Laryngocoele

By P. M. Stell (Liverpool) and A. G. D. Maran (Edinburgh)

The laryngocoele was first described by Larrey, surgeon to Napoleon's Army in Egypt, in 1829. He described compressible pouches related to the thyrohyoid membrane in the Muezin, who were used to call the faithful to prayer from the minaret and he described these swelling as 'goitres aeriennes'. Many of the patients became so hoarse that they ultimately became aphonic, at which point they were relegated to caring for the temple baths.

Virchow, apparently unaware of Larrey's description, named this affliction 'laryngocoele ventriculatis' in 1867, and since that time there have been almost 100 articles in the English literature concerned with this topic. Most of these deal with single cases, or at the most a handful, so that in an attempt to build up an overall picture of this lesion, all of the clinical accounts in the English literature have been reviewed, and the appropriate clinical data abstracted.

Material and method

Eighty-three articles have been written in the English language; of these, 81 reports of definite laryngocoeles have been read personally, while two, those by Pearsall (1897) and Shambaugh (1914) could not be traced since they were in obscure journals not obtainable in this country. Several reports such as that of El-Hakeem (1951) and El-Mofty (1959) have been rejected since it appeared that they related to laryngeal cysts or pharyngeal pouches rather than true laryngocoeles. Two pseudo-laryngocoeles were also not included—one traumatic in origin reported by Simpson (1938) and one resulting from perforation of the thyroid cartilage by a tuberculous process allowing air into the soft tissue of the neck, reported by Kilpatrick (1940).

Furthermore, the report by Holinger (1961) has also not been included, since this refers to neonates. Laryngocoeles in neonates have not been seen by any other author, and it appears that Holinger's patients could equally as well have suffered from a laryngeal cyst. His adult cases (Holinger *et al.*, 1954) have however been included. The following details were abstracted: age, sex, race, presenting symptoms and their duration, type, number of laryngocoeles, pathological and histological data, details

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of associated conditions, details of mis-diagnosis and treatment. The 83 articles reviewed related to 130 patients; nine personal cases of the authors are also included, making a total of 139 case reports available for study.

Results

The results as regards age, sex, and race are shown in Tables I and II from which it emerges that this disease is commonest in white men in their fifties, and appears to be uncommon in women and other races. The only author to report laryngocoeles in babies is Holinger (1961); he did not describe the histological findings on these laryngocoeles and it appears possible that they were true cysts.

TABLE I.

SEX AND AGE INCIDENCE

	Male	Female
0 – 9	2	–
10 – 19	2	–
20 – 29	16	–
30 – 39	12	4
40 – 49	20	2
50 – 59	28	4
60 – 69	16	7
70 – 79	4	0
80 – 89	2	1
Not stated	19	–

TABLE II.

RACE

White	59
Negro	6
Arab	7
Not stated	67

It is difficult to assess the incidence of this condition, but it is obviously rare. In the years 1965 to 1973 the authors have seen nine laryngocoeles, and at the same time have seen about 500 patients with laryngeal carcinoma. The incidence of laryngeal carcinoma is of the order of 1,000 cases per year, so that it appears that laryngocoeles have an incidence of approximately 1 per 2,500,000 population per annum in this country.

It is well known that laryngocoeles may be internal, external or combined, that they may be unilateral or bilateral and that they may become infected, forming a pyocoele. The relative frequency of these events is shown in Tables III, IV and V, which show that this disease is most often unilateral, that the combined type is the most common, and that about 10 per cent of laryngocoeles become infected.

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TABLE III.

NUMBER OF LARYNGOCOELES

Unilateral	106
Bilateral	19
Not stated	14

TABLE IV.

TYPE OF LARYNGOCOELE

Combined	50
External	34
Internal	21
Not stated	34

TABLE V.

PYOCELES

13

The presenting symptoms are shown in Table VI, from which it is obvious that the two common presenting symptoms are hoarseness and a swelling in the neck. Twenty-two patients have presented with respiratory embarrassment, however, and it is remarkable how this could occur relatively suddenly over the space of a few days or even hours, in a patient who had previously only had mild symptoms for months or years; this pattern of events has caused the death of two patients, and almost that of a third. The length of time the symptoms had been present varied enormously from a few days to 40 years, but there did not seem to be any point in recording this information.

TABLE VI.

SYMPTOMS

Hoarseness	70
Swelling in the neck	65
Stridor	22
Dysphagia	11
Sore throat	6
Snoring	4
Pain	2
Cough	3
None	1
Not stated	15

As regards histology, virtually every report where such details were given, shows that laryngocoeles are lined by columnar ciliated epithelium with sometimes patches of squamous metaplasia. They can thus be usually distinguished from laryngeal cysts, which are often lined by squamous epithelium.

