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The aetiology of lateral cervical (branchial) cysts: past and present theories

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Abstract

Four theories have been suggested to explain the aetiology of lateral cervical cysts. Ascherson (1832) suggested that the cysts arose due to incomplete obliteration of branchial cleft mucosa, which remained dormant until stimulated to grow later in life. His (1886) suggested these cysts were vestiges of the precervical sinus. Wenglowski (1912) believed lateral cervical cysts developed from the third pharyngeal pouch (thymopharyngeal duct).

A number of investigators during the 19th century noted the close relationship between lateral cervical cysts and lymphoid tissue (Lucke, 1861). Luschka (1848) suggested that cystic degeneration of cervical lymph nodes was the mechanism by which lateral cervical cysts were formed. This theory received little support until King (1949) studied the histology of a large number of lateral cervical cysts and concluded that these cysts resulted from cystic transformation of cervical lymph nodes.

The evidence for and against these theories of aetiology is discussed. The debate is centred on a study of 20 patients with lateral cervical cysts operated on in the Department of Otolaryngology, Bedford Hospital, between January 1986 and December 1991. In all twenty cases the wall of the cyst was found to be composed of lymphoid tissue, histologically identical to that present in lymph nodes. The mean age of presentation was 31 years, and in no case was a tract or cord found which connected the cyst to the skin or pharynx.

The evidence strongly suggests that lateral cervical cysts develop from the cystic transformation of cervical lymph nodes. Mechanisms by which this may occur are discussed.

Key words: Branchioma, aetiology

Introduction and Historical Review

Cysts presenting in the lateral aspect of the neck were first described by Hunczovsky (1785). Since that time, there has been a wealth of names for these cysts: dermoid cyst of the sheath of the internal jugular vein (Langenbeck, 1859); deep-seated atheromatous tumour (Schede, 1872); congenital hydrocele of the neck (Smith, 1866); hygroma colli (Luschka, 1848); atheromatous cysts of lymphatic glands (Lucke, 1861); branchial cyst (Ascherson, 1832); tumour of the branchial cleft (Virchow, 1866); lateral lympho-epithelial cyst (King, 1949); benign cystic lymph nodes (Bhaskar and Bernier, 1958). This variety of names reflects the lack of agreement as to their definition and aetiology. A number of differing opinions are stated in recent articles with respect to both aetiology (Wild *et al.*, 1987; Doi *et al.*, 1988) and definition (Maran and Buchanan, 1978; Howie and Proops, 1982).

In order to understand how the present confusing position has been reached, it is necessary to consider the evolution of theories over the last century.

At the beginning of the nineteenth century Rathke (1828) described the development of the pharyngeal arches in the human fetus. Shortly after this, Ascherson (1832) described 11 cases of branchial fistulae (Figure 1). Ascherson equated the development of lateral cervical cysts with that of branchial fistuale, and suggested cervical cysts were the result of imperfect obliteration of a pharyngeal cleft. The theory was based on Ascherson's impression that lateral cervical cysts developed at the site occupied by the branchial apparatus in the embryo. During the rest of the 19th century increasing support developed for this 'Branchial Theory'. Without providing any new evidence a number of investigators re-emphasized this link between the pharyngeal arches and cervical cysts (Langenbeck, 1859; Heusinger, 1864).

The second theory of aetiology was suggested by His (1886). His considered that branchial fistulae were related to the cervical sinus rather than the pharyngeal clefts or pouches. In a similar manner to the 'Branchial Theory' this 'Precervical Sinus Theory' was extended to include lateral cervical cysts (Work, 1972; Sadler, 1990).

The third theory proposed the third pharyngeal pouch as the site of origin of the lateral cervical cyst. Wenglowski (1912) dissected the necks of 78 embryos, 144 infants and 59 adult cadavers and showed that pharyngeal cleft tissue was not represented in any adult tissue inferior to the hyoid bone. Thus any cyst lying below this level could not be derived from a pharyngeal cleft. Therefore the 'Branchial Theory' could not explain a considerable

