A PHARYNGEAL POUCH OF LARGE SIZE REMOVED BY OPERATION

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*The Clinical part of this case is described by Mr. Godlee, the
Pathological for the most part by Mr. Bucknall.*

Received 8th April—Read 14th May, 1901

THE patient whose case forms the subject of this paper came under Mr. Godlee's observation accidentally on May 18th, 1900. He had then a swelling, which looked exactly like an enlarged gland, on the left side of the neck at the level of the hyoid bone in front of the sternomastoid. It was, however, soft, and on percussion gave a tympanic percussion note, and was clearly anchored to the top of the larynx, the movements of which it followed during deglutition. On squeezing it a squeaking

sound was produced, and a most uncomfortable sensation was experienced by the patient, which shot up to the ear, and which lasted for some days. The pressure caused an obvious diminution in the size of the tumour. It was clearly, therefore, a case of pharyngeal or possibly œsophageal pouch.

He was seen once more during the next few days, and in view of the serious symptoms that usually, sooner or later, are said to supervene in these cases, he was urged to allow the pouch to be removed while it was still small, but he could not at this time make up his mind to accept this advice.

He was not seen again until December 31st, 1900, when he was admitted to University College Hospital, and the following history was elicited:

The patient, who is thirty-one years old, had been in the habit of smoking to excess, and had habitually taken more alcohol than was good for him. Twelve years previously he had received a blow from a fist on the left side of the neck, which was painful, but was followed by no special symptoms at the time. Two years later he began to feel something working up and down the left side of the neck on swallowing. It was not always present, but occurred from time to time, but no fresh symptoms occurred until about three years ago.

During the last three years he had been liable to acute attacks, lasting usually about a fortnight, during which a lump developed at the left side of the neck, and swallowing became difficult and painful, until at last he could only swallow liquids. At the end of the attacks the lump disappeared, sometimes gradually, sometimes suddenly during the night. He had had about seven such attacks, and also slighter ones which he had cut short by squeezing the lump, which always caused the expectoration of a quantity of phlegm, and almost daily he had coughed up what he described as "a piece of flesh," about one inch long and curled up, which sometimes stuck in his throat, and caused him to choke and vomit.

A full feeling in the neck warned him some twenty minutes before the commencement of an attack.

The last attack began on December 21st, and resulted in a much larger and more painful swelling than had ever occurred before, and the skin for the first time became red over the swelling, though the redness was evidently not inflammatory. On admission the swelling presented the appearance shown in the photograph (Pl. XVII). It reached the middle line in front, and the posterior border of the sterno-mastoid behind, overlapping and pressing back this muscle. It reached upwards to a point just above and behind the angle of the jaw, and downwards to the clavicle, the upper border of which it just overlapped. He said that it had extended still further back a few days previously. It projected fully an inch and a half from the normal surface of the neck.

The tumour was very painful and tender, the skin was movable over it, and it was freely movable over deeper parts, except near the upper part of the larynx, to which it was fixed, and at which part it appeared to be thickened, though the rest of the swelling was evidently a very thin-walled cyst. The carotid vessels passed behind it. The percussion note all over it was tympanitic. When he held his breath and blew he could slightly inflate the tumour, which procedure produced a slight change in the percussion note.

There was no enlargement of the cervical glands, and no sign of pressure on nerves, though the voice was harsh and metallic. A laryngeal examination showed the parts to be normal, and that the cords moved freely. The temperature was normal.

Operation.—The pouch was removed on January 2nd, 1901, by a long incision over the most prominent part of the tumour. It was a delicate matter to dissect the thin skin from the surface of it, and whilst doing so the lump collapsed. It was not apparent that the wall had been pricked, but it is possible that this may have been the case. The anterior border of the sterno-mastoid with

the spinal accessory nerve entering it was drawn back, and the thin lower part of the cyst was then opened in order to make it easier to dissect it from the subjacent structures. The dissection was less difficult when the thicker upper part of the pouch was reached, and this was found to consist of two portions communicating by a rather small opening. At last the narrow pedicle was reached which passed through the thyro-hyoid membrane ; but very careful probing failed to discover the actual communication with the pharynx. The superior laryngeal nerve was not seen. The thyro-hyoid was stretched over this deeper part of the cyst, which lay altogether in front of the large vessels. The tubular pedicle was then secured by catgut stitches, a fresh portion of it being divided as each stitch was passed. Finally the stump was invaginated by four stitches passed after the manner of Lembert's sutures, and the larger wound was closed except immediately over the site of the pedicle, where a small plug of gauze was placed in case the stitches should give way.