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As regards aetiology, the patient's habits relative to any hobbies or occupations requiring blowing are shown in Table VII, which shows that

TABLE VII.

'BLOWING' HOBBIES OR JOBS

Not relevant	39
Possibly relevant	4
Not stated	96

at most four patients were subject to these habits; in two, this history was irrelevant, the laryngocoeles appearing at the same time as the patient took up trumpet playing, which thus merely served to bring an already existing laryngocoele to the patient's notice. There are also several cases where the patient first noticed a lump after a coughing bout, which also suggests that such a bout filled an already existing laryngocoele.

Of much more importance as regards aetiology is the co-existence of a carcinoma or papilloma of the larynx (Table VIII). It appears that the

TABLE VIII.

CO-EXISTING PATHOLOGY

Carcinoma	
False cord	2
'Larynx'	3
Vocal cord	1
Subglottis	1
Papilloma	2

carcinoma probably acts as a valve, allowing air under pressure into the ventricle. Asymptomatic laryngocoeles can occur, however, in about 5 per cent of patients with laryngeal carcinoma as shown in the studies of Meda (1952) and of Pietrantoni *et al.* (1959). This matter has also been investigated in France by Cachin and his colleagues (Gerand-Marchant *et al.*, 1969). They found external laryngocoeles in 16 per cent of laryngectomy specimens where the operation had been carried out for laryngeal carcinoma, as against 2 per cent in laryngectomy specimens from patients with carcinoma of the piriform fossa. Furthermore, they investigated the height of the ventricle in these two groups and found that the ventricle in patients with laryngeal carcinoma was deeper than in patients with pharyngeal carcinoma, the difference being statistically significant. They attributed this difference to the increased air pressure consequent upon obstruction produced by the laryngeal carcinoma.

Probably because of the rarity of this lesion, it has several times been mis-diagnosed (Table IX). When it was mistaken for a branchial cyst, little harm resulted, but several patients have unfortunately been irradiated. The patient in whom the diagnosis was missed, died of asphyxia

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TABLE IX.

MISDIAGNOSIS

Branchial cyst	3
Irradiated as laryngeal carcinoma	5
Diagnosis missed, leading to death	1
Gas gangrene	1

caused directly by the laryngocoele, and this emphasizes the importance of an X-ray of the neck of every patient who is hoarse.

The specific investigation in this condition is a plain radiograph of the neck which shows the characteristic appearances. There are a few reports of demonstrations of a laryngocoele by tomography or by injection of contrast medium into the sac (Blewett, 1939), but these appear to be unnecessary. It appears to be important to X-ray the patient with the glottis open, since the detection rate falls markedly when the larynx is under tension (MacFie, 1966). A high incidence of asymptomatic laryngocoeles, diagnosed radiologically, has been reported in patients with laryngeal carcinoma (Meda, 1952; Pietrantoni *et al.*, 1959), in bandsmen (MacFie, 1966), and in patients with a chronic cough (Felisati *et al.*, 1958). These appear to be incidental findings and do not require further attention.

The vast majority of laryngocoeles which have been treated, have been managed by an external approach, in which the neck of the sac has been followed medial to the thyroid cartilage and divided and stitched at the level of the ventricle. There are occasional reports of management via a laryngofissure, but usually when the diagnosis was thought to be carcinoma of the larynx (Freedman, 1938). The technique which is apparently popular in France (Bourdon, 1954), of injecting tri-chloracetic acid into the sac, does not appear to have become popular in Anglo-Saxon countries, although there is one report of carbolic acid injection from the early part of this century (Ingals, 1914). A few internal laryngocoeles have been managed by endoscopy (Holinger, 1961). The authors have used an external approach, based on Ogura's technique (1958) for supraglottic laryngectomy, in which the upper part of the thyroid cartilage is removed on the same side to provide access to the neck of the sac.