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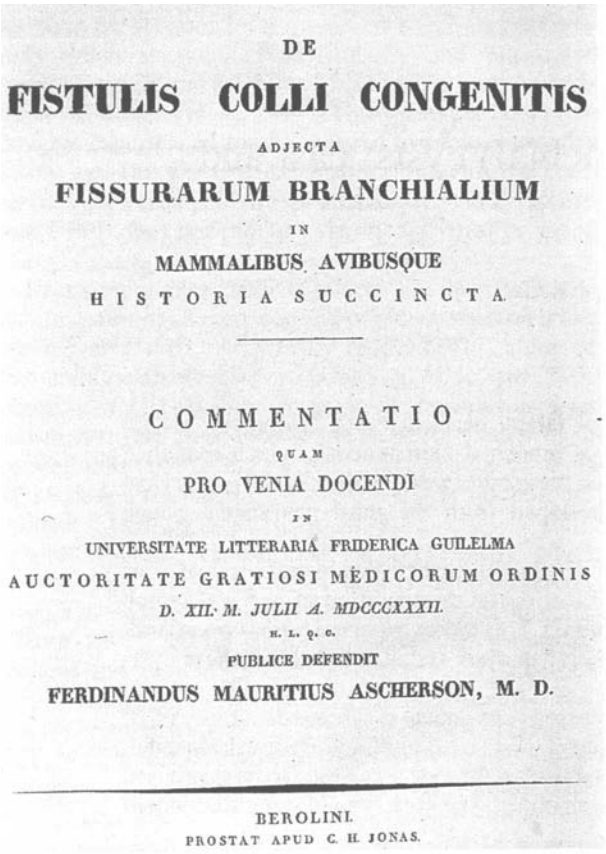

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FISTULIS COLLI CONGENITIS

ADJECTA

FISSURARUM BRANCHIALIUM

IN

MAMMALIBUS AVIBUSQUE

HISTORIA SUCCINCTA.

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Fig. 1
The title page of Ascherson's book (1832), in which the *Branchial Theory* is first discussed.

number of cervicl cysts. Wengłowski went on to describe the development of the thymus from the third pharyngeal pouch via the thymopharyngeal duct (Figure 2). He suggested that incomplete obliteration of the thymopharyngeal duct resulted in a lateral cervical cyst. Wengłowski's work was reviewed in English by Meyer (1932).

The final theory of aetiology suggests that lateral cervical cysts represent cystic lymph nodes. As early as the mid 19th century Professor A. Lucke (1861) noted that the external appearance of a lateral cervical cyst showed a great similarity to a hypertrophied lymphatic gland. Luschka (1848) described a lymph node lying between the external and internal carotid arteries (ganglion caroticum). He suggested that lateral cervical cysts resulted from cystic degeneration of this gland. However it was not until the work of King (1949) that the 'Lymph Node Theory' received much support. King studied 76 cervical

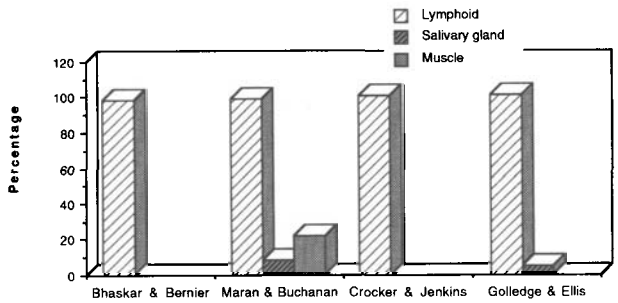

FIG. 3
The constituents of the wall of lateral cervical cysts; this study and a review of previous studies.

