature fat tissue and fibrous tissue indicated a fibrolipoma. In some places, there were hamartomatous blood vessels surrounded by very thick connective tissue (Fig. 4). Other areas of stroma gave the appearance of myxomatous degeneration which

Fig. 4. An area showing fibrolipoma HE. ×100
Fig. 5. An area showing myxomatous degeneration PAS. ×200
Fig. 6. An area showing cartilaginous tissue Silver. ×100
Fig. 7. An area showing bone and dilated blood vessels HE. ×100

stained blue with alcian blue-PAS staining (Fig. 5). Cartilaginous tissue was also seen with calcium deposition (Fig. 6). Some areas contained relatively mature bone tissue with proliferation of dilated blood vessels (Fig. 7). In summary, the tumor was mostly fibrolipoma. However, there were other components, namely, cartilage, bone and hamartomatous blood vessels without malignancy, so that it was diagnosed as a so-called benign mesenchymoma. For 16 months postoperatively there has been no recurrence.

DISCUSSION

The term mesenchymoma, according to Stout (1), describes a mixed tumor composed of two or more elements of mesenchymal derivation such as fibrous tissue, fat, smooth muscle, striated muscle, blood vessel, bone, cartilage or myxomatous tissue, but excluding fibrous tissue. Fibrous tissue is excluded because it is the main component of the tumor.

Genetic factors are poorly understood. Consideration of mesenchymoma occurring in the utero-vaginal resion led Amolsch (2) to attribute it to an abnormality in congenital genetic process. The heterogenic theory that undifferentiated mesenchymal tissue in the embryonic stage has strayed into other tissue organs or that it has remained in its undifferentiated congenital form seems reasonable.

This tumor occurs at all ages. Kaufman and Stout (3) reported 6 cases of benign mesenchymoma already present at the birth. There have been 25 cases (benign and malignant) in Japan (Table 2) (4–21). The youngest malignant reported was 11 months old (8), whereas the oldest (20) was 76 years. Of the benign cases the youngest was 4 years 4 months old (5). In respect to incidence related to sex differences, Kaufman and Stout (22) reported 12 cases of male, 26 of female and one was not reported, whereas Nash and Stout (22) reported female cases to be about twice male cases. In Japan, 7 cases were male against 17 cases of female; two cases of benign mesenchymoma were female.

The kidney, extremities and urogenital organs are sites of high occurrence. These are also sites where congenital deformities are frequent, suggesting perhaps a congenital aetiology for this tumor. The occurrence rate is low in the oral cavity (23), and there have been no cases reported in Japan so far. Sterns *et al.* (24), and Muldoon (25) reported malignant mesenchymoma occurring in the mandible. Benign mesenchymoma developing in the tongue has been reported by Cracovaner (26) and Hatziotis *et al.* (27). Junuska *et al.* (28) reported 2 cases developing at the mouth floor. Most cases are isolated and develop in a localized form, but Kuroda *et al.* reported a rare case developing in a multiple fashion.

Macroscopic findings differ according to which elements composing the tumor mass are dominant, so there is no characteristic external appearance.

Tumor components most often seen are fat, blood vessels and smooth muscle. Lymphoid or myxomatous tissue is rare. Eight cases of mesenchymoma reported by Stout in 1948 were all classified as malignant, but angiolipoma of the kidney and mixed tumor of the lung with hamartomatous characteristics are considered benign. In other words, tumors in which each element can be distinguished histologically are considered benign whereas those with a strong heterogeity where this is not possible are taken as malignant. Nash and Stout reported 460 cases diagnosed as mesenchymoma till 1959; 72.8% or 335 cases were malignant. From birth to age 15, 50 of 86 cases (58.1%) were malignant, although Le Ber and Stout (29) reported that the benign cases they studied never turned malignant. However, increases in the rate of recurrence and the change to malignancy (3, 12, 14, 30, 31) microscopically have been reported. Moreover, one case showing infiltration microscopically has also been reported.

Treatment may consist of local excision, but Ewing and Harrison (31) state that amputation at a certain level must be made for tumors of an extremity. Great care is necessary to avoid a partial extirpation, because this tumor tends to recur. Irradiation therapy is not very effective because the mesenchymoma comprises cells of different radiosensitivity. Irradiation after operation, however, is effective in some cases.

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