ON
PERSISTENCE OF THE THYREOGLOSSAL
DUCT,

WITH
REMARKS ON MEDIAN CERVICAL FISTULÆ AND CYSTS
DUE TO EMBRYONIC REMNANTS.

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At the present time, judging from the literature of the subject, there is considerable confusion in the matter of fistulæ and cysts in the middle line of the front of the neck. This contribution is made with the hope that these various conditions, arising from the persistence of foetal or ancestral rudiments, may be more clearly understood. Lannelongue and Achard tend to regard all median cervical fistulæ as branchial in origin. More recently von Kostanecki and Mielecki, and Kanthack have been most dogmatic in their assertions of this as the sole mode of causation.

Several specimens of indubitable persistence of the thyreoglossal duct have come into my hands by the kind-

ness of the surgeons who treated the cases. These specimens will be described, and an inquiry instituted into the cases which have hitherto been recorded: especially because the influence of von Kostanecki and Mielecki has been such that practically all cases are now huddled together under the heading "Branchial Fistulæ or Cysts."

CASES.

CASE I. Persistent Thyreoglossal Duct; Secondary Fistula; Excision; Relapse. The specimen about to be described was removed from a well-developed male patient æt. 19, a water-side labourer by occupation.

The history is that a swelling appeared in the middle line close above the sternum, immediately after an attack of scarlatina, when the patient was four years old. This swelling was incised; and ever since there have been relapses of fistula-formation, followed by healing. On admission to Guy's Hospital (on November 27th, 1893) there was a rounded swelling, about 4 cm. in diameter, containing yellow pus, close above the sternum in the middle line; from this a rounded cord could be traced strictly in the median line up to the hyoid bone. The mass was excised by my father as high as the lower border of the hyoid bone, beyond which point the probe would not pass.

The part removed is a somewhat pyriform mass, about 7 cm. long, and tapers from about 3 cm. below to 1 cm. above in breadth and width. The lower portion consists of a mass of ragged granulation-tissue enclosing the cavity which contained the pus; from this cavity a probe can readily be passed up the entire length of the specimen.

The mass presents a groove, better marked anteriorly but traceable posteriorly, which separates it into bilateral portions, a right and a left. On section there is found to be a lumen in each portion: that on the *right* being the one through which the probe passes from the pus-

containing cavity; that on the *left* apparently ends blindly below, as will be further described. Both lumina are freely open at the upper end of the specimen. The pus-containing cavity is lined by ragged, soft, vascular granulation-tissue, and is irregularly partially divided into a number of loculi by incomplete septa of this material; it possesses no epithelial lining. The lining of granulation tissue extends for some distance up the right lumen, but at the upper part it becomes replaced by a ciliated cylindrical epithelium. The left lumen is also lined with ciliated cylindrical epithelium, except at its upper and lower extremities; at the upper end some squamous epithelium appears; inferiorly its lining ends as a number of solid cords of epithelial cells; here it is surrounded by soft granulation-tissue. A probe passed down this lumen (after hardening) entered one of the loculi of the pus-containing cavity; it would seem probable that a false passage was made leading from the somewhat widened lower extremity of the left lumen.

The bulk of the mass consists of tough lamellæ of fibrous tissue, arranged both longitudinally and circularly; some of the circularly disposed lamellæ form a well-marked septum between the two halves of the specimen. At the upper part this septum, though still well defined, becomes much thinner, and the two lumina approach one another. At the lower part the septum becomes lost in granulation tissue. This tough fibrous tissue contains very numerous bloodvessels. Surrounding the smaller vessels are collections of leucocytic cells. In the present specimen there can be no doubt that they are of inflammatory origin; they merge into the granulation tissue of the pus-containing sac, and also into the sub-epithelial collections of adenoid tissue.

At the upper end of the specimen there are numbers of striated muscle fibres, the majority of which have a longitudinal direction. Many of these are probably fibres of the sternocleidohyoid muscles. Some few, however, are cut very obliquely in transverse sections; a few

pass into the septum between the lateral parts of the mass (*vide* Plate IV, fig. 1, *mf.*), these apparently belong to the musc. levator corporis thyreoidiei.

There are also a few small nerves; it is to be noted that the fistulous track was sensitive.

The upper part of the right lumen and nearly the whole of the left lumen are lined by ciliated epithelium, and give off cœcal diverticula, so that in transverse section there is an irregularly branched cavity more or less surrounded by a varying number of other smaller cavities; the lowest part of the left lumen is a single cavity without surrounding diverticula; at its extremity it gives off a number of processes which end as solid cords of cells. The ciliated epithelium consists of long cylindrical cells with oval nuclei; the nuclei are situated at various levels, leaving a free border of finely granular protoplasm, which is limited by the hyaline refringent border usual in ciliated cells. Each cell possesses numerous cilia (fig. 2).

Besides the shrinkage spaces between juxtaposed cells, there are a few clear spaces due to the presence of goblet cells loaded with mucous material, and provided with nuclei.

Scattered here and there wandercells are seen making their way through the epithelium. One or two of the diverticula are lined by nonciliated mucous cells, cylindrical in shape with peripheral nuclei; in appearance they recall somewhat the cells of the mouths of the gastric glands.

Beneath the epithelium is a variable layer of fine fibrous tissue, for the most part studded with leucocytes. In many places (*vide* fig. 1, *l.*) these leucocytes are so numerous that the tissue becomes like adenoid tissue.