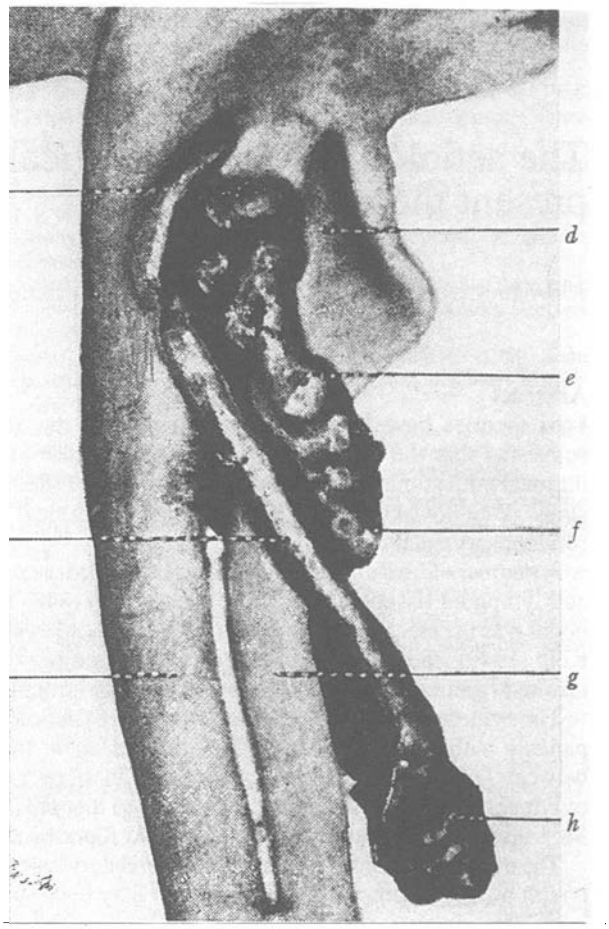

Fig. 2
Picture showing one of Wenglowski's models of the developing thymopharyngeal duct (*b*).

cysts. Based on their site, histology and a review of the embryological basis of the 'Branchial Theory', he concluded that cervical cysts had no direct relationship with any structure in the embryo. Instead he emphasized the close relationship between these cysts and lymphoid tissue and therefore suggested the name lateral lymphoepithelial cyst.

Further support for the association between cervical cysts and lymphoid tissue was provided by Bhaskar and Bernier (1959). They reviewed the histology of 468 cysts: 452 had a wall composed of lymphoid tissue (Figure 3). On the basis of this they concluded that cervical cysts developed as a result of cystic transformation of cervical lymph nodes.

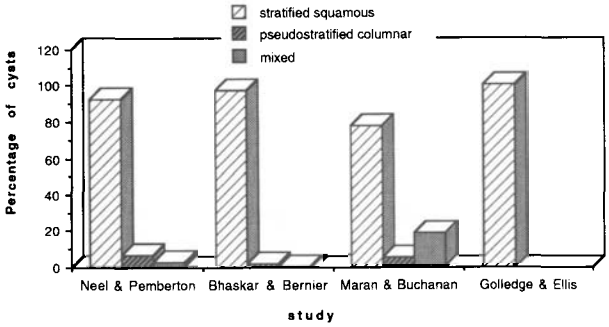

FIG. 4
The epithelium lining the lateral cervical cyst; this study and a review of previous studies.

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TABLE I
THE AGE OF PRESENTATION OF PATIENTS WITH LATERAL CERVICAL CYSTS: THIS STUDY AND A REVIEW OF PREVIOUS STUDIES

	Mean age (years)	Median decade	Range (years)
Golledge & Ellis (1994) (n = 20)	31	Third	14–50
Nee Pemberton (1945) (n = 319)	35	Third	1–84
King (1949) (n = 79)	28	Third	1–80
Bhaskar & Bernier (1959) (n = 468)	27	Third	5–65

A more recent review by Maran and Buchanan (1978) disclosed similar histological findings, greater than 90 per cent of the cysts studied were surrounded by lymphoid tissue (Figure 3). However, they pointed out that cystic transformation of lymph nodes is not known to occur in other anatomical sites.

Hence there are principally two explanations for the formation of the lateral cervical cysts. First, that they represent a remnant of the structures lying in the lateral aspect of the embryo (pharyngeal cleft/pouch, cervical sinus, thymopharyngeal duct), i.e. the congenital theories. Second, that they develop as a result of cystic transformation of cervical lymph nodes. Despite the lack of evidence in support of the congenital theories, a number of recent publications (Finn *et. al.*, 1987; Doi *et al.*, 1988; Gosain and Wildes, 1988; Sadler, 1990; Morrish and Manning, 1991) and standard textbooks (Boyd, 1986; Ellis, 1987; Walter and Israel, 1987; Sadler, 1990; Cheeseman *et al.*, 1992) continue to emphasize this view.

The aims of this study were to review the clinical and histological features of lateral cervical cysts excised at the Otolaryngology Department in Bedford General Hospital during the last six years. Then utilizing the findings of this investigation, together with evidence from previous studies, to reconsider the definition and aetiology of cysts which arise in the lateral aspect of the neck.

Material and methods

The medical records and histology of 20 patients who had lateral cervical cysts excised between January 1986 and December 1991 at Bedford General Hospital, were reviewed. The presenting features, length of history, signs on examination, operative findings and microbiological reports were noted. In addition the histology was reviewed.