The wound, however, healed perfectly, complete asepsis was maintained ; the last part of the plug was removed on January 12th, and healing was complete on January 19th.

The temperature remained normal throughout.

A careful examination of the pouch after removal was made by Mr. Bucknall. The wall of the smaller upper portion was three millimetres thick, that of the lower larger part varied from one to six millimetres.

The structure of the wall was fibrous, without any muscular elements except where some fibres of the platysma were incorporated with the outer surface. The lining was throughout glistening. That of the larger sac fell into folds like those seen in an hypertrophied bladder, but that of the smaller part was almost smooth, though showing this tendency to rugosity to a less extent.

Microscopically the outer part of the wall was made up of dense fibrous tissue with few nuclei. Within this was a layer of more nuclear connective tissue, with numerous dilated venules, and in part showing signs of recent inflammation. On this, and resting on a definite basement membrane, lay the remains of the lining epithelium, which had in most parts desquamated, and was only to be found intact in the deeper crypts (Pl. XVIII). Where perfect this consisted of about three to six layers of columnar cells with oval nuclei, the superficial layers being ciliated. The deeper layers, which alone remained over the greater part of the inner surface, were columnar or cubical; the superficial ones were long and regular, and ciliated with definite free borders united to form a plane inner surface to the sac, where they were not shed or concealed by débris. The inflammatory cells, which had in parts burst through the basement membrane, had evidently been the cause of the desquamation. Close beneath the epithelial lining lay several nodules of lymphoid tissue, and here also at one spot a group of acini were cut across, resembling in every detail of their structure a mucous salivary gland. The large cubical cells composing the lining of each acinus displayed various stages of secretive activity. A few ducts were also visible (Pl. XIX).

It is possible to obtain records of nearly two hundred cases of pharyngeal pouch and fistula. They probably all arise in connection with one or other of the visceral clefts, and the following varieties occur which depend upon the position of the obliteration of the cleft:

- (1) It may remain open throughout its entire length, giving rise to the complete fistula opening into the pharynx and on the side of the neck.

- (2) No external opening may be formed, and a pharyngeal pouch with a blind extremity may remain.

- (3) The internal aperture may become shut off from the pharynx, leaving a blind external fistula.

(4) Both internal and external openings being closed, a "dermoid" cyst of the neck may remain.

In more than a third of all the cases the fistula was complete, and of the remainder the majority presented an external fistula, either blind or with the internal orifice so concealed that it could not be found.

In several a pharyngeal pouch was observed to burst externally, giving rise to the formation of a complete fistula; and conversely the external orifice of a fistula has been known to close, giving rise to a pharyngeal pouch, or the internal orifice once patent has become occluded, and a blind external fistula has remained.

These facts point among others to the unity of origin of the various conditions. The internal orifice, when present, has always been found in the pharynx, and not, as at one time was stated, in the larynx. This has been proved by dissection in three cases, by passing the probe in twelve, by the laryngoscope in three, and by injection from below in seventeen. It is usually small, and like the orifice of Stenson's duct situated on a small papilla, but in one case it was large enough to receive the last joint of the finger. In those cases in which its anatomical position has been accurately defined, it opened on the side wall of the pharynx close to the margin of the tonsil, and frequently in connection with the fold of the palato-pharyngeus.

