was gripping and spasmodic but not very severe. There was no nausea or vomiting, and no alimentary or urinary symptoms.

On examination a purulent discharge was seen coming from the umbilicus, and just below it there was a small spheroidal swelling in the abdominal wall. Cystoscopy and cystography were normal.

Operation.—A midline incision with an ellipse was made around the umbilicus. A urachal cyst was defined and dissected free. The urachal cyst was followed down to the dome of the bladder, where it was clamped and divided. The bladder wall was repaired in two layers with catgut. The obliterated hypogastric arteries were also dissected out, the whole procedure being performed extraperitoneally. The linea alba was repaired with wire and the skin closed about a corrugated drain.

Pathology.—Microscopically there was no epithelial lining to the cyst, and no lumen was present in a transverse section of the distal end of the urachus. The appearances were those of an infected urachal cyst.

Comment

Cases 1 and 2 fall into Long's group 4, and Cases 3 and 4 into group 2. They show several interesting points in the ways they presented, in diagnosis, and in treatment. Case 1 presented first as acute retention of urine, and there was difficulty in deciding how this arose. A possible explanation is that inflammation in the urachal relic caused a block in the lymphatic territory, which it presumably shares with the bladder-neck, since both derive from a common ancestor—namely, the ventral cloaca. This would cause oedema around the neck of the bladder, and a "congestive stricture" would result. Some support is given to this theory by the finding, on cystoscopy, of "a prominent anterior commissure" to the prostate which was probably due to oedema, since it had completely disappeared at the final examination. The ring of bullous oedema in the dome of the bladder which gave the first clue to the diagnosis was very striking indeed. Case 2 presented as an inflamed umbilical hernia with strangulation, and only at operation did the true nature of the condition become apparent. Cases 3 and 4 complained of a discharge from the umbilicus with an associated midline swelling, and a pre-operative diagnosis was made with confidence.

Case 1 was treated by saucerization of the cyst and track combined with diathermy cauterization of the epithelial lining. In Cases 2 and 3 the umbilicus and the urachal cyst and track were excised with their covering peritoneum; while in Case 4 the cyst and track were dissected out without opening the peritoneum. The final result was similar in each case.

In the last year one other case has been seen, which, although not capable of complete proof, is highly suggestive of urachal pathology.

A housewife aged 64 complained of a painful swelling in the lower abdomen of one week's duration. She had recently had an attack of influenza and bronchitis, but no symptoms referable to her genito-urinary or alimentary tracts. On examination there was a large rounded swelling in the anterior abdominal wall, extending up from the symphysis pubis, half-way to the navel. There was central fluctuation beneath red and oedematous skin, and obviously pus lay underneath. The mass was incised and a large quantity of foul-smelling pus escaped from a subcutaneous abscess. The latter led through a hole in the linea alba to another abscess cavity lying behind the recti muscles but in front of the peritoneum, which was intact. The abscess was saucerized and drained through two lateral stab drains with suture of the midline incision about a central drain. She made an uneventful recovery, and when seen eight months later the lower abdomen was quite healed and soft. Pathological examination of the pus showed numerous pus cells but no organisms. There was no growth on culture.

The midline situation of the abscess, its "collar-stud" nature, and the fact that the communication between its two compartments was so strictly midline suggested an

infected urachal cyst as a possible diagnosis. Of course, the alternative diagnosis of an infected haematoma brought on by her recent coughing is equally admissible.

These cases seen in a short period seem to indicate that the urachal remnant is a more frequent cause of disease in the umbilicus and anterior abdominal wall than is perhaps generally realized.

Summary

The development of the urachus and the pathogenesis of urachal cysts are discussed.

A note is added on their incidence, diagnosis, and treatment.

Four cases of infected urachal cysts and possibly a fifth case are described and the clinical and operative findings commented upon.

I should like to express my gratitude to Mr. D. P. Marks, of Stratford-upon-Avon Hospital, and Mr. Henry Clarke, of Luton and Dunstable Hospital, for permission to publish these cases and for assistance with their management; to Mr. L. R. Broster for encouragement and criticism in writing this paper; to Dr. J. Bradley Watson for the pathological reports; and to the librarian of the B.M.A. Library for considerable help with references.