Results

As in previous reviews, the median age of presentation was in the third decade (see Table I), the range of ages being 14 to 49 years; 11 of the 20 patients were female and 12 of the cysts presented on the right side. The length of history was usually short, the longest being 16 months, the shortest being one month, and the mean being five months.

Most patients complained of constant neck swelling (55 per cent). However, an appreciable number presented with a history of rapidly increasing neck swelling and pain which had responded to antibiotics (40 per cent). In 17 patients (85 per cent) the cyst was anterior to the upper third of the sternocleidomastoid, in two patients (10 per

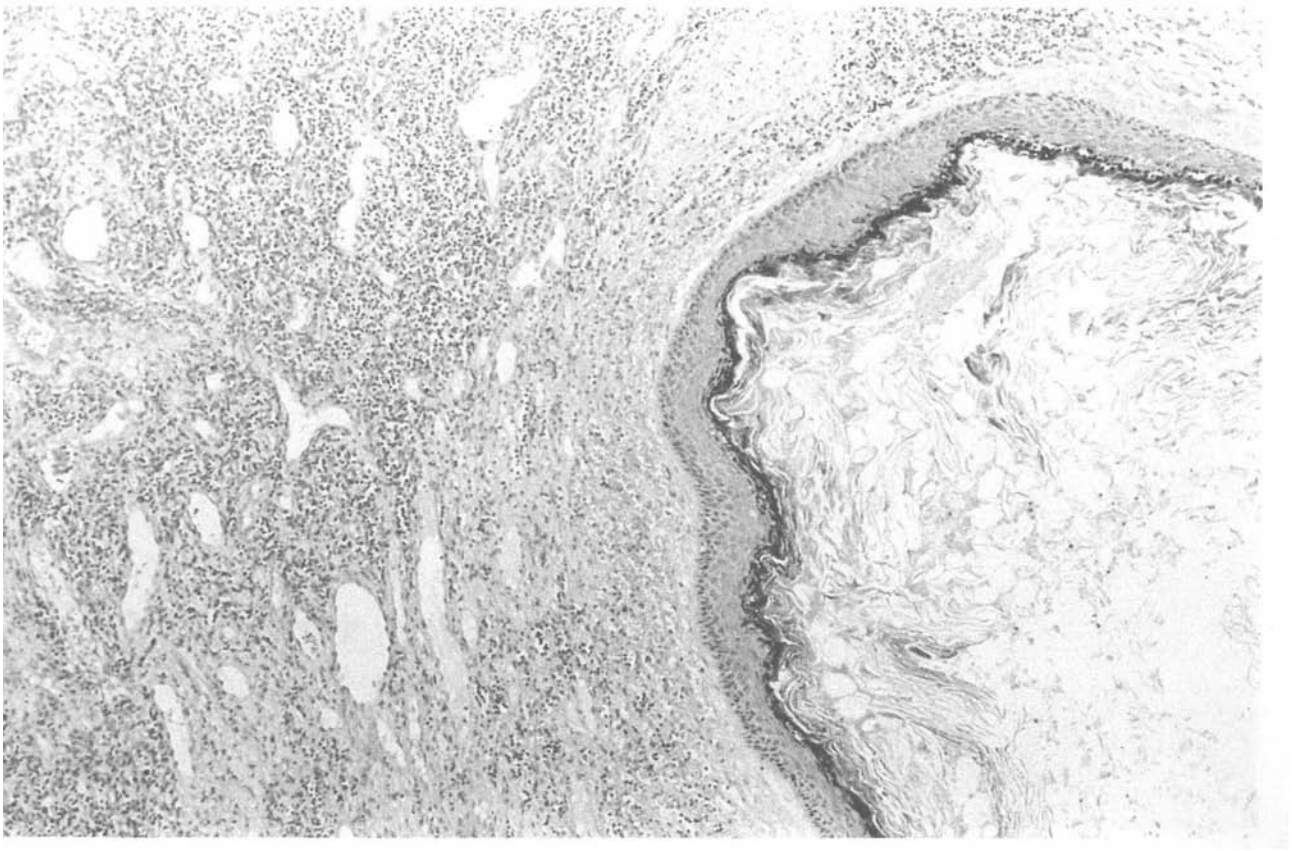

Fig. 5
Photomicrograph of a medium power view of the lining epithelium of a lateral cervical cyst. (H & E; × 80).