The external orifice, if it exists at birth, is situated at some point along the anterior border of the sterno-mastoid; it is single, and is sometimes guarded by a subcutaneous nodule of embryonic cartilage. The most common position is just above the sterno-clavicular articulation, but it may be found at some higher point along the anterior border of the sterno-mastoid, and it has been noted opposite the cricoid, on a level with the top of the thyroid, close to the great cornu of the hyoid bone and opposite the angle of the jaw. If it results from the bursting of a dermoid cyst or pharyngeal pouch, as a rule it is still generally situated at the edge of the

sterno-mastoid, but the pouch may have reached some other position before it burst, and several external fistulous apertures may in some cases be formed.

Where a complete fistula exists some varieties in its relation to surrounding parts have been noted, but in a general way it may be said to arise from the pharynx, near the great cornu of the hyoid bone, and to run down with the carotid vessels beneath the sterno-mastoid as far as the external orifice. Pharyngeal pouches and blind external fistulæ, as a rule, follow the same course.

The lumen of the canal is usually small, and its course is tortuous, especially above, and the whole pouch or fistula may form little more than a fine cord-like band extending down the neck. But in those cases in which no external orifice exists, or in which such orifice becomes blocked or closed, there is a tendency for distension to occur, and a swelling is then formed in the side of the neck which may possess the typical features of a pharyngeal pouch; that is to say, it forms a thin-walled, resonant, globular tumour, moving with deglutition, perhaps becoming more tense on forcible expiration, and capable of reduction in size on pressure. It is very rare for food to pass into the cyst, as usually happens in cases of true œsophageal pouch; it is, however, common for mucus to be expectorated, as happened in the present case.

It is interesting to try to determine which of the visceral clefts is concerned in the development of such a cyst. But the problem is a difficult one, and the difficulty is increased by the uncertainty existing at the present time with regard to the structures that correspond to the particular clefts and arches; and it must be added that, as neither of us is a skilled embryologist, all information on these matters had to be taken at second hand.

It is generally held that in the first (or mandibular) arch the lower jaw is developed; in the second the styloid process, the stylo-hyoid ligament, and the small cornu and part of the body of the hyoid bone; in the third the great cornu of the hyoid bone.

The relation of the larynx to the visceral arches has not been determined.

As the neck of this pouch passed through the thyro-hyoid membrane, it is clear that it could not have been

FIG. 1.

I—V. Visceral clefts.
1—6. Visceral arches.
s. Styloid process.
s.h. Stylo-hyoid ligament.

s.c. Small cornu of hyoid.
g.c. Great cornu of hyoid.
H. Body of hyoid.

connected with any cleft above the third (Fig. 1). It remains to be seen whether, supposing it was connected with the third, it bears a right relation to vessels and nerves.

The relation of nerves to the upper clefts is held to be

constant, and is shortly as follows (Fig. 2). The nerve that is associated with each cleft divides at the dorsal end, and is distributed partly to the arch above (anterior) and partly to the arch below (posterior). Thus the seventh nerve is distributed partly to the first arch and partly to

FIG. 2.

- I—V. Visceral clefts.
 1—6. Visceral arches.
 v—x. Cranial nerves.
 M. Third division of v nerve.
 F. Facial nerve.

- G.P. Glosso-pharyngeal nerve.
 S.L. Superior laryngeal nerve.
 R.L. Recurrent laryngeal nerve.
 Va. Vagus nerve.

the second; the ninth (glosso-pharyngeal) partly to the second and partly to the third; and the tenth (vagus) partly to the third (external laryngeal) and partly to the fourth arch and to lower parts of the body. Unfortunately the position of the external laryngeal was not

ascertained at the time of the operation ; the main trunk of the vagus was certainly behind the neck of the pouch. It is not possible, therefore, to say whether the external laryngeal was in front of the neck, as it ought to have been if the pouch were connected with the third or some lower cleft ; it is, however, almost certain that it did occupy this position.

The relation of the carotid arteries to the clefts is not yet definitely settled, and the issue is confused by the fact that the embryo of the chick has been so much employed in the study of embryology, and that the development of the carotids does not appear to be the same in birds as in mammalia. Another source of confusion arises from the fact that the vessels are displaced downwards (towards the thorax) in the course of development.

