CENTRAL SALIVARY GLAND TUMORS OF JAWS

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Nine cases of central salivary gland tumors of the jaws were seen during the period 1962–1968. Four patients were in the sixth decade; they all presented with swelling of one of the jaws. Pain was unusual. X-ray of the mandible revealed either a diffuse rarefaction or a multiloculated cystic appearance. Four patients had a cylindroma, 3 had mucoepidermoid carcinoma, and 2 had an adenocarcinoma. Four patients had been treated earlier by local excision or radiotherapy, and, in each case, further progress of disease was slow. However, the overall cure rate has been low, as has been described in the literature. These tumors have a clinical course which is distinct from the course of other common jaw tumors, viz. adamantinoma and squamous cell carcinoma. Because of this and their frequency they merit a separate identity in the classification of jaw tumors.

TUMORS OF SALIVARY GLAND ORIGIN HAVE been frequently reported as arising centrally in the mandible and maxilla. However, these tumors have not been accepted as a distinct entity in the classification of jaw tumors.

Bhaskar[4] described 2 cases of central mucoepidermoid tumors of the mandible; he felt that most tumors described as malignant ameloblastomas up to 1955 were probably tumors of salivary gland origin. Smith et al.[10] reviewed 58 cases of cylindroma, 3 of which arose in the maxillary antrum. Brown and Lucchesi[6] reported a case of central mucoepidermoid tumor of the mandible, and recently, Bradley[5] described a case of cylindroma in the mandible. Smith et al.[11] reviewed 22 cases of mucoepidermoid tumors of jaw bones from the literature, and described 9 cases of their own. Karmarkar[8] reported 2 cases of central mucoepidermoid tumors of the mandible from Bombay.

This report presents 9 cases of central salivary gland tumors of the jaw bones. These patients were seen between 1962–1968, at the All-India Institute of Medical Sciences, New Delhi. These were central tumors, as there was no breach in the oral mucosal surface, and in either case, the mandible and maxilla were expanded on more than one surface.

Incidence (Table 1): During the same period, a total of 24 cases of jaw tumors were seen. Squamous cell carcinoma of the maxillary antrum and squamous tumors infiltrating the mandible from the floor of the mouth were excluded. In this group of 24 patients, 7 had adamantinoma, and 9 had central salivary gland tumors. Of the latter 9, 4 were in the mandible and 5 were in the maxilla.

All 4 mandibular tumors were in female patients, and all 5 in the maxilla were in males. Though the youngest patient was 25 years old, the tumors tended to occur in the fourth, fifth, and sixth decades (Table 2).

Clinical presentation (Table 3): The patients presented with a swelling of one of the jaws (Fig. 1). Pain was present in only 3, and it was not severe. When first seen, the symptoms were present for less than 18 months. In 8, they were present for less than 6 months. The swelling in each case was smooth, rounded, firm and uniformly expanding the jaw. The oral mucosa over the swelling was intact. In the mandible, 3 of the swellings appeared to begin near the angle, while one was distinctly located in the body. This last patient had a swelling of the parotid which preceded the jaw swelling, but the

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TABLE 1. Tumors of Jaws (1962–1968)

	Mandible	Maxilla
Adamantinoma	7	0
Osteoclastoma	2	0
Salivary	4	5
Sarcoma	0	3
Reparative granuloma	1	0
Fibrous dysplasia	0	1
Mucocoele	0	1
TOTAL:	14	10
TOTAL:	24	

2 were quite separate. This last patient probably had a central secondary deposit in the mandible from a primary in the parotid. She also had a facial palsy. Another patient with maxillary tumor had orbital spread and ophthalmoplegia. Proved lymphatic metastasis was demonstrated in one patient in the postoperative period. Pulmonary metastasis occurred in 2 patients.

X-ray appearances in the maxilla were atypical. There was expansion and erosion of the bony walls of the antrum with a soft tissue shadow (Fig. 2). In the mandible, the radiologic appearances were more specific. There was an area of rarefaction and erosion of the bone with indistinct margins (Fig. 3). In only one patient was there an expansion of the bone with a multiloculated cystic appearance (Fig. 4). This patient had an adenocarcinoma of the mandible.

Histology (Table 3): Four patients had a cylindroma (adenoid cystic carcinoma)—one in the mandible and 3 in the maxilla. Three patients had a mucoepidermoid carcinoma, one of them in the mandible and 2 in the maxilla. The eighth patient had an adenocarcinoma in the mandible, and the last patient with a concurrent swelling of the parotid and mandible had an adenocarcinoma at both sites (Figs. 5–7).