Here and there, where the epithelium is stretched over projecting nodules of adenoid tissue, it becomes much thinned out, or even lost altogether; thus showing the probable history of the lower portion of the right lumen, where only a granulation-tissue lining remains.

Kanthack considers the adenoid tissue about the foramen cœcum to be of a chronic inflammatory origin, as it is not present in the foetus or young infant, and is largely increased in cases where the tonsils are enlarged ; such, no doubt, is its nature in the present case.

In the sections taken close to the extreme upper end of the specimen there are a few patches of squamous epithelium forming the lining of diverticula of the left lumen (*vide* fig. 3). This is evidence that some part of the ductus lingualis is also persistent. Further evidence is afforded by the recurrence of the fistula, with which the patient left the hospital at his own desire.

He was readmitted on 3rd February, 1894. On February 9th, under an anæsthetic, a probe was passed as far as the lower border of the hyoid bone, and the fistulous track was again excised to that level. An unsuccessful attempt to pass a probe down the ductus lingualis from the mouth was also made.

Three days after the operation stringy mucus exuded from the upper part of the wound. An injection of iodine solution was ordered. On the 18th, when the injection was given, the patient suddenly coughed and said he could taste the fluid, with the sensation of his mouth being "all crinkled up." A few days later an unsuccessful attempt was made with a solution of methyl blue and quinine, in order to test the question of patency, both objectively and subjectively. The patient refused further treatment, and left the hospital. The mucus that exuded was examined microscopically, and found to contain squamous epithelial cells ; no cylindrical cells were observed. The portion excised at the second operation was found to be devoid of epithelial lining.

The commencement of the trouble with scarlatina is extremely suggestive that infective material from the faucial region made its way down a patent canal.

It will be remembered that the foramen cœcum is lined by squamous epithelium. I have not been able to discern any transition to the squamous type in the right lumen.

This specimen differs from those *branchial* fistulæ mentioned by Baumgarten, Neumann, &c., in which the epithelium below is squamous (derived from the sinus præcervicalis of His, and the epiblastic branchial pouch), and above is ciliated; the positions being reversed.

In a series of sections taken near the upper part, in the right side on its mesial aspect (*i. e.* close to the septum), is a small tract of thyroid gland follicles with their colloid contents (*vide* fig. 4). These follicles are not enclosed in a capsule as are the accessory glands, but are intimately associated with a diverticulum, or groove in the right lumen.

Neither at the operation, nor from microscopical examination of snips of the lower parts of the specimen, was any evidence obtained of a connection with the thyroid body, or a lobus pyramidalis. In the lower part of the specimen there are no thyroid gland follicles.

It seems certain that this specimen owes its origin to the *thyreoglossal duct*. This duct, by the researches of His and others, has been shown to bifurcate at its extremity, and the two lumina in this specimen doubtless belong to the forked portion of the duct. The point of bifurcation in this case is above the lower border of the hyoid bone. In Marshall's case¹ it is figured at a lower level. In His's case this point is at the upper part of the thyrohyoid membrane. It would appear from these cases that the locality of the bifurcation is somewhat variable. Besides the anatomical position about the median plane, and the bilateral structure, the presence of thyroid gland follicles is convincing evidence in favour of development from the ductus thyreoglossalis.

The investigations of von Kostanecki and Mielecki, supported by Kanthack, have introduced a further complication in the diagnosis of cysts and fistulæ of the middle line of the neck; namely the possibility of their origin from the *sinus præcervicalis* of His, a cavity which is formed by the overlapping of the hyoid arch in front of

¹ There is some reason to doubt the genuineness of this case, *vide infra*.

the third and fourth visceral arches, and includes the apertures of the external pouches of the never completed second, third, and fourth branchial clefts. As the sinus præcervicalis extends across the middle line, it is obviously possible that median fistulæ may result from the persistence of its aperture, which normally becomes obliterated (true congenital fistulæ present at birth appear to be exceedingly rare, if indeed there are any really authentic cases); or from the nonobliteration of the included portion, and the secondary formation of a fistula owing to distension of the sac. These authors assert that the only difference that there is between *median* and *lateral* fistulæ is the median position of the aperture of the former.

In median fistulæ it is clear that the diagnosis of the persistence of the sinus præcervicalis can only be made when squamous epithelium occurs about the aperture of the fistula, as this structure is derived from the epidermis, unless a transition from squamous to ciliated epithelium is hypothesised.

It therefore appears to me that several of the cases which von Kostanecki and Mielecki have tabulated as median branchial cysts and fistulæ are in reality of thyreoglossal origin, as will be discussed more fully later.

It is probable that all such cases, in which the lumen passes up in the *mid-line* behind the hyoid bone, and is lined with ciliated epithelium, originate by the persistence of some portion of the ductus thyreoideus or lower part of the ductus thyreoglossalis. Confirmatory evidence may be obtained either by dissection or by microscopical examination; in the former case, connection with a *lobus pyramidalis*, or with the *ductus lingualis*, or with *accessory thyroid bodies*, or by the passage of probes or injected fluid up to or through the base of the tongue (*foramen cæcum*) in the middle line; in the latter case, by the discovery of a bifurcated condition, or of thyroid gland follicles, or of squamous epithelium in the upper end replacing the ciliated epithelium.