The accompanying diagrams (Fig. 3) show what, as far as we can ascertain, is considered by many good authorities to be the developmental relation of the carotids to the arches. According to this view, the external carotid is developed from the ventral aortic vessel, and thus passes in front of the visceral arches ; the internal carotid from the third arch and that part of the dorsal aortic vessel which is above (anterior) to it.

If, then, this pouch were developed from the third cleft, its neck ought to have passed behind the two carotids. But such was not the case. Both carotids were behind the neck, and only one explanation suggests itself—that the pouch was quite small at the time when the vessels were being displaced downwards (towards the chest), and that in its subsequent growth it pushed up in front of the external carotid.

If we turn to the descriptions of other observers, it is found that it is not infrequent for the neck of the pouch to pass between the external and internal carotids, and that the relation of some of them to the second cleft appears to have been clearly made out.

Thus König, Watson, Heuter, Rehn, Tricomi, and

Gussenbauer, all noted that the canal passed beneath the lower border of the digastric, and Watson and Heuter traced it between the external and internal carotid arteries to the pharynx, the seventh nerve lying above it, and the ninth nerve and the stylo-hyoid ligament below

FIG. 3.

E.C. External carotid.
I.C. Internal carotid.
I—V. Visceral clefts.

1—6. Visceral arches.
s. Subclavian.
Ao. Aorta.

it. This seems to have been the course of the canal in the very few cases that have been dissected post mortem. Some others have been traced into the same position by operation, but though a few others, like the present case, have been traced up to the thyro-hyoid membrane in an operation, the exact position of the internal opening has

not been ascertained. In Watson's case a branch of the ninth nerve innervated the walls of the pouch, and in the case recorded by Berger probing of the fistula caused cough and stopping of the pulse and other symptoms of distress thought to be due to irritation of the superior laryngeal nerve. In Cavazzani's case vomiting followed the use of the probe.

In most cases the walls were found to consist of fibrous tissue, and striped muscular fibres have been noted by some observers. The lining epithelium was either of the stratified flattened variety, or columnar and ciliated as in the present case. König noted two canals side by side, one lined with flattened and the other with columnar ciliated epithelium, and other observers have described pouches lined with flattened epithelium below and columnar and ciliated at the upper part,—that is to say, the part of the pouch nearest the pharynx.

The facts pointing to the congenital origin of pharyngeal pouches and fistulæ, and to their probable connection with the visceral clefts, may be summarised as follows :

- (1) They are frequently hereditary.
- (2) They are often bilateral.
- (3) Though they often do not appear till early adult life, they may be present as fistulæ at birth or very early in life, and it is probable that they are often overlooked, as the symptoms they give rise to are slight and may take some time to develop.
- (4) The peculiar course of the canal.
- (5) The presence of a lining of ciliated epithelium, although they are derived from a portion of the pharynx normally covered with squamous epithelium.
- (6) The fact that the neck of the canal or pouch often passes between the carotids in the position of the second visceral cleft.
- (7) The fact that striated muscle has been found in the wall of such pouches and fistulæ, and that in one case a band of striated muscle ran from the end of the

pouch down the course usually pursued by complete fistulæ.

(8) The fact that in several cases glands having a similar structure to the normal mucous salivary glands have been found opening into the lumen of pouches and fistulæ. This may account for the discharge noted in cases like the present. One of us has met with an instance, many years ago, of a cyst, then supposed to be developed in connection with the bursa beneath the hyoid, which was, however, lateral in position, and which it was found impossible to dissect out. The patient, who was under the care of Lord Lister, was a distinguished advocate, and he was much troubled after the operation by a mucous discharge from the fistula, which occurred when he was speaking in court. The fistula, after many years, closed.

(9) The fact that around the lumen a considerable quantity of lymphoid tissue was noted by König in three cases, and also by others, while cartilage has occasionally been met with in the walls of such fistulæ.