Previous treatment (Table 3): Three of the patients with maxillary tumors had previous treatment. One of them (A.R., cylindroma) had biopsy and radiotherapy which slowed down the progress of the disease. Maxillectomy was done 2½ years later. A second patient (N.D., cylindroma) had an antrostomy and removal of tumor and was symptom-free for 16 months. The third patient (M.S., cylindroma) had local excision and radiotherapy and was symptom-free for 3 years. One of the patients with mandibular tumor (R.M.D., adenocarcinoma) had biopsy and radiotherapy. Although the tumor did not regress completely, the patient did not seek medical advice again until 5 years later, when a mandibular resection was done.

Management (Table 3): One patient with a mandibular swelling (V.B., cylindroma) was inoperable due to the local extent of the disease and pulmonary metastasis. Radical excision was done in the other 8 patients, 2 of whom (A.R. and S.S., maxillary cylindromas) died in the postoperative period because of their advanced age and poor general health.

Among the 6 survivors, one patient (Sh) with a mandibular mucoepidermoid tumor developed cervical metastasis, first on one side one year after operation and then on the other side 3 years later. Radical neck dissection was done on one side, but the patient refused to have it done on the other side. Another patient (R.M.D., adenocarcinoma, mandible) developed local cervical and pulmonary metastasis a year after radical surgery. She remained in good health 18 months later. One patient (M.S., cylindroma, maxilla) had tumor spreading along the optic nerve, and the excision was incomplete. Of the remaining 3 patients, 2 remained well one year and 3 years, respectively, when last seen. These had an adenocarcinoma of parotid with mandible (Ba)

TABLE 2. Central Salivary Gland Tumors of Jaws

Site	Site, sex, and age distribution: Sex M	Sex F	Age in yrs	Decade
Mandible	0	4	50, 40, 25, 70	3rd decade — 1 4th decade — 2 5th decade — 1
Maxilla	5	0	52, 60, 38, 60, 60	6th decade — 4 7th decade — 1

TABLE 3. Central Salivary Gland Tumors of the Jaws (Clinical Data)

No.	Name	Sex	Age (yrs.)	Site	Swelling	Pain	Duration of symptoms when first seen	Spread	Histology	Previous treatment	Symptom-free interval	Management	Results
1.	N.D.	M	52	Max.	+	—	2 mos.	—	Cylindroma	Local excision;	16 mos.	Maxillectomy	?
2.	A.R.	M	60	Max.	+++	—	3 mos.	—	Cylindroma	R'therapy	2.5 yrs.	Maxillectomy	Postop. death
3.	M.S.	M	38	Max.	+	+	4 mos.	Orbit	Cylindroma	Local excision; R'therapy	3 yrs.	Rt. radical Maxillectomy. Exenteration orbit	Spread along optic nerve
4.	Kh.	M	60	Max.	++	—	4 mos.	—	Mucoepid. ca.	—	—	Maxillectomy	Satisfactory 3 yrs.
5.	S.S.	M	60	Max.	++	+	1 mo.	—	Mucoepid. ca.	—	—	Maxillectomy	Postop. death
6.	Sh.	F	50	Mand.	++	+	5 mos.	Lymphatic: Contralateral -1y. Ipsilateral -3y.	Mucoepid. ca.	—	—	Mandibulectomy + rib graft. Lt. radical neck dissection. Right refused	Died 4 yrs. from onset.
7.	V.B.	F	40	Mand.	++	—	4½ mos.	Pulmonary deposit	Cylindroma	—	—	Inoperable	? Died
8.	R.M.D.	F	25	Mand.	+++	—	6 mos.	Hypoglossal nerves	Adenoca.	R'therapy	5 yrs.	Mandibulectomy.	Local recurrence & pulmonary deposits after 1 yr.
9.	Ba.	F	70	Mand. Parotid	+ +	—	18 mos.	Facial nerve	Adenoca.	—	—	Parotidectomy, mandibulectomy	Satisfactory for 1 yr.

Max. = Maxilla; Mand. = Mandible.