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TABLE II
THE SITE OF LATERAL CERVICAL CYSTS: THIS STUDY AND A REVIEW OF PREVIOUS STUDIES

	Golledge & Ellis (1994)	Neel & Pemberton (1945)	King (1949)	Bhaskar & Bernier (1959)
Anterior/deep to upper 1/3 of sternomastoid	85	73	74	41
Anterior/deep to middle/lower 1/3 of sternomastoid	10	22	16	15
Submandibular triangle	5	0	7	10
Parotid region	0	0	3	3
Anterior triangle	0	0	0	15
Posterior triangle	0	0	0	3

cent) it was anterior to the middle third of the sternocleidomastoid and in one patient (five per cent) it lay in the submandibular triangle (Table II). All 20 cysts were treated by excision under a general anaesthetic. In no case was a tract or cord connecting the cyst to the skin or pharynx noted. Most of the cysts (17) were found to lie just deep to the investing cervical fascia and sternocleidomastoid. In two cases the cyst was adherent to the internal jugular vein, hence resection of the vein was necessary. In one case the cyst was adherent to the submandibular gland, which was removed in addition to the cyst, and found to be histologically normal.

Review of the histology showed all 20 cysts to have a stratified squamous epithelial lining (Figures 4 and 5). Of particular interest was the finding that, in all cases, the wall of the cyst contained lymphoid tissue with recognizable lymphoid follicles (Figures 3 and 6). In half of the cysts the lymphoid follicles showed evidence of reactive hyperplasia.

Microscopy of the cyst contents showed pus in five (25 per cent) cases. However, significant bacterial growth only occurred in one case (five per cent). This is probably related to the widespread preliminary use of antibiotics by the referring GPs.

Discussion

At the outset it should be made clear that the aetiology of the lateral cervical cysts remains unproven. It is possible that there are in fact a number of different mechanisms by which the cysts, which arise in the lateral aspect of the neck, are formed.

In order to discuss the aetiology of lateral cervical cysts it is necessary to have a clear definition. Since definition and aetiology are inseparably intertwined, there have been changing opinions, as theories regarding aetiology have evolved, concerning which cysts should be included in the 'branchial collection'.

Early descriptions of cervical cysts concentrate on anatomical site and consistency. Bailey (1922) described the cyst as a cystic swelling which lies on the deep surface of the upper half of sternocleidomastoid. Howie and Proops (1982) reiterated this, defining 'branchial' cysts as lesions which lie at the junction of the upper third and lower two-thirds of the sternocleidomastoid. However, this only stimulated a number of case reports describing cysts which arise outside this classical position, in particular cysts associated with the parotid and submandibular salivary glands (Fujibayashi and Itoh, 1981; Scott, 1987;