As this pouch was obviously of the pharyngeal variety, no reference is here made to the so-called pressure pouches of the œsophagus, but it is at least possible that some of these should be placed in the same category.

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Pouches similar in many ways to the one forming the subject of this paper are described by Watson, Pertick, Zuckerkandl, Kirchner, Schmidt, Kostanecki, and Kopstein. The references of the first six will be found in Kostanecki's paper, mentioned above.

DISCUSSION.

Sir FELIX SEMON.—Perhaps the most important part of the paper refers to the unity of the various forms of congenital malformation met with in the front of the neck. Formerly it was held that these various forms, diverticula of the pharynx, complete or incomplete fistulæ in front of the neck, and cysts, were of different origin, and it is the great merit of Kostanecki that he, in 1890, for the first time established the unity which has been so admirably and clearly demonstrated in this paper. That is a point of great practical importance. I was astonished to hear that as many as two hundred cases are already on record. I myself have only seen two such cases, of which one belongs to the variety described in this paper, the second being a case of incomplete fistula. It was one in which there was a slight mucous discharge on the right side of the neck on a level with the larynx in front of the sterno-mastoid. It was possible to probe it up to the pharynx, where it ended blindly, but its origin was obviously close to the tonsil. I explained to the patient that a cure could only result from dissecting out the whole of the fistulous tract, and that apparently discouraged him. My second, or rather my first case, was one which I am not likely to forget, for I found myself in the position of the sorcerer's apprentice in Goethe's poem, who could not get rid of the spirits he had invoked. The patient was a young gentleman who wished to enter the army. He had a swelling on the left side of the neck which his medical attendant thought was thyroid, but on examining him it was obvious that, unless there was thyroid tissue quite separate from the main body of the gland, it could not be that, for it was situated between the hyoid bone and the lower border of the cricoid cartilage, occupying almost the position shown in Mr. Godlee's diagram, but was not nearly as large. It was egg-shaped, distinctly moveable, did not seem to have any pedicle, was not tender to the touch, and showed some indistinct fluctuation. I thought it was probably a simple cyst, and I probed it, evacuating a considerable quantity of what looked like ordinary pus, and the tumour collapsed. Within a week it filled again, and a large quantity of caseous *débris* came away. I enlarged the opening, but the tumour always immediately refilled. I need not weary you with the clinical details beyond saying that it would not close in spite of every treatment. With the finest probe I could not detect the length and direction of the canal. I had a consultation with Sir Frederick Treves, who considered

it a sebaceous cyst. By his advice I laid it open, scraped the walls, and burned it with the actual cautery. This also was unsuccessful, and ultimately it was dissected out completely by a country surgeon, who informed me that the sac ended blindly above the hyoid bone. The case, which was observed long before the publication of Kostanecki's paper, taught me the important lesson that a fluctuating tumour in front of the neck, of the exact nature of which we are uncertain, should not be probed, unless we are prepared to go the length of dissecting the whole thing out if need be. In conclusion, I should like to ask what are the serious symptoms which the authors state are likely sooner or later to supervene. Do they refer to the possibility of this complete or incomplete fistula becoming later a so-called branchiogenous carcinoma?

Mr. GODLEE.—I may mention at once that this phrase was introduced at a time when we were thinking more of œsophageal pouches, in which inflammatory troubles are common.

Mr. BUTLIN.—I have never seen a cyst of this kind, and therefore know little about them. The only thing at all resembling this which I have seen is a specimen in the museum at St. Thomas's Hospital to which the author has referred in his paper. When I examined this specimen years ago in connection with *œsophageal* pouches I could not make it out, and was much surprised to find that it was quite on one side of the neck, and ran down from a very much higher point than do œsophageal pouches. On the other hand, I have dissected out a branchial fistula, which opened near the sterno-clavicular articulation, into which a probe was passed three or four inches upwards. I dissected it without any particular difficulty, and it was by no means a serious operation. It reached right up to the posterior palatine arch, where I invaginated it into the mouth, tied the neck, and then cut it off. The wound healed by first intention, and the boy had no further trouble. That was in 1897. I wish I had taken notes of the operation myself, for the dresser only stated that the tube lay in front of the external carotid artery, and said nothing about the internal carotid artery and the nerves. Naturally, in doing an operation of this kind, one limits one's self to the removal of the trouble without searching about to discover the relations. With regard to *œsophageal* pouches and their possible relationship to these pouches, I may say from a very extensive experience of them—for I have removed not less than seven and have seen at least twelve—they never present any external tumour like this in the neck. Then, again, they are all built so exactly upon one type, open upon the same place, and are so much the same size that they seem to have no relation at all with branchial fistulæ, and I cannot help thinking

