CLINICAL RECORDS

INTRALINGUAL NEURILEMMOMA

By J. A. HARPMAN and A. P. PRIOR (Warwick)*

THIS note may prove of interest owing to the unusual nature of and the diagnostic and surgical problems presented by the case here described and discussed.

Case History

A 23 years old man had suffered, for four years, from progressive fullness of the suprahyoid area, muffling of the voice, difficulty in swallowing solids and, especially when he had a cold, some dyspnœa. He had been thought by several doctors to have enlarged submaxillary salivary glands; actually, these glands were not diseased, but rendered prominent because they were pushed downwards with the floor of the mouth. Examination of the pharynx was rendered difficult by a swelling within the root of the tongue; because of this, the larynx could not be seen by indirect laryngoscopy—nor could it have been reached by direct laryngoscopy. The mucous membrane at the root of the tongue appeared yellowish and thinned—in contrast to its appearance in a patient with a lingual thyroid, in whom large blood vessels were to be seen in this situation. The latter patient had had serious buccal hæmorrhages; the present patient had had none. Soft tissue lateral X-rays of the region of the root of the tongue in these two cases also presented different appearances: in both cases there was obviously a swelling within the posterior part of the tongue, depressing the epiglottis, but in the case under consideration the swelling appeared smooth and rounded; in the case of lingual thyroid it appeared less regular (Figs. 1 and 2). A radioactive iodine test proved the one patient to have a lingual thyroid without thyroid tissue in the normal situation, and the other to have only normally placed thyroid tissue. A diagnosis of benign neoplasm, possibly cystic, was made.

Surgery was obviously required. It was also clear that the first step of the operation would have to be a tracheotomy under local anæsthesia; this was done through a short transverse incision just above the suprasternal notch; a size 6 cuffed MacGill's endotracheal tube was passed into the trachea, and a pentothal, nitrous oxide, oxygen and trilene general anæsthetic administered (Dr. D. Rigg). How to approach the neoplasm had given rise to much consideration. Ian Aird (1949) states that a lingual thyroid can be satisfactorily excised through the mouth, by splitting the tongue down its midline. In this case the nature of the tumour was not known, it lay very far back in the tongue and it was large. It was not thought that an intraoral approach would be sufficient for adequate removal, nor that it would be safe if gross bleeding were to occur. Sir James Walton (1943) recommends, for larger cysts and tumours in the substance of

* From the Departments of Ear, Nose and Throat Surgery and Pathology, Warwick Hospital.

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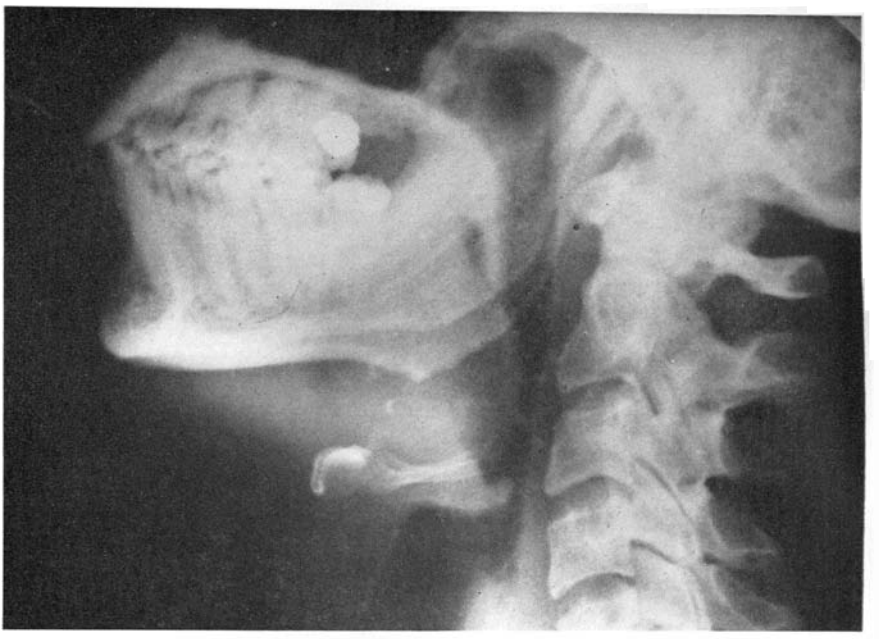


FIG. 1.
Lateral radiograph of lingual thyroid.