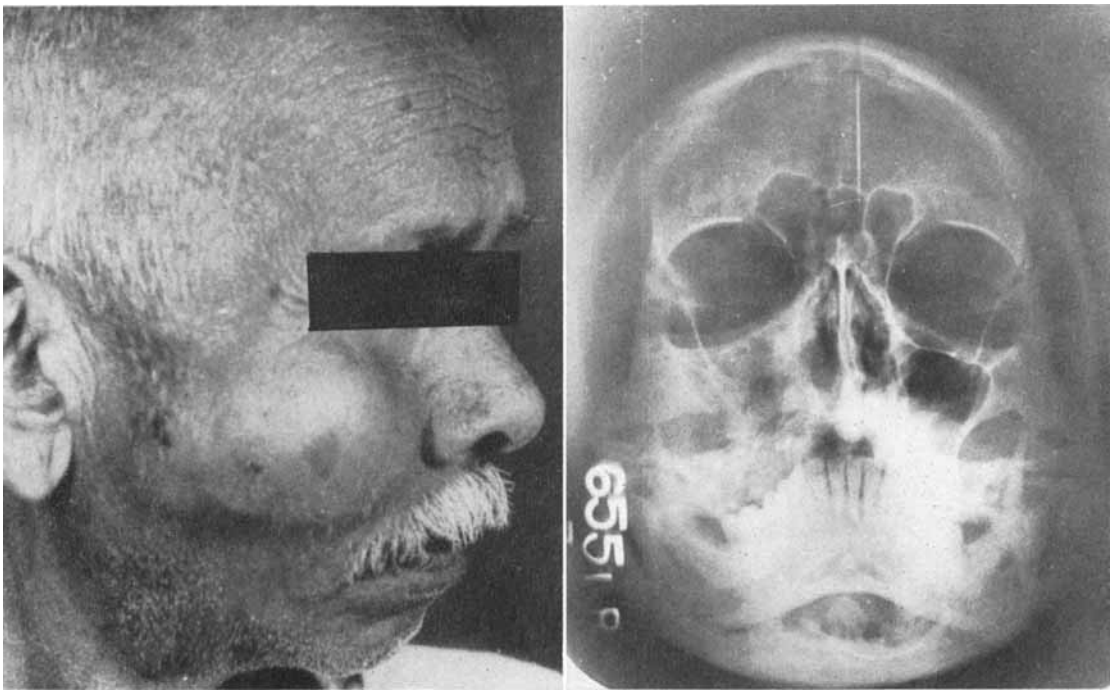

FIG. 1 *(left)*. An elderly patient with a mucoepidermoid carcinoma of the maxilla.
FIG. 2 *(right)*. Appearance of a mucoepidermoid tumor in the right maxilla.

and mucoepidermoid carcinoma of maxilla (Kh), respectively.

DISCUSSION

It would appear from the material presented that central salivary gland tumors of the jaws have a distinct clinical identity. They occur in the fourth, fifth, and sixth decades, almost the same age group as squamous cell tumors, but much later in life than adamantinoma and giant cell tumors. In the current series, they were seen more often than the adamantinoma which, how-

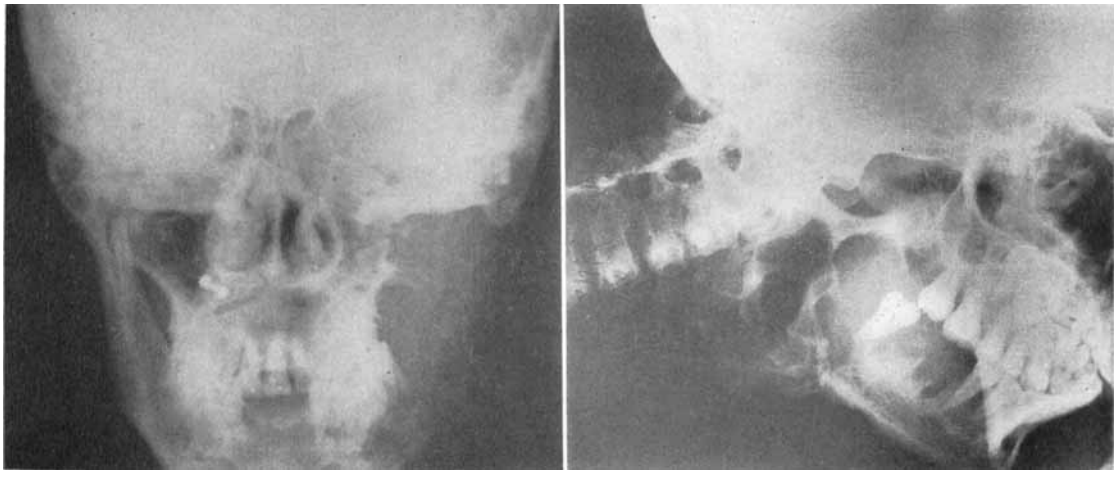

FIG. 3 *(left)*. X-ray mandible of a patient with cylindroma of mandible, showing diffuse erosion and rarefaction of the bone. There appears to be no expansion of the cortex. The changes are in the ascending ramus, behind the last molar.
FIG. 4 *(right)*. X-ray mandible of a patient with adenocarcinoma, showing a multiloculated cystic appearance, confined to the part behind the last molar.