Operative technique

The operation is conducted under general anaesthesia with endotracheal intubation. A collar incision is made over the thyroid cartilage and skin flaps including the platysma are elevated up to the hyoid bone superiorly, and down to the lower border of the thyroid cartilage. The fundus of the sac is defined and dissected free. The strap muscles are retracted laterally but it may be necessary to divide the sternothyroid and thyrohyoid muscles at their attachments to the thyroid cartilage for adequate access. A perichondrial flap is then elevated by incising along

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the superior border of the ipsilateral thyroid cartilage to the thyroid notch, then down the midline and along the lower border of the thyroid cartilage laterally. The perichondrial flap is elevated leaving an inferior attachment. The upper half of the thyroid cartilage is thus exposed and with a Stryker saw or dental fissure burr, the cartilage is divided horizontally (Fig. 1), taking care not to damage the superior laryngeal neurovascular bundle or enter the lumen of the larynx. The upper portion of the thyroid cartilage is removed to allow adequate access to the intralaryngeal part of the sac and its neck (Fig. 2). The sac is then divided at its origin and the stump invaginated. The perichondrial flap is sutured to the remains of the thyrohyoid membrane and the neck wound closed with drainage.

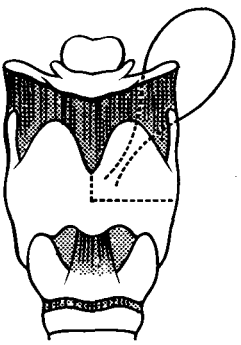


FIG. 1.
Cartilage cuts marked out.



FIG. 2.
Sac of the laryngocoele exposed.

Discussion

The ventricle of the larynx was first described by Galen, and it was also referred to by Morgagni (1741), and indeed was for long known as the sinus of Morgagni. Arising from the anterior end of the ventricle lies the saccule which proceeds upwards between the thyroid cartilage and the ventricular fold, often ending as high as the upper border of the thyroid cartilage. The saccule was first described by Hilton (1837), but he used the term ventricle to describe the cavity of the larynx, and the term saccule to describe what is now described by the term ventricle. He describes the saccule by the term laryngeal pouch. Air sacs occur in a vast number of animals (Watkins, 1935)—for instance, the cheek pouch of monkeys, the fish pouch of pelicans, the tracheal sacs of emus, the syrinx of male quacking birds, to name only a few. Laryngeal air sacs are also found in many animals, mostly mammals, but also in some frogs and toads, and in one fish, the garnet. They become more developed the higher the animal is in the evolutionary scale, being most highly developed in certain anthropoid apes, but they do not usually appear in man. Lateral

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laryngeal sacs arising from the ventricle of Morgagni occur only in the Siamang gibbon, the chimpanzee, the orang-utang and the gorilla, being more developed in males (as is the syrinx in ducks, and the laryngocoele in the human). Median laryngeal sacs also occur in some animals, for example, the tree toad, and the gurnet, and laryngeal sacs arising from the ventricle itself (i.e. not the saccule), occur in many monkeys; these sacs often expand into the laryngeal skeleton, but are obviously not homologues of the human laryngocoele. Negus (1929) has shown that the lateral sacs do not have a vocal function, but are a means of enabling the animal to rebreathe while holding its breath for long periods, particularly in animals who make great exertions with the muscles of the chest wall fixed.

One of the main points of interest in laryngocoeles is their aetiology. It is frequently stated that they occur in patients who blow during their work, such as glass blowers, or during their hobbies, such as trumpeters. This statement is almost certainly incorrect for the following reasons:

(*a*) there is only one case on record (Freeman, 1952) in which the laryngocoele came on after many years of a job requiring blowing.
(*b*) the vast majority of laryngocoeles are unilateral.
(*c*) they are rare.
(*d*) their recurrence after simple removal has never been recorded.

It appears virtually certain, therefore, that they are an atavistic remnant corresponding to the lateral laryngeal air sacs of the higher anthropoid apes, and that on occasion they become manifest suddenly in response to a sudden increase of intra-laryngeal pressure, caused by coughing, straining at stool or trumpet playing.

Summary

All cases of laryngocoele in the English literature have been reviewed and a further nine added, making a total of 139. The sex incidence is 5 : 1 in favour of men, and the maximum age incidence is in the sixth decade. Approximately two out of three laryngocoeles are unilateral and they may be combined, external or internal, with roughly equal frequency; about 8 per cent become infected and present as pyocoele. The commonest presenting symptoms are hoarseness and a lump in the neck, with almost equal frequency.

There is only one case on record where the laryngocoele could have been caused by prolonged and repeated blowing against resistance. This factor appears to play no part in the cause of laryngocoele though increased air pressure in the larynx may make an already existing laryngocoele manifest; this increased air pressure may be caused by a co-existing carcinoma. Laryngocoeles appear to be an atavistic remnant from the higher apes, particularly those who use their arms with the thoracic cage fixed whilst swinging through the trees.

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Ear, Nose and Throat Infirmary,
Myrtle Street,
Liverpool L7 7DF.

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