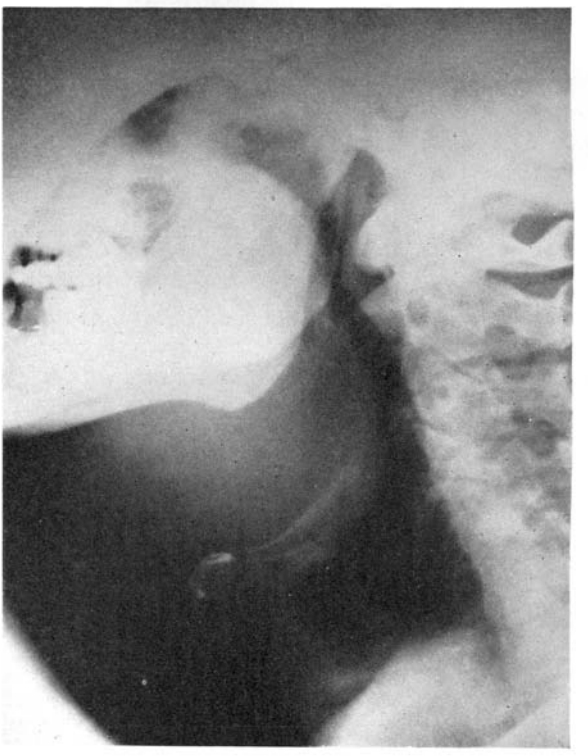


FIG. 2.
Lateral radiograph of the present tumour.

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the tongue, an approach through a slightly curved transverse incision at the level of the hyoid bone; a flap of skin and platysma is then dissected upwards, and the lesion excised after dividing the mylohyoid raphe and separating the fibres of the geniohyoid and genioglossus muscles. This method it was decided to adopt. A vertical incision in this situation may be easier, but is liable to leave an ugly scar. Trotter's median division of the tongue, lower jaw and suprahyoid region (e.g. Gardham, 1943) was considered and discarded for cosmetic reasons, because such a large exposure was thought unnecessary and for fear of bone infection—I have seen chronic low-grade osteomyelitis of the mandible after similar surgery under intensive antibiotic cover.

The pharynx was packed with gauze to push the tumour downwards and to deal with blood that might enter it.

In making the long transverse incision, care was taken to preserve the lowermost branch of the facial nerve on each side. It was found, after splitting the mylohyoid raphe and separating the geniohyoid and genioglossus muscles of the two sides, that the tumour was encapsulated and solid (Fig. 3). It was too large to be removable through the exposure so far obtained. The nature of the tumour not being known, it was not considered wise to remove it by opening it and removing it piecemeal. Dividing the hyoid bone in the midline afforded little additional room. The medial parts of the right mylohyoid, geniohyoid, genioglossus and hyoglossus muscles were divided; this division is best done gradually with a scalpel, and not with diathermy, to facilitate recognition and preservation of hypoglossal nerve and lingual artery branches. The additional exposure thus obtained still not being sufficient, the medial parts of the same muscles on the left side were divided, but to a lesser extent. The tumour could then be readily shelled out (Fig. 4). Before dividing the muscles, I had thought of elevating them subperiosteally from the hyoid bone and retracting them upwards (as advised, for instance, by Klopp and Delaney (1950)), but it seemed very doubtful whether this would give enough exposure. A small opening was made in the thinned mucosa over the dorsum of the base of the tongue; it was closed with a catgut ligature. The intrinsic muscles of the tongue were very thinned; it had been intended to suture them together to obliterate the space left by the removal of the tumour, but there was so little of them that it was thought unwise to pass deep sutures through them for fear of strangulating main branches of the hypoglossal nerves. The divided muscles were reconstituted with a few catgut sutures; the tumour bed was drained with a gauze wick after careful diathermy coagulation of all bleeding points; the platysma was sutured with catgut and the skin closed with Michel clips.

Post-operatively, the patient was fed for a week through a naso-œsophageal feeding tube that had been introduced pre-operatively. There was considerable post-operative swelling; the larynx did not become visible on direct laryngoscopy for 10 days; the tracheotomy tube was then left out. Sero-sanguineous fluid oozed through the drainage opening for three weeks. There was considerable interference with the mobility of the tongue. Two months later tongue movements, speech and swallowing were still improving.

Pathological Examination

Macroscopically the specimen consisted of a spheroid mass of material which measured 6 cm. at its greatest diameter. It appeared to be encapsulated. A few

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FIG. 4.