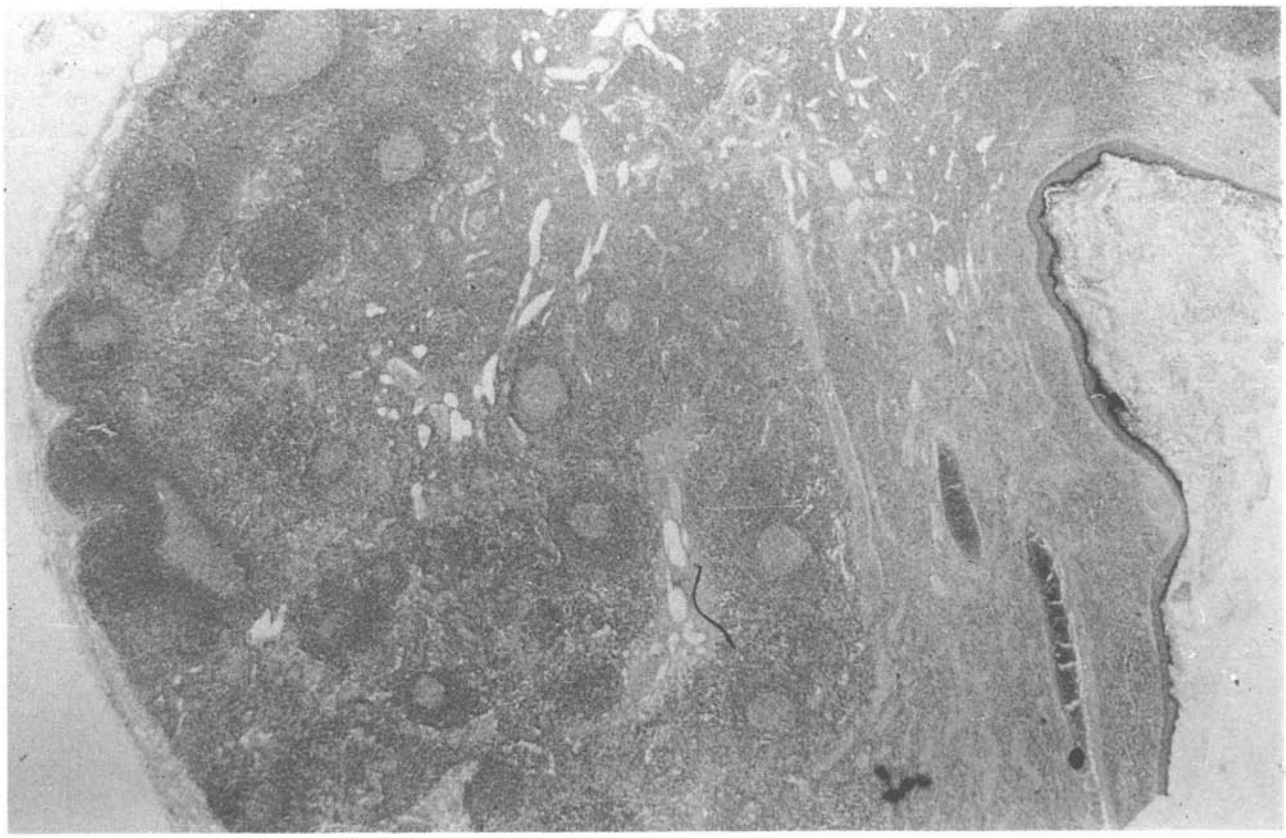

Fig. 6
Photomicrograph of a low power view of the wall of a lateral cervical cyst, illustrating the lymphoid follicles. (H & E; × 10).

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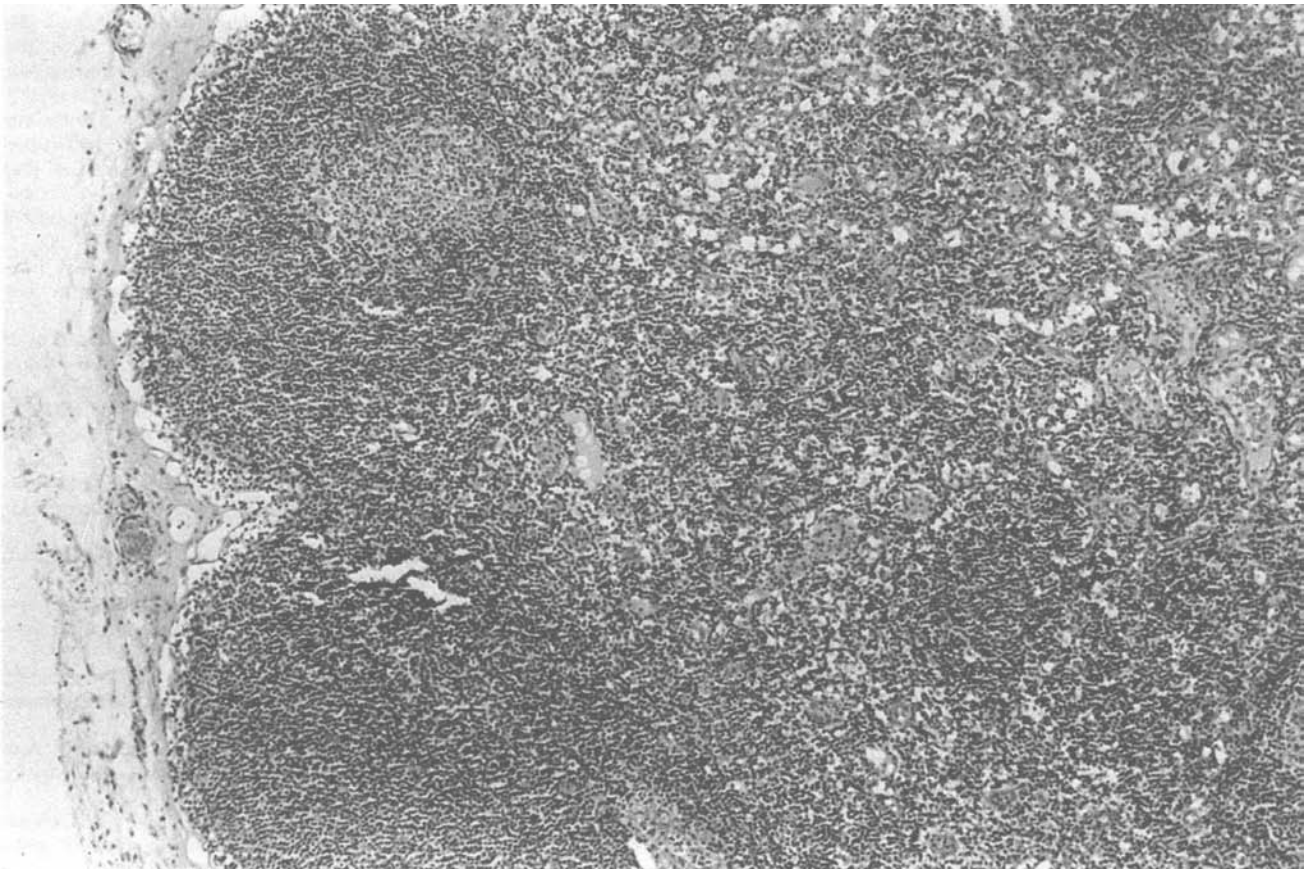