that they are a disease or a condition of an entirely different character.

Mr. A. E. BARKER.—I may mention a case which will add to the list which Sir Felix Semon has made of his cases. It presented many features in common with the one described except that it was larger and was on the other side of the neck, but there were certain points of difference, and this may add to its interest. The patient was first seen by Sir Felix Semon, but the patient was an old friend of mine, and he asked me what had best be done. I expressed the opinion that it was a dermoid cyst or pouch connected with one of the branchial clefts, and I based my conclusion on the following points:—(1) I was aware that the right auricle was badly developed; (2) down in the episternal notch I noticed a little tuft of hair, and over the centre of this a tiny depression with a little hardness around it; (3) between the two points under the lobe of the ear was a tumour about the size of the bell over the electric light. There was this feature, that the tumour varied very much in size, being sometimes inconspicuous and at others quite large. It had the soft doughy feel of an incompletely filled sac. I could not empty it, and the patient said it sometimes filled rapidly. Then, again, he felt very ill at the time, although in general good health. This suggested to me that there was an auto-intoxication going on. In view of the position of the tumour right in front of the sterno-mastoid, and of the other signs, I thought it was a dermoid, and I removed it. The relations are fresh in my memory. I made an incision from the lobe of the ear to over the episternal notch, bifurcating below to enclose the dimple. I came down upon the sac and dissected it off the carotid vessels with some difficulty, for it was closely adherent to them as the result of past inflammation. I stripped the common carotid for two or three inches and gradually got it out. It came out whole, and it narrowed further and further down to about the size of a lead pencil alone. I traced it up to just below the tonsil, and with my finger in the mouth and my other fingers grasping what I may call the pedicle, I could feel that they were close together, only the mucous membrane intervening. I put a silk ligature round it and tied it. Before doing that I made a little hole in the upper part, and the probe came right against my finger below the tonsil. I have no doubt that there was an opening there. I had sections made of the narrower part, and they showed what the author's case also shows. The point of interest is that we diagnosed it by an examination of the skin below. What cleft this actually belonged to I cannot tell. I took it to be the lowest cleft, but that may or may not be correct. Since it was taken away my friend tells me that his constitution has been quite revolu-

tionised. The incision, of course, is a very formidable one, but it healed throughout by first intention. I may mention that I have had several cases, which have been recorded in the 'Transactions of the Clinical Society' many years ago.

Mr. GODLEE.—I was rather surprised to hear Sir Felix Semon say that he had only seen one or two cases of the sort in his large experience. I was shown one of these cases quite early in the course of my experience, and since then I have seen a good number of such fistulæ opening into the neck. Mr. Butlin's remarks are always interesting, and one of the most interesting things he has said is that he has never seen a case of this kind, although he has met with so many of œsophageal pouch. When I saw the case first I must say I thought it was one of œsophageal pouch, because I have heard more about them, also because there was a history of a blow on the side of the neck, and the theory is that some of these pouches are in some way due to injury. The dangers I alluded to have been described in connection with œsophageal pouches.

Mr. BUTLIN.—You never see an œsophageal pouch come as high up as this one did.

Lining of pouch shewing columnar ciliated epithelium.

WALL OF POUCH shewing—

(a) Lining of columnar ciliated epithelium ; (b) Loose connective tissue ; (d) Dense outer layer of fibrous tissue ; (c & e) Acini of salivary glands in the wall of the pouch.