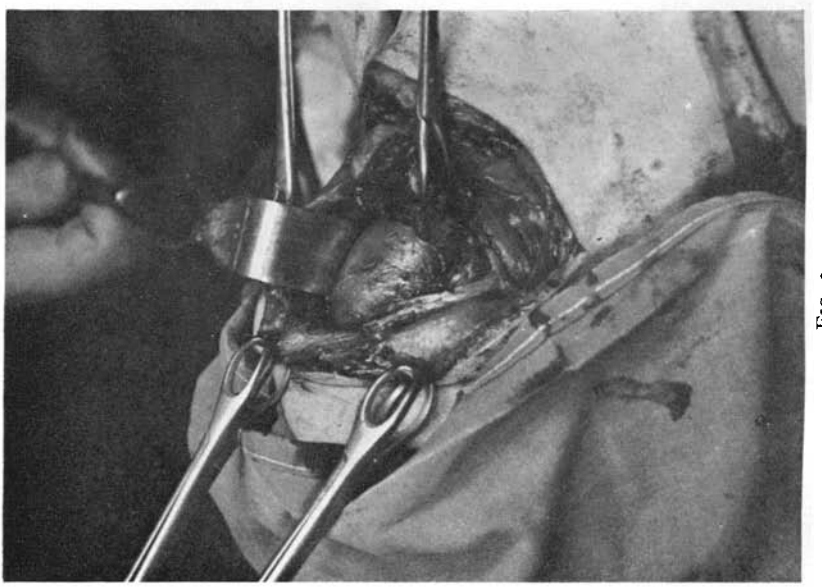


FIG. 3.

FIGS. 3 and 4.

Photographs taken during the operation. The patient's head is towards the viewer's left. In Fig. 3 the right submaxillary salivary gland and the right anterior belly of the digastric muscle are seen as well as the partially divided right suprahyoid muscles and the partially exposed tumour. Fig. 4 shows the tumour "delivered".

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fine tags were adherent to the outer surface. An incision had been made into the equator of this sphere and this divided it almost into two. The cut surface presented a fine fibrous capsule which enclosed and surrounded a mass of yellowish tissue with a greasy feel and a shining homogenous surface (Fig. 5).

Microscopically the presence of a fibrous capsule was confirmed. There was a slight, delicate, vascular stroma. The tumour consisted essentially of elongated strap-like cells. These were generally arranged in sinuously twisted cords. There were a number of reticulum fibres which passed between the cells, and very often elongated cells showed a delicate argyrophil boundary. The nuclei were elongated and showed a curious vacuolation. Regimentation was not an obvious

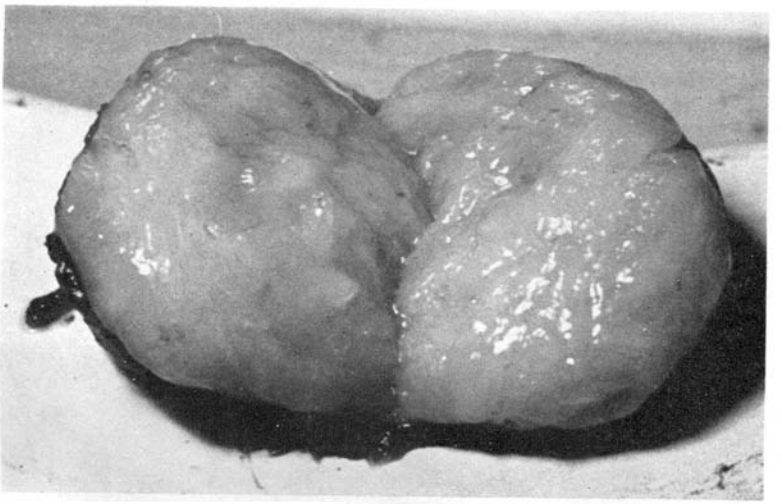


FIG. 5.
Photograph of the bisected neurilemmoma.

feature but could be found with moderate ease. Stained by Van Gieson the elongated cells took on varying tints of yellow. Microcysts and other areas of degeneration were not found. No distinct nerve elements were found. In view of the morphology of the cells and their tinctorial characters, the shape and arrangement of the nuclei and of the absence of any evidence of relation to muscle cells, it was concluded that this spindle cell tumour had the characters of a neurilemmoma. It was fasciculated and reasonably orderly. Microcysts and mucin could not be demonstrated and regimentation of nuclei was seen. Thus the neurilemmoma belonged to Type A (Figs. 6, 7, 8, 9).

Comment

A question that arises is: would Trotter's median approach have resulted in less interference with the mobility of the tongue?

Neurilemmomas in the tongue have been reported previously (Willis, 1953; Stout, 1935, 1949, and others quoted by him). A slightly fluctuant feel of the tumour, and occasional macroscopic appearance like a lipoma, are recognized;

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FIGS. 6-9. Photomicrographs of histological sections of the tumour.