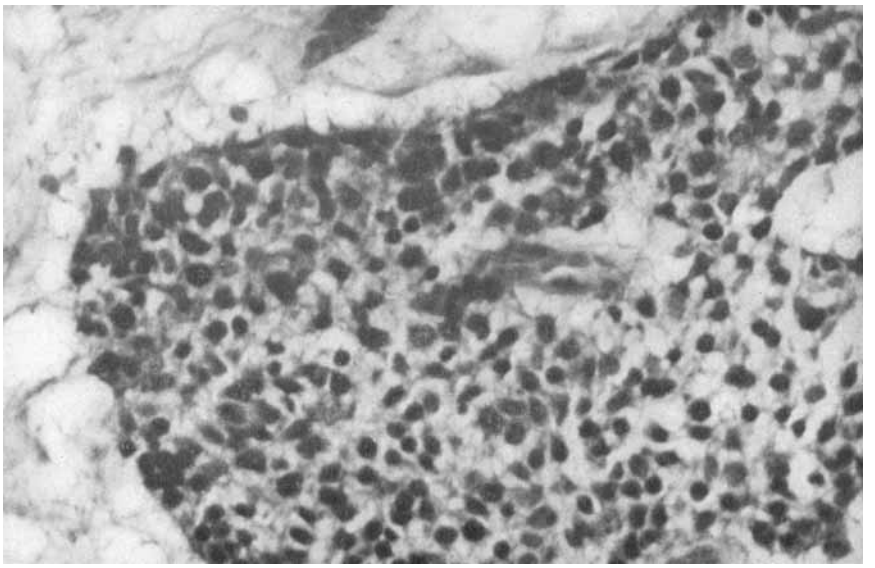

FIG. 5. Details of tumor cytology in case 7 (V.B.). The features are characteristic of cylindroma with small uniform cells having darkly staining nuclei (H and E, ×320).

ever, was entirely confined to the mandible. Like squamous cell cancers, salivary gland jaw tumors have a shorter history of onset. This distinguishes them from adamantinoma, where the duration of symptoms is much longer. However, like adamantinoma, salivary jaw tumors are central in origin and have an intact oral mucosa over them. Presenting clinical symptom is a swelling, and pain is not a regular feature as it is in squamous cell tumors. The rate of growth can be slowed down for several years by radiotherapy (and this would probably depend on histologic features). Limited surgical excision may be followed by a greatly delayed recurrence. They may spread to the lymph nodes and, unlike squamous cell tumors, may metastasize to the lungs. Cure rate is low, although these tumors are slow growing. Thus, although there are a few features where these tumors may resemble squamous cell carcinoma, there are many distinguishing features between the 2.

Smith et al.,[10] in their review of 3 maxillary cylindromas among a group of 58 such tumors, stated that few patients can be cured, though many survive for long periods. Radiotherapy was only temporarily effective. Duration of the disease was much longer than in other adenocarcinomas. Only half the patients with local recurrence were dead within 5 years of appearance of recurrent cylindroma. All these facts emphasize the slow progress of the tumor after treatment and its low cure rate. Smith and others[11] presented 9 of their own cases of mucoepidermoid carcinoma of the jaws and reviewed another 22 from the literature. They described the radiologic appearance as generally multilocular cystic. The average duration of symptoms was 10 years in their cases. They state that a benign mucoepidermoid tumor does not exist. Brown and Lucchesi's[6] mucoepidermoid tumor of the mandible showed a multicystic appearance and was treated by repeated curettage for 14 years, when it was considered to have acquired a malignant character.

Because of the small number of cases, it is difficult to draw conclusions about the differences in behavior between cylindromas and mucoepidermoid tumors of the jaws. However, it is reasonable to expect that their natural history would correspond to that of identical tumors at other sites. The 3 maxillary cylindromas in the series of 58 such tumors reviewed by Smith et al.[10] appeared to behave like similar tumors at other sites. The cylindromas in this series, and the case quoted by Bradley, had a similar behavior. The mucoepidermoid tumors of the jaws, however, do not appear to be as mild as the cases reviewed by Bhaskar and Bernier[3] in the minor and major salivary glands. They believed that such neoplasms in minor and major salivary glands could be divided into benign and malignant varieties, and that local excision produced a cure in 80% of patients. On the other hand, the mucoepidermoid jaw tumors in this series, and those