Difficulty arises in cases such as that described by

Raymond Johnson (Case 1) in which a probe could be passed "more than half an inch above the hyoid bone," the lumen being lined by "a thick layer of stratified epithelium having in parts a distinct papillary arrangement. There are no glands." Kanthack describes numerous racemose glands opening into the ductus lingualis (foramen cœcum). As these mucous glands are abundant just above the hyoid bone, it is reasonable to suppose that they would also be found below it in fistulæ derived from the thyreoglossal duct. Such a condition has yet to be described. In the present state of knowledge it does not appear to be possible to distinguish whether the condition arose from the permanence of the *ductus lingualis*, or of the *sinus præcervicalis* alone, without such minute dissection as could only be undertaken post mortem. The extent to which a persistent sinus præcervicalis may extend up above the hyoid bone is unknown. Hence, though suggestive of such an origin, it does not appear scientific to assign this case definitely to either category.

The presence of squamous epithelium lining the lower part of a fistula must also always suggest the fallacy that it has grown in over bare granulations from the external epidermis, and is therefore quite a secondary condition, having no relation whatever to the transient sinus præcervicalis. In many recorded cases the funnel-like aperture of the fistula was deepened by the traction exerted by movements of the hyoid in swallowing, &c.

CASE II. *Persistent Thyreoglossal Duct; Excision; Cure.*—This specimen was removed in 1889, by my father, from a female child six years old.

There was a recalcitrant fistula in the middle line below the hyoid bone. After excision and cauterisation the wound healed well, and the patient has had no further trouble (February, 1894).

Unfortunately the specimen was not carefully hardened for histological study, so that sections are not

quite so good as could be wished. It is now 1 cm. in length; before shrinkage it was much longer. A probe readily passes through the fistulous track. Microscopical transverse sections at the middle of the specimen show two distinct lumina, each with a differentiated fibrous sheath blending into a common sheath. The connective tissue is less dense and hard than in Case I. One lumen is large, and through it the probe was passed; the other is smaller ($\cdot 2$ mm. $\times$ $\cdot 05$ mm.). Both are lined with the remains of cylindrical epithelium; the preservation of the specimen is not sufficiently good to allow cilia to be seen, whether there were any must be left open to doubt.

Transverse sections at the extreme upper end show the two lumina; [the larger ($1\cdot 1$ mm. $\times$ $0\cdot 1$ mm.) has diminished so that it just occupies the field of a half-inch objective].

Between the two a third cavity can be traced connecting the others.

In all the three lumina the epithelium is partly cylindrical and partly squamous.

There are no circumferential diverticula as in Case I, nor are thyroid follicles seen.

This specimen is to be regarded as a persistent thyreoglossal duct because of its median position, its forked lumen, and the squamous character of the epithelium at the upper extremity.

CASE III. Persistent Thyroid Duct; Isthmian Bronchocele; Excision; Recovery.—For this specimen, and for the permission to publish it, I am greatly indebted to the unfailing courtesy and kindness of Mr. Jacobson, under whose care the patient was. Nowhere before, I believe, has a similar specimen been described; unfortunately only a small piece is extant, which was preserved and embedded in paraffin by my late brother.

Clinical aspect.—A girl, $\text{æt. } 18$, was under Mr. Jacobson's care for a goître in 1888. The tumour extended from the lower border of the thyroid cartilage to within half

an inch of the sternum ; laterally it passed under the right sternomastoid and reached the anterior border of the left.

At the operation the isthmus alone was found to be affected. "A process running upward on to the thyroid cartilage" had to be cut. The connection with the left lateral lobe was merely fibrous ; some healthy gland tissue was removed from the right lateral lobe with the tumour.¹

The tumour was "almost entirely colloid with cysts of large size." The microscopical sections show several cysts, the largest being about 1 cm. in diameter. The cysts are of three kinds :

1. Cysts lined with *cubical* epithelium, empty or with finely granular contents.

2. Cysts lined with somewhat *cylindrical* epithelium, in two or three layers of cells, which secrete mucoid material. In some of these the secretion has remained discrete, and forms long curved threads of granular material more or less wound up inside the cyst (fig. 5). The condition may be compared to the cylinders of oil paint which exude from the collapsible tubes used by artists. Here and there, there are indications of the presence of cilia, but they are not quite sufficiently well preserved to be made out definitely.

3. Cysts lined with *cylindrical ciliated* epithelium. The epithelium is similar to that described in Case I, so that fig. 2 would accurately represent it, although actually drawn from the other specimen. The contents, where present, are a somewhat coarsely granular material with débris of cells.

The stroma consists of a fine loose reticulum of fibrillæ, with sparse branched and rounded cells. The meshes of the spongework are filled with an infiltration of colloid material, which in some parts forms considerable cell-less masses. In appearance this colloid material resembles the contents of thyroid follicles ; but it is loose in the reticulum, and not contained in epithelium-lined spaces, a condition not infrequent in ordinary goïtrous tumours. The data

¹ Before operation the right lobe was thought to be involved.

for diagnosis in this case consist of (1) the ciliated cystic cavities; (2) the clinical note that at the operation a process was found to run upon the thyroid cartilage; (3) the undoubted connection with the isthmus of the thyroid body.

It does not appear to be possible that the ciliated spaces can be other than unconverted portions of the lower part of the ductus thyreoideus. Whether or no they were connected with a patent canal, extending towards, or actually to the foramen cœcum, must remain unknown.