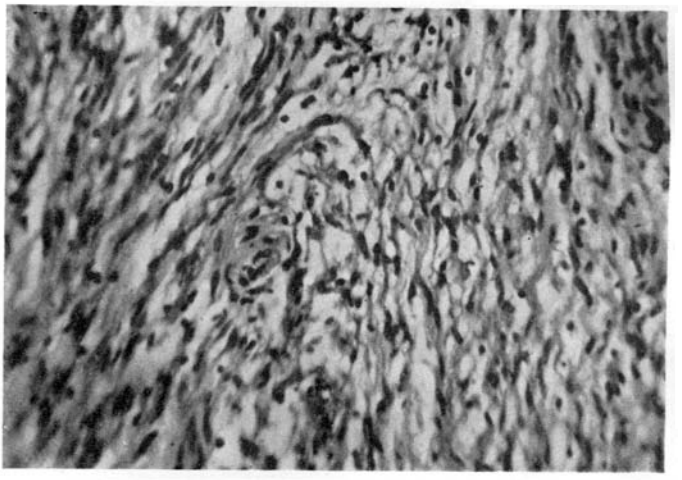


FIG. 6.
Hæmatoxylin and eosin. × 220. Shows constituent cells arranged in fasciculi.

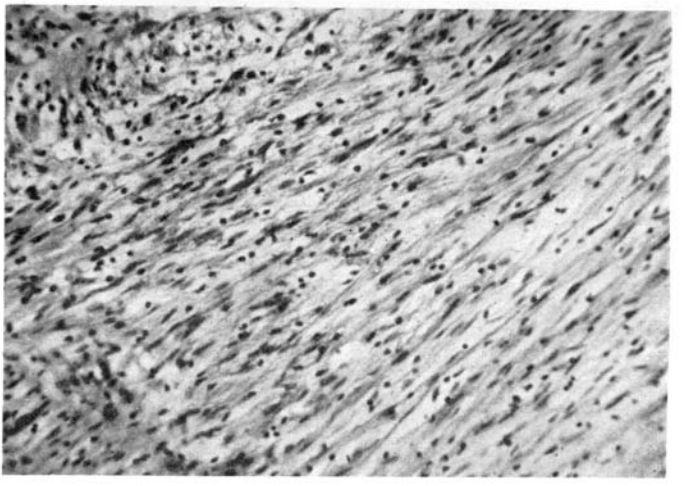


FIG. 7.
Iron Hæmatoxylin and Van Gieson. × 160. Elongated strap-like cells showing a regularity of arrangement. Regimentation of nuclei.

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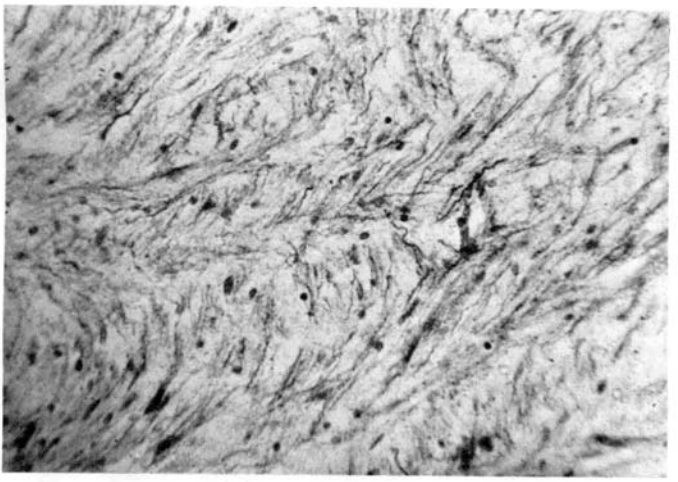


FIG. 8.

Laidlaw's Silver. × 220. Argyrophil borders to cells and reticulum inter-cellular bridges.

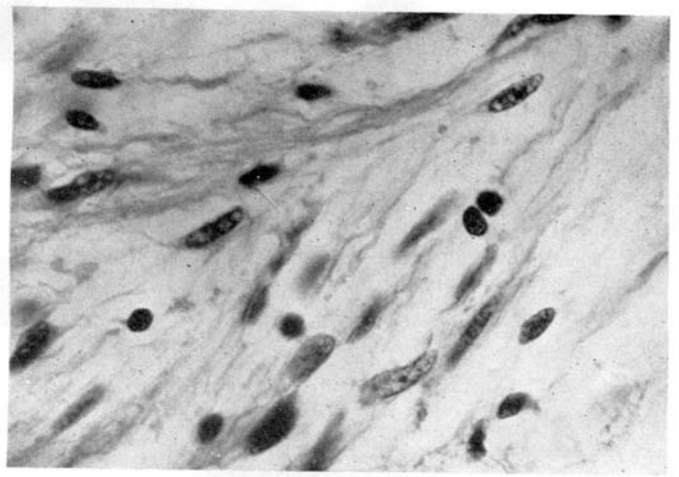


FIG. 9.

Iron Hæmatoxylin and Van Gieson. × 630. The elongated nuclei of the tumour cells are shown and some of these have vacuoles. The nuclei are clearly distinguished from the lymphocytes which are also present in this field.

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so is the type of capsule. The tumour in our case is unusual because of its size and the surgical problem this presented. These tumours are benign.

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