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LINGUAL THYROID

BY

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Lingual thyroid is a rare occurrence. It appears that Hickman recorded the first case in 1869. Buckman (1936) collected 242 cases from the literature up to and including 1935, but Montgomery (1935, 1936), studying probably the same material, reduced this number to 144 which he accepted as genuine lingual thyroids. Crispell and Parson (1950) stated that up to 1949 a total of 276 cases had been reported. Between 1949 and 1951 I was able to find seven more reports of lingual thyroid, including the case described below. In all these cases the lingual thyroid was causing symptoms; the incidence of silent cases will never be established.

Montgomery recognized as authentic lingual thyroids only those cases in which a specimen was examined and proved histologically to consist of thyroid gland tissue and when the lesion was situated in the tongue substance between the epiglottis and the circumvallate papillae. From the embryological point of view, it is natural to expect that the most common site for its occurrence in the tongue should be in the region of the foramen caecum.

According to Montgomery (1935, 1936), Goetsch (1948), and Nordland and Nordland (1950) "lingual thyroid" and "lingual goitre" should not be confused with "aberrant thyroids," as they accept the view of Warren and Feldman (1949) that the latter are metastatic deposits from carcinoma of the thyroid gland.

A great majority of reported lingual thyroids were histologically diagnosed to be benign. A definite malignancy in lingual thyroids was reported in few

instances (Graham and McWhirter, 1947). Most cases resembled the normal thyroid tissue, but adenomatous changes have been observed in some. In most of them, normal type of epithelium predominated, and in others the foetal type was present. Simple goitre of colloid type has also been seen.

Lingual thyroids or goitres are detected usually when their size is such that they give rise to symptoms. It has been postulated that thyroid insufficiency is responsible for most, if not all, simple hypertrophies of lingual thyroid tissue. No case has been reported with hyperthyroidism. The thyroid insufficiency may arise when a special stress such as puberty, pregnancy, or lactation exists, and in the total absence of the thyroid in the normal anatomical position these factors excite a compensatory hypertrophy of the ectopic thyroid tissue. This view is well supported by the histories of the recorded cases. The menopause was also described as a possible factor. Most of the patients were females.

The interesting feature in the following case is the unusual presenting symptom of haematemesis, which was misleading enough to cause considerable delay in establishing the diagnosis. The rarity of this condition precludes learning from one's own experience. The main object of this paper is to draw attention to its existence and to give guidance in management of a similar case.

Case Report

A married woman aged 30 was admitted as an emergency case in December, 1950, because of haematemesis, but giving no previous history of vomiting or dyspepsia. She was of small stature, 4 ft. 8 in. (142 cm.), fairly well nourished, and of limited intelligence. She had two children, both delivered by caesarean section. She was now six months pregnant. There had been amenorrhoea for the past seven years. Abdominal examination revealed no epigastric tenderness. Her pulse was rapid, of poor volume; heart sounds were good; blood pressure was 120/70. Her blood count was: Hb, 50%; colour index, 0.7; red cells, 3,500,000; white cells, 20,000 (polymorphs 85%, lymphocytes 10%, large monocytes 4%, eosinophils 1%). Three pints (1.7 ml.) of blood was transfused, following which her blood count was brought to normal limits. She remained under observation for about a fortnight, and as there were no further symptoms she was discharged and advised to attend the antenatal clinic. She was delivered of a stillborn baby by caesarean section on March 15, 1951.

She was well until June, 1951, when she was again admitted as an emergency because of another haemorrhage. This time, however, she said that she "felt fluid at the back of her throat" which she kept swallowing. She vomited about $\frac{1}{4}$ to $\frac{1}{2}$ pint (140 to 280 ml.) of blood in the ambulance. At the same time she noticed that her voice became "thick." Examination of the mouth with the help of a laryngeal mirror revealed a pale-pink, globular, and firm mass, approximately 2.5 cm. in diameter, which was attached to the tongue in the region of the foramen caecum. The mucous membrane running over it was intact, and there were numerous dilated veins which were probably responsible for the haemorrhage. The suprasternal space appeared to be much emptier than usual, and the trachea was easily palpable. Thyroid could not be felt in the neck. A lateral x-ray film of this region confirmed the existence of a well-defined swelling at the far end of the tongue. The opinion was expressed that the mass was most probably a lingual thyroid.

Her family history revealed that both parents are rather small, approximately the same height as she is. She has three sisters and one brother. One sister and the brother are of normal height, but two other sisters are not much taller than she. Her two boys are of right size and weight

for their age. She knew that she was thyroid-deficient, as her family doctor had been giving her thyroid tablets from 2 years of age. Her voice became thicker after the first haemorrhage (December, 1950), when she was six months pregnant, and during the last month or two of pregnancy she began to have difficulty in swallowing.