Speculations as to the association of the persisting rudiment with the colloid infiltration of the isthmus would be at once idle and useless.

This is the only instance I know of, in which there exists pathological enlargement of the isthmus with persistence of the thyroid duct.

Streckeisen describes (Fall 104, p. 234) a universal cystic goître in which there was a ciliated cyst the size of a pea, at the attachment of the musc. hyogl. to the thyroid gland.

REMARKS.

The subject of mid-line cervical cysts and fistulæ is scantily treated of in the text-books on surgery,¹ by English, French, and German authors; and not only is this the case, but the information given seems hardly satisfactory. Such must be my excuse for entering more fully upon this interesting subject—a subject, moreover, which has often been treated with prejudice.

The following classification includes all the possible forms of *cysts and fistulæ of congenital origin in the mid-line of the neck*; the basis is upon embryological grounds.

1. DERMOID CYSTS AND FISTULÆ (epiblastic):

- A. Those derived from Ductus Lingualis.
- B. Those derived from Sinus Præcervicalis.
- C. Those derived from Sinus Præcervicalis with epiblastic portion of gill pouch. (The epiblastic and

¹ The best account is perhaps that of Broca.

hypoblastic portion of the gill cleft, normally, never communicate in the development of the human embryo.)

- d. Those of independent origin, unconnected with any normal embryonic rudiment.

2. MIXED DERMOID AND MUROID CYSTS AND FISTULÆ (epi- and hypoblastic):

- A. Those derived from Ductus Thyreoglossalis.
B. Those derived from Sinus Præcervicalis with Gill-cleft, including the hypoblastic portion.

3. MUROID CYSTS AND FISTULÆ (hypoblastic):

These, when in the mesobranchial region, that is, limited to the middle line, can owe their origin solely to the Ductus Thyreoidæus.

1. DERMOIDS.¹

A. *Those derived from Ductus Lingualis*.—A very considerable number of cases of dermoid cysts of the floor of the mouth have been collected by Lannelongue and Achard (Chap. 'Kystes Dermoides du Plancher Buccal,' p. 230). These are all above the level of the hyoid bone. According to their structure they appear to be of two kinds: (1) those with simple squamous epithelial lining, whose contents are only sebaceous matter; and (2) those in which hair-follicles, hairs, and sebaceous glands, and sometimes papillæ are found. As instances of the former we may cite the cases described by Barker, while an instance of the latter is to be found in Stephen Paget's case, where the lining of the cyst wall is described as "true skin with a few fine short hairs," and some pigmentation—the anatomical attachments not being given. A few cases are recorded in which there was a distinct attachment to the mandible (*e. g.* Ozenne, Obs. 2). It appears, then, that two forms of dermoid cysts occur here, viz. those derived from *ductus lingualis* and those of *independent*

¹ Some authors suggest the term "epidermoids." However, the word "dermoid" is so well understood to mean squamous epithelial-lined cysts, that the refinement appears to be hardly necessary.

origin. The ductus lingualis is so remote from a region producing hairs, and, moreover, such glands as it possesses being mucous in structure, we should be disposed to rank all dermoid cysts of the floor of the mouth supplied with hairs and sebaceous glands as of *independent origin*; and conversely, at any rate, many of those with simple walls as derived from the *lingual duct*. Unless definable attachment to the hyoid bone or mandible exists, there would appear to be no criterion to distinguish them; except that, in the lingual duct cases, at all events, the genio-hyoglossi muscles are separated, whatever be the state of these muscles in the other conditions (*vide* Ahlfeld's case of ectopia linguæ). Apart from the above remarks on their ontogeny, the full accounts of Ozenne and Barker, and Lannelongue and Achard, render further discussion superfluous here. It is to be noted that they are above the hyoid bone, and that, though usually in the middle line, it is possible for them to assume a somewhat lateralised position by muscle pressure, as Barker has pointed out. This author suggests the sinus præcervicalis as a source; it is uncertain whether this structure can rise above the upper level of the hyoid, probably it never does.

Streckeisen (Fall 102) has found a little dermoid cyst between the hyoid and the upper end of the pyramis thyreoidea, rather in front of, but attached to the latter. It seems impossible to place this in either category (sinus præcervicalis or ductus lingualis).

B & C. *Those derived from Sinus Præcervicalis with or without the Epiblastic Branchial Pouch.*—These are situated below the hyoid bone, and are attached to it. They are lined by squamous epithelium, and are often supplied with hairs and sebaceous glands. Fistulæ appear to be secondary to rupture of a cyst in a large majority of the cases.

Though it is not known at what level the ductus lingualis ceases, the evidence (His) is in favour of its termination being situated above, or at the thyrohyoid membrane.

Raymond Johnson has described a case of middle-line fistula lined with pavement epithelium, in which a probe could be passed upwards "more than half an inch above the hyoid bone,"—presumably above its lower border. It seems probable, therefore, that this case is to be considered as a persistence of the sinus præcervicalis, rather than of the ductus lingualis. Between sinus præcervicalis dermoids, and independent hypohyoid dermoids there is no diagnostic criterion; the close relation of the sinus to the hyoid bone, to the growth of which it largely owes its existence, would tend to make us regard all examples, with attachment to this bone, as owing their development to this sinus.

The extensive bibliography collected by Lannelongue and Achard concerning median hypohyoid dermoids, suggests that it will be sufficient to notice the few cases in which the branchial rudiment also remained. This is shown by the lateralised passage from a median aperture.