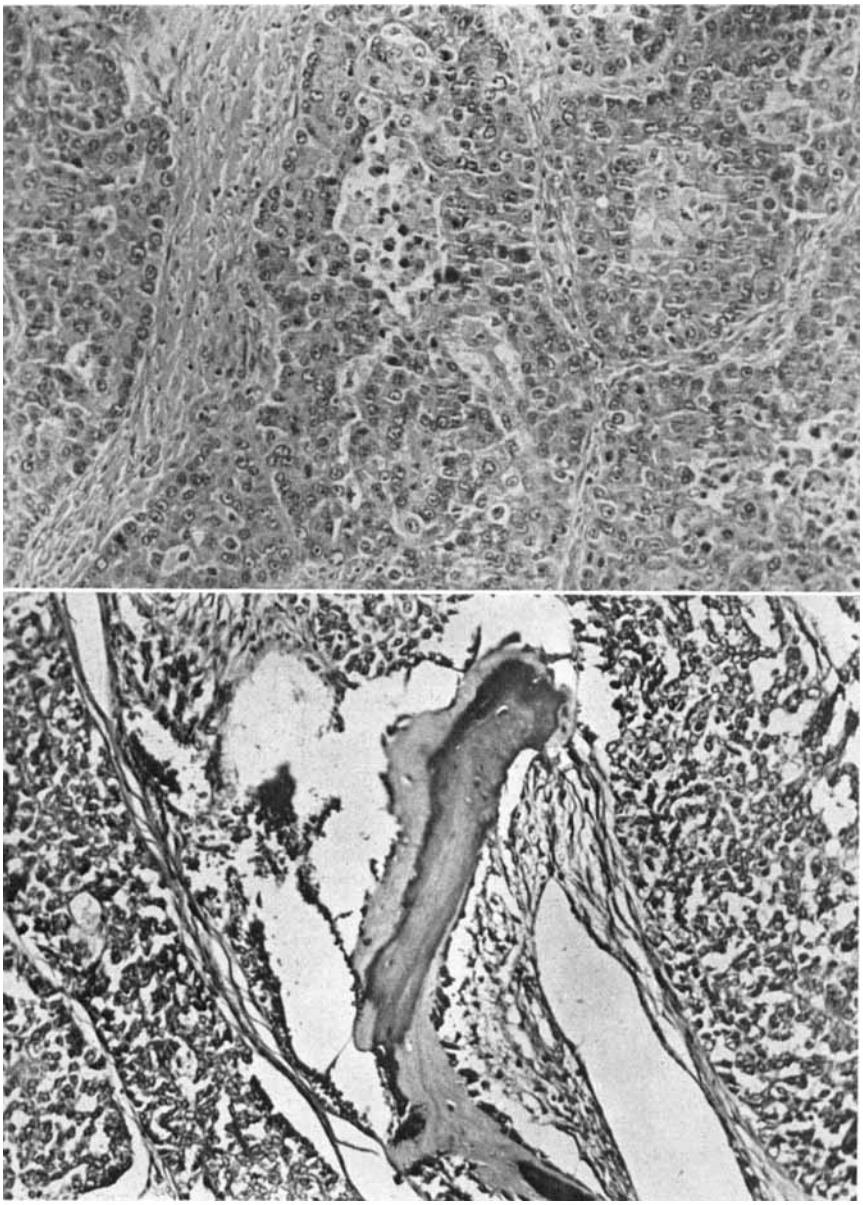

FIG. 6. Mucoepidermoid carcinoma of the maxilla; large mucoid cells with foamy cytoplasm are seen in the center of closely packed squamoid cells (H and E, ×160).

FIG. 7. Intramandibular mucoepidermoid carcinoma. Sheets and groups of tumor cells are seen in between bone trabeculae. In a few areas, mucoid cells are seen (H and E, ×160).

reported in the literature, appear to behave more like malignant tumors with a low cure rate. In fact, Smith et al.,[11] who reviewed 33 mucoepidermoid jaw tumors, remarked that a benign mucoepidermoid tumor does not exist.

In an attempt to describe the origin of these tumors, Bhaskar[4] said that the source of origin in the mandible is probably the mucus glandular inclusion in the retromolar area. Occurrence of ectopic salivary tissue in the mandible producing a translucent defect has been reported by Richard and Ziskind,[9] Araiche and Brode,[2] Hayes,[7] and Amaral and Jacobs.[1] In the maxilla, these tumors most probably arise from ectopic salivary gland tissue in the lining of the antrum. Bhaskar has made a plea that these tumors of the jaws be recognized as a distinct clinical entity.

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