Because of the risk of a haemorrhage, a diagnostic test with radioactive iodine was advised instead of biopsy. A tracer dose of 100 μ c. of I^{131} was given orally, and the measurements were started six hours later with a Geiger counter. These demonstrated that the iodine was taken up by the mass in the posterior third of the tongue, and that there was no evidence of any uptake in the neck. The 48-hours urinary excretion of I^{131} was about 60%. The B.M.R. was +1%. The diagnosis of a lingual thyroid was therefore confirmed. In view of the history of thyroid deficiency, treatment with Lugol's iodine was begun. After six weeks of this treatment the swelling showed no change in size. As it was thought that surgical removal would involve a great risk, especially of haemorrhage, it was decided to treat her with radioactive iodine. Because of her age, only 5 mc. of I^{131} was administered in September, but as there was no change in size of the thyroid after three months a further dose of 20 mc. was given in January, 1952. Its urinary excretion increased to 78% in the first 48 hours. The B.M.R. taken at the same time was -19%. Even after this dose the swelling did not appear to shrink, but in fact it looked larger. She began to complain of more difficulty in swallowing, dysphonia was more pronounced, and she had two more small haemorrhages. Her weight increased slightly and her face was somewhat puffy. At the end of March her B.M.R. was -24%. It was then decided that the thyroid had to be removed surgically. Operation was carried out on May 12; the tongue was pulled well out, and, with a finger supporting the tumour from behind, access was surprisingly good. An incision was made over the anterior part of the swelling, which was then shelled out without any difficulty. Haemorrhage was easily controlled by interrupted sutures.

The post-operative course was uneventful, and she was discharged after nine days. The pathological report was as follows: "The specimen is well encapsulated, roughly spherical in outline, and about 4 cm. in diameter. The cut surface suggests thyroid tissue. Section confirms that it consists of thyroid tissue which shows marked variation in structure from active epithelial hyperplasia to involution and quiescence. There is no evidence of malignancy."

On August 12 her swallowing was good and her voice was normal. Myxoedematous signs and symptoms became, however, much stronger. The B.M.R. dropped to -38%, and she was given thyroid extract. Since then she has remained well and symptom-free.

Diagnosis

According to Goetsch, lingual thyroid has to be differentiated from adenoma, angioma, salivary tumour, fibroma, non-thyroid cysts, and the rare instances of malignancy such as sarcoma, lymphosarcoma, and carcinoma. Apart from the characteristics associated with thyroid insufficiency, which are frequent, most symptoms are caused by the pressure and obstruction of the lingual swelling. Various degrees of dysphagia, dysphonia, and dyspnoea are the most common symptoms. Occasionally, tightness in the throat or a choking sensation is the only complaint. Recurrent haemorrhage from the mouth was one of the symptoms recorded.

It is important to remember that in examination of the mouth extreme protrusion of the tongue is essential, as a small nodule can easily be overlooked. Inspection with a laryngeal mirror should not be omitted. Lateral x-ray films of the soft tissue of the face and neck will help in locating the lesion.

Nachman *et al.* (1949), and Crispell and Parson (1950), diagnosed their cases with a tracer dose of I^{131} and physical

measurements with a Geiger-Müller counter. In my case this test was found to be most helpful, and it is strongly recommended because of its accuracy, simplicity, and absence of risk. Biopsy, the obvious procedure in diagnosis of most tumours, seems to be superfluous. It is worth noting that the results of the tracer dose corresponded with those of the B.M.R. in this case.

Treatment

Lingual thyroid has never been successfully treated medically. Montgomery tried Lugol's solution without success. In some goitres thyroid extract could perhaps be of some benefit, but it is doubtful whether it would adequately reduce the symptoms caused by obstruction, nor would it eliminate a recurrence of a haemorrhage. The only other methods available are radiotherapy and surgery. It seems hardly justifiable to treat a simple goitre with high dosage of deep x rays, which would result in severe skin and mucosal reactions. Small dosage would be of little if any effect.