CASE 1 (Berkeley Hill).—F., æt. 15. Small sinus opening in mid-line by thyroid cartilage. Probe passes half an inch upwards towards angle of jaw. Initially tumour. No histological examination recorded.

CASE 2 (Lefort).—M., æt. 15. Aperture in mid-line at level of hyoid. History of tumour when eleven years old. Track of sinus obliquely upwards, backwards, and to the right towards great cornu of hyoid bone. Excision; persistence of fistula; eventual cure by electrolysis. No histological examination is recorded.

In the absence of knowledge of the microscopical characters it is impossible to assign these definitely here, or under the next heading (Mixed Dermoid and Mucoid).

2. MIXED DERMOID AND MUCOID.

A. *Of Thyreoglossal Origin*.—These are more conveniently treated under the next section. For although dermoid lining occurs above, the main part of the aberrant cavity possesses a mucoid lining.

B. *Of Branchial Origin* (lateral track with median aperture).—So far as I am aware, no such case has been

as yet investigated with the microscope. But it can hardly be doubted that dermoid lining existed in the peripheral part, and mucoid in the proximal part, in the following cases, for the reason that the fistulous track was demonstrated to be pervious into the pharynx. The only cases I know of in which the histology was inquired into, and the mixed condition found, were fistulæ with lateral aperture (*vide* Cusset, Baumgarten, and Neumann).

CASE 1 (Meinel).—F., æt. 34. Median aperture half an inch above sternum. Probe goes upwards and to the right. Pervious to food from pharynx.

CASE 2 (Jenny).—F., æt. 24. Aperture exactly in middle line, opposite lower border of thyroid cartilage. Probe passes up and back to right angle of mandible. Track pervious to pharynx by injections. Tumour first; spontaneous rupture; fistula formed.

CASE 3 (Berg).—M., æt. 21. Aperture in mid-line above incisura cartilaginis thyreoideæ. Probe passes 1 cm. rightward. Pervious to pharynx by injection. Initially tumour. No histological examination.

3. MUCOID.

In the mesobranchial region of His, *i. e.* the median region limited laterally by the gill clefts,—there exist the following structures capable of giving rise to cysts other than dermoids.

1. The acinous glands opening into the ductus lingualis.
2. Remnants of the hypoblastic portion of the thyreoglossal duct—the ductus thyreoideus.

There are a few isolated cases of mucous cysts of the tongue situated in the middle line close to the mucous membrane. Hadden records one, but gives no histological description. There is another specimen in the Guy's Hospital Museum, situated between the position of the foramen cœcum and the epiglottis. Albert also records a case in which there were two cysts in this region. The description, however, is very meagre, and no histological examination is recorded.

A few cases are recorded as accessory thyroid glands

in the tongue (Rushton Parker, Hickman, Butlin, and Wolf). These have been adversely criticised by Kanthack, who considers them adenomata of tubular glands; and he has reason on his side, for it is impossible to recognise thyroid tissue unless the typical colloid-containing follicles are present. Butlin gives no indication of any such structure in his figure. However, Warren records a case in which typical thyroid gland structure was found microscopically.

Typical accessory thyroid glands ("glandulæ hyoideæ," &c.) have been carefully described, especially by Streckeisen in a most excellent paper (*vide* also Zuckerkandl, Kadyi, and Merten); those that occur within the trachea and larynx (Paltauf) do not concern us here, as they are lateral, and appear to be offshoots from the lateral thyroid rudiments, derived from the region of the fourth branchial bar.

True accessory thyroid glands may occur *above, in front of, behind, below,* and actually *within* the hyoid bone.¹ Similarly, ciliated cysts may occur in the same regions, and many such cases were discovered by Streckeisen in the post-mortem room. Neumann describes an instance simulating a ranula. The examination of a portion of the cyst-wall showed that the lining consisted of ciliated cylindrical epithelium. He recommends removal of a part of the cyst-wall in all cases of suspected ranula for obvious reasons. The rationale of these ciliated cysts is not far to seek. Bochdaleck jun., Krause, Streckeisen, and Rosenberg have found ciliated cells at the lower end of the ductus lingualis, and I can confirm this from personal observation in the post-mortem room. The ciliated cylindrical lining is found both at the termination of the ductus as well as at the openings of the ducts of the mucous acinous glands.

Kanthack was unable to find other than squamous

¹ The terms *supra-, infra-, subhyoid, &c.*, used by some authors are avoidable hybrids; a classical friend tells me there is no objection to the words *epihyoid, hypohyoid, retrohyoid, &c.*

epithelium in his researches ; this, no doubt, was owing to his imperfect method of only examining microscopically cases in which the presence of a duct was suspected under a dissecting lens. Streckeisen, on the other hand, decalcified the hyoid bone and prepared sagittal microscopical sections of the whole region. In a case that I examined it was impossible to detect the presence of the extremely minute duct in the hyoid region under a dissecting lens ; but it became perfectly obvious even to the naked eye upon injection of watery Berlin blue solution from the foramen cœcum.

It is possible for one of these epihyoid mucoid cysts to form a median fistulous track opening above the hyoid bone. Houel records such a case (which von Kostanecki wrongfully quotes as "subhyoid"¹) ; microscopical examination revealed the presence of a ciliated epithelium.

Streckeisen and Schlange describe enclosure of the duct by the hyoid bone. Out of the few cases I have examined I found one in which this condition was present.

Dermoid and mucoid cysts may be present together. Ozenne has described one case (Obs. I, p. 281) in which a hair-containing dermoid was found in the epihyoid region, and below and behind it there was a little mucous cyst ; he does not give any account of the histology of the latter.

It will be remembered that the ductus thyreoglossus bifurcates at its extremity, that is, the thyroid duct (or hypoblastic portion) ; indeed by some authors this is considered as an originally paired structure. I have already adverted to the uncertainty of the position of the point of bifurcation. It appears that the two lumina are persistent in the first two cases described above. This condition is either a rare one, or else the cases recorded in the literature have not been thoroughly examined ; as indeed is inevitable in those which have been treated by cautery, caustics, or electrolysis. Ribbert's case (*vide infra*) seems to be almost the only case hitherto recorded in

¹ Houel's expression is "sushyoidienne."

which more than a single lumen exists ; he considers it of branchial origin.

The diagnosis of fistulæ and cysts derived from the ductus thyreoideus depends upon—

1. The position strictly in the *middle line*.
2. The presence of *ciliated cylindrical epithelium*, which may be more or less replaced by squamous as the ductus lingualis is approached.
3. Permeability to injections or probes through the *foramen cæcum* ; this is rare in the adult, although injections have been successful in the young, interruption of the continuity of the lumen usually taking place at the thyrohyoid membrane owing to movements and tension (His).
4. The presence of *paired lumina*, the remains of the original bifurcation.
5. The presence of *thyroid gland follicles*, or accessory glands intimately associated with the duct.
6. Connection with the *thyroid gland* at the lower extremity.

Of these points the first is absolutely essential. The second is probably almost equally necessary, with the reservation that it is possible that the ductus lingualis (squamous lining) may reach below the hyoid, without persistence of ductus thyreoideus. The other four points are of importance, if present. The presence of a lobus pyramidalis is so common that it would be unsafe to argue from the coincidence unless a given cyst or fistulous track has some essential connection with it.

It is to be noted that the cyst or fistulous cord moves with the hyoid bone on deglutition in nearly all recorded cases—a condition which is a prerequisite of thyroid gland tumours according to textbooks on surgery. The hypo-hyoid dermoids also move with the hyoid bone.

The level of the aperture of a fistula is of no importance. The thyreoglossal duct, descending from the level of the first and second branchial clefts, may have a fistulous aperture anywhere, from a little above the hyoid

bone to the sternum. Since the early days of the recognition of branchial fistulæ, attempts have been made to mark out the positions upon the neck where the second, third, and fourth clefts will open. It would hardly be worth while referring to the matter here, were it not that such diagrams have been passed on from one textbook to another. A glance at the embryological history of the neck shows how futile such attempts must be. The only means of distinguishing the several clefts is by their internal apertures in the pharynx and by their relations to the nerves, as has been done in a few cases.

The following cases appear to belong, with great probability, to the category of cysts and fistulæ derived from the ductus thyreoideus.

1. EPIHYOID REGION.—Houel's and Neumann's cases already referred to above, as also those observed by Streckeisen. Delens records many probable cases, but gives no histology (*vide* postscript).

2. HYPOHYOID REGION.—A. Several cases of median fistulous tracks, with ciliated lining, passing *upwards* have been recorded (Jouon, 1864; Nélaton, 1866; Faucon, 1874; Affre, 1875; Köhler, 1876; Ribbert, 1882; Tillaux, 1885; Arndt, 1881; Strübing, 1892; Schlange, 1893).

All these cases are classified by their authors as branchial fistulæ; Strübing alone suggests the possibility of a thyreoglossal origin. There is no evidence that branchial clefts can exist in the middle line; though, by means of the squamous-lined sinus præcervicalis, they may acquire a median aperture. In Affre's case, there is no possibility of any remnant of sinus præcervicalis, since the exuberant rim of the fistula was shown to be coated with ciliated cells. Ribbert's case appears to be very similar to the one I have described above (Case I). Two tubes were demonstrated by the naked eye; microscopically a third was found between the other two. (It is to be remembered that the ductus thyreoideus is described as breaking up rapidly into a number of anastomosing

branches, so that the original bifurcation soon becomes imperceptible.) The lumina of these tubes were found to be complicated by folds, and by cæcal diverticula. The lining membrane consisted of cylindrical cells, for the most part ciliated. Squamous epithelium was also present, "especially in the smaller tubes;" but the description is not sufficiently clear as to the exact disposition of this structure.

Arndt regards his case as a palingenesis from the hypobranchial groove of *Amphioxus*; ciliated epithelium occurs, however, in the thyroid gland of a slightly less remote ancestor, viz. the ammocete larva of the lamprey. Though embryologists have not described the appearance of cilia in the development of the Mammalian thyroid body¹, yet we may consider ciliated epithelium a normal feature of the ductus thyreoideus when any of it persists at the end of the ductus lingualis (Bochdaleck, &c.).

His and Marshall have described cases as persistent thyreoglossal ducts, their diagnosis being based upon naked-eye anatomy. His made no microscopical examination. Marshall, to whom I am much indebted for showing me his specimens, was unable to obtain sufficiently satisfactory microscopical sections to put his specimen beyond all doubt.