Treatment with radioactive iodine, mainly because of its simplicity and its effectiveness in other thyroid conditions, would seem to be an obvious method to explore. Hamilton and Lawrence (1942) have demonstrated that the administration of I^{131} to rabbits was followed by a decrease in size of the normal thyroid. Since then this isotope has been successfully used in the treatment of toxic goitres in various centres. Buck and Guthrie (1949), and Schilling *et al.* (1950) have reported cases of lingual goitres treated with I^{131} , but, although decrease in size was claimed, in neither case was the resolution of the swelling complete, and some of the symptoms persisted. Myxoedema was produced in each case. A study of the concentration of I^{131} in thyroid by radioautography revealed that the absorption of iodine depended greatly on the type of predominant tissue, and it was seldom homogeneous. It was said that thyroids with abundant colloid are more likely to concentrate the isotope. In my case no beneficial effect whatsoever was observed. Once the lingual thyroid reaches the dimensions when the patient starts to have more than occasional discomfort, and especially when a haemorrhage occurs, one is forced to remove the tumour and disregard the thyroid function in order to eliminate the symptoms.

Furthermore, it is generally accepted at present that no radioactive substance should be given in therapeutic doses to young patients. As the result of treatment with this isotope is doubtful it seems ill-advised to break this rule in the case of a lingual thyroid, which occurs mostly in children or young adults, unless there are strong contra-indications to surgery.

It was stated by some authors that surgery is technically difficult and because of anaesthetic hazards and the difficulty in controlling the bleeding it should be replaced by therapy with radioactive iodine. My experience in this single case was to the contrary. In fact, the extirpation of this goitre was characterized by absolute simplicity in every respect. Others who have also chosen the oral route in recent years found no apparent difficulties, and one wonders if modern anaesthesia and surgery would not remove the above-mentioned objections. The extraoral route was adopted by some in unusually large tumours. The most commonly expected sequel is thyroid insufficiency, which, however, can be easily controlled by giving small doses of thyroid extract. This complication might be avoided in cases of thyroids which are not encapsulated, and it seems reasonable to follow the advice given by Goetsch, that complete removal or destruction need not be attempted.

Summary

A case of lingual thyroid is reported in detail, and a short survey of the literature regarding this rare condition is made with reference to its incidence and pathogenesis. Its diagnosis and treatment are more fully

discussed, and evaluation of radioactive iodine as a diagnostic and therapeutic agent is attempted. On the basis of recorded cases in the literature and from my own experience it is concluded that I^{131} serves as the most valuable test towards the diagnosis, but in treatment the surgical approach is found to be preferable.

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Medical Memorandum

"L.E." Cells in Acute Disseminated Lupus Erythematosus Treated with A.C.T.H.

Many cases of acute disseminated lupus have now been treated with A.C.T.H. or cortisone (Montgomery and McCreight, 1949; Brunsting *et al.*, 1951; Haserick *et al.*, 1951). However, the initial detection of L.E. cells in this disease after treatment with A.C.T.H. or cortisone has rarely been recorded (Haserick, 1951; Michael *et al.*, 1951). The following case showed this phenomenon.

CASE REPORT

A single woman aged 23 was admitted to hospital on August 21, 1951, with a two-months history of a rash around the eyes, cheeks, nose, and "V" of the neck. She had taken four tablets containing phenolphthalein a few days before the rash appeared. Five weeks later she complained of malaise, nausea, shivering, pains in the limbs, and swelling of the face. The following week the rash had spread to involve the hands and feet. Her past history was uneventful, apart from an attack of "influenza" which was treated with sulphonamides at the age of 17 years. She denied being given sulphonamides on any other occasion, and gave no history of sensitivity to sunlight.

On examination she appeared seriously ill. Her temperature was 103° F. (39.4° C.), pulse 120, respirations 20, and blood pressure 110/60. A violaceous erythema extended over her face, ears, scalp, and "V" of the neck, and scaling and crusting were present on her cheeks. The swelling on her face was such that her eyes could be opened only with difficulty. The pulps of the fingers and toes showed erythematous patches, and telangiectases were present around the nail-folds. The mucous membranes of the mouth and lips were superficially eroded. Moderate painless enlargement of the axillary, cervical, and epitrochlear glands was present, and the spleen was just palpable on inspiration. No abnormality was detected in the chest or locomotor or nervous system.

Investigations.—Hb, 12.58 g. per 100 ml.; red cells, 4,200,000; white cells, 4,000, with a lymphopenia and toxic granulocytic changes; platelets, 90,000 per c.mm. (Lempert). The iliac marrow smears showed changes consistent with a severe toxic process. Plasma proteins, 7.19 g. per 100 ml.