B. Cases of median hypohyoid fistulous tracks or mucous cysts in which no microscopical examination was made are recorded by Dzondi (1829, cases 2 and 3); Riecke (1845), Luschka (1848), Larrey (1852), Chamart (1850), Forget (1852), Hamilton (ten cases) (1870), Bellouard (1875), de St. Germain (1875), Gosselin (1876), Köhler (1876), Cusset (1877), Fischer (1880), Demons (1885), Volkenrath (1888), König (1889), Karewski (1893).

Of these, the series of cysts described by Hamilton are the most interesting; unfortunately he gives no histological account, but as they all contained clear mucoid material, they were all probably of thyroid duct origin, though he

¹ Wölfler figures cells from the teased middle thyroid rudiment of a 15 mm. dog embryo, which appear like ciliated cells, but he gives no description.

regarded them as bursal in causation, and their contents synovial.

c. Cases in which a median fistulous track led *downwards* towards the sternum. Of these I have been able to collect four, namely, Pooley (*vide* Hamilton) (1870), Roth (1878), Berg (1883), Cusset (1886).

Pooley's case (recorded by Hamilton) is most interesting. Pooley attended a primipara and immediately after delivery the child was noticed to have an abrasion on the front of the skin of the neck, in the median line, over the thyrohyoid space, "as if a scratch with the nail." There was then found to be a little sinus in the mid-line which would admit an "ordinary" probe half an inch downwards and backwards, and a second aperture just above it, one eighth inch deep, as tested by the same appliance. "The openings had the ordinary mucous appearance of such orifices." Whether this track was opened traumatically is left doubtful. The child died when two months old, and no dissection was permitted, nor were any scrapings examined microscopically.

Cusset's case was not examined histologically.

Roth had the opportunity of dissection in a subject with mid-line cervical fistula, down which a probe passed as far as the sternum. Flakes obtained by probe showed both squamous and ciliated cells. Transverse section at middle of the specimen showed cylindrical-celled lining and mucous acinous glands. It was considered as a product of the fourth branchial pouch; and no mention is made of connection to hyoid bone or thyroid body.

Berg found both cylindrical and squamous epithelial cells in the flakes brought away by probing. Examination of the excised track showed a duct, lined with ciliated cylindrical epithelium, which was surrounded by eight or more rounded "acini," also lined with cylindrical epithelium. He believes it to be derived from the fourth gill cleft, "which is not to be doubted when the position and the presence of ciliated epithelium are considered"—a statement which invites challenge.

In the light of those cases I have examined, I should regard all these as almost indubitable instances of persistent ductus thyreoglossus. For it is extremely difficult to imagine how a branchial cyst could come to occupy such a position ; whilst such a ciliated cyst as Streckeisen found attached to the thyroid isthmus, or Case III recorded above, if it happened to rupture, could easily give rise to a similar condition.

PROGNOSIS AND TREATMENT OF THYROID DUCT PERSISTENCE.

Whilst it is true that some recorded cases of cyst have disappeared *suâ sponte*, yet as a general rule the prognosis and treatment are interdependent. It must also be remembered that a mere puncture of one of these cysts may give rise to a troublesome fistula ; for instance (Larrey's case), a soldier was troubled with the pressure and friction of his tunic against the "tumour," so he pricked it with a needle. After this he had a secreting fistula, for four years under surgical treatment ; it was eventually cured by iodine injections.

Cases have been treated by incision, injection, caustics, actual cautery, electrolysis, and excision ; in some, success attended the treatment more or less immediately ; in others the patient was lost sight of—with his fistula. In some recorded cases the perseverance both of the patient and of the surgeon is remarkable. Thus Gosselin describes a case which was severally treated by incision and excision ; then nearly all the known caustics were tried in succession, at intervals of some few weeks for a period of two years. The patient still had the fistula patent, and M. Gosselin was still hopeful that caustics would eventually cure him. It is obvious that such unsuccessful attempts are due to incompleteness of the interference with the epithelial-lined track, the reason whereof is the narrowness of the lumen as it passes behind the hyoid bone, or indeed actually perforates that bone. Recognising this fact, Schlange has cut through the hyoid bone and by "forcibly retracting the two halves,"

obtained a good view and room enough to deal with the tissues in the middle line far up towards the root of the tongue. He states definitely that no trouble was experienced from this procedure, either at the operation or after it. Indeed, in one case he removed a piece one centimetre broad from the middle of the hyoid bone. The extraordinary obstinacy of some of these fistulæ in the past, together with the sound basis of anatomical conditions, as well as the absence of disagreeable results (immediate or remote), make it probable that section or resection of the hyoid bone will be resorted to without compunction in the future, when less complete measures have failed.

From attempts to trace the ductus lingualis in the post-mortem room, I should be inclined to think that the injection of a saturated solution of Berlin blue into the fistula would be useful before proceeding to excise. The blue precipitates and leaves an unprobeable and otherwise invisible canal perfectly obvious as a thin dark blue line. By this method it would be easy to discern whether the upper limit of the fistula had been reached.¹

FURTHER REPUTED ORIGINS OF MEDIAN CYSTS AND FISTULÆ.

Besides the above conditions, the following other reputed causes of median cysts and fistulæ remain to be shortly considered.

1. *Enlargement or Cystic Degeneration of Præepiglottic Mucous glands* (Nélaton). It has been adversely criticised that the thyrohyoid membrane must have become perforated for these to present below the hyoid bone. There is no evidence in favour of this view, except in so far as that the "glands in front of the epiglottis" and the glands opening into the lingual duct are identical.

2. So-called *Tracheal Herniæ*. Several cases have been collected by Eldridge; none of them appear to be entirely

¹ Any excess of the blue can be removed by a weak alkaline solution.

satisfactory—moreover air has been blown through fistulæ, e. g. Fischer's case. The common paired condition suggests ductus thyreoidæus, or bilateral persistence of branchial pouches as the real condition. Until a dissection has been performed these tracheal herniæ must be considered doubtful.

3. *Enlarged Bursæ about the hyoid bone, thyroid cartilage, and geniohyoglossi.*

Several bursæ are described in these situations, and these have been pronounced guilty of cyst-, and fistula-formation.

Verneuil's classification is the one most frequently adopted, and which has crept into most text-books, English and foreign, viz.

1. *Bursa mucosa infrahyoidea seu thyreochoidea* (Bourse séreuse de Boyer).

2. *Bursa mucosa suprahyoidea.*

3. *Bursa mucosa thyreoidea.*

Notwithstanding the common belief that these structures are the causes of median-line cysts and fistulæ, I have been unable to find a report of a single authentic case.¹ The merits of the belief are well characterised by Verneuil's statement in 1877, at the discussion on Houel's case, "on devra s'appuyer comme caractère fondamental sur l'épithélium à cils vibratiles qui les tapissent ; alors que les autres (fistules branchiales) ne présentent que de l'épithélium pavimenteux." A bursal cyst with ciliated lining would indeed be a prodigy !

In conclusion, it is hoped that the evidence given that the thyreoglossal duct may persist is sufficiently convincing to prevent the reappearance of any such denials as have been published in the past.

The characters of three cases have been described, and

¹ The case recorded by Verneuil of epihyoid "bursal" cyst (op. cit. infra, p. 192) is hardly satisfactory, as the absence of epithelium attached to the cyst-wall is amply accounted for by the macerated condition of the tissues of dissecting-room subjects.

the means whereby they may be identified have been pointed out. It is suggested that many of the cases of "*median branchial cervical fistula*" which have been published are in reality cases of *persistent ductus thyreoglossus*.

The thorough examination of all specimens met with in the future is much to be desired ; more especially to determine whether bursal enlargements, which have been described about the hyoid bone, are not pathological creatures of the imagination.

Postscript.—Since the above paper was written, Dr. Buscarlet has kindly sent me a copy of his thesis. He records (1) a case of median epihyoid fistula lined with ciliated epithelium (*vide* pp. 215 and 217 *supra*). (2) A case of median hypohyoid fistula lined similarly. (3) A case of epihyoid dermoid fistula lined with squamous epithelium (*supra*, p. 210).

All three appear to be derived from the ductus thyreoglossus ; the former two from the ductus thyroideus, the latter from the ductus lingualis. It is therefore hardly desirable to call them "*Fistules Branchiales*," since the author admits their origin from the "*Canal de Bochdaleck*" or thyreoglossal duct of His.

In each case operative interference was practised more than once, owing to recurrence of the fistula.

Messrs. Whitehead, Hardie, and Morton have each recorded cases of mucoid fistula in the median hypohyoid region. In one case (Hardie's) ciliated epithelium was found ; the other cases were not examined microscopically. I have to thank these authors for kindly replying to my enquiries.

The specimens obtained in Cases I and II have been presented to the Museum of the Royal College of Surgeons.

Case I formed the basis of a clinical lecture by my father ; this is reported in the '*Clinical Journal*,' vol. iii, 1894, p. 150, and in the '*Guy's Hospital Gazette*.'

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(For report of the discussion on this paper, see 'Proceedings of the Royal Medical and Chirurgical Society,' Third Series, vol. vi, p. 83.)

DESCRIPTION OF PLATE IV,

On Persistence of the Thyreoglossal Duct; with Remarks on Median Cervical Fistulæ and Cysts due to Embryonic Remnants (HERBERT E. DURHAM).

FIG. 1.—Transverse section ($\times 8.4$) at the upper end of the specimen removed from Case 1. (3 in. obj. cam. luc.). On the right side (R) are seen three cavities representing the right lumen and two diverticula. On the left side (L) there are several cavities representing the left lumen and its diverticula. *art.* Arteries. *cd.* Débris of cells and mucus. *d.* Septum between the right and left sides. *F.P.* False passage made for purposes of orientation. *l.* Lymphoid or adenoid tissue, represented diagrammatically with fine dots. *mf.* Striated muscle fibres. *s.* Situation of the thyroid follicles.

FIG. 2.—Ciliated cylindrical epithelium from one of the duct cavities. ($\frac{1}{8}$ in. obj. cam. luc.)

FIG. 3.—Squamous epithelium from one of the diverticula of the left lumen. ($\frac{1}{8}$ in. obj. cam. luc.) *ct.* Connective tissue.

FIG. 4.—A thyroid follicle from the neighbourhood of the right lumen. The colloid contents show the hemispherical depressions commonly seen in sections of the thyroid body. ($\frac{1}{2}$ in. obj. cam. luc.)

FIG. 5.—Portion of cyst in Case 3, showing the threads of secretion (*m.*) attached to the epithelial cells; also the hyaline interruption between the granular thread and the surface of the cells (*ep.*). The loose reticular tissue (*rt.*) with colloid infiltration (*c.*) is seen below. (1 in. obj. cam. luc.)