Fig. 7
Photomicrograph of a medium power view of the wall of a lateral cervical cyst, illustrating the appearance of a reactive lymph node. (H & E; × 80).

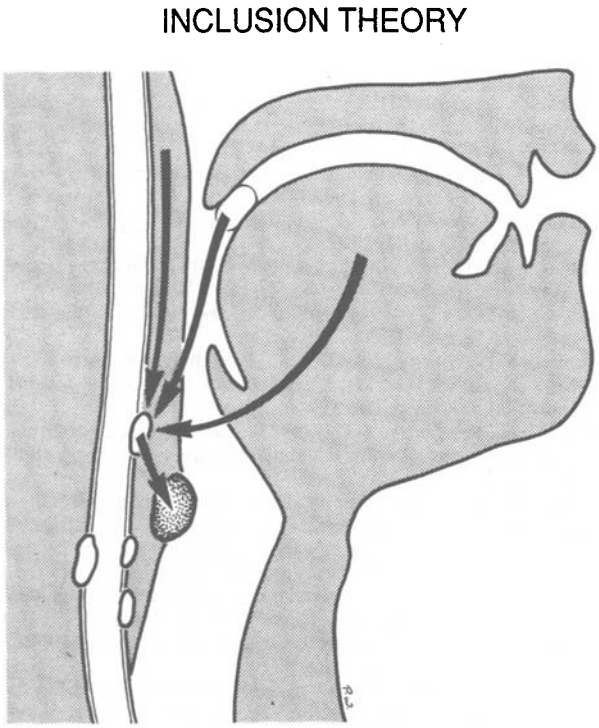

FIG. 8
Diagram to illustrate how squamous epithelium from the palatine tonsil could enter a cervical lymph node via the lymphatics and stimulate cystic transformation to a lateral cervical cyst.

Wyman *et al.*, 1988). In more recent publications, histology has become increasingly important in the definition of lateral cervical cysts (Bhaskar and Bernier, 1959; Maran and Buchanan, 1978). Thus, as made clear by King (1949), two factors are important in the definition of a lateral cervical cyst. Firstly, position, the cyst must lie outside the midline, i.e. any position in the lateral aspect of the neck; secondly, the histology – the cyst is lined with squamous or rarely columnar epithelium and is surrounded by lymphoid tissue. These defining features of lateral cervical cysts are illustrated by Figures 3 to 7.

Of the four hypotheses regarding the aetiology of lateral cervical cysts the evidence strongly favours the 'Lymph Node Theory'.

Most patients present in the third decade or older (Table I). In contrast to the classical description, a considerable number of cysts lie at sites outside the upper third of the sternocleidomastoid (Table II). Those presenting inferior to the hyoid bone, in the parotid region and in the submandibular triangle cannot easily be explained by the congenital theories. At operation it is usually not possible to demonstrate a tract connecting the cyst to the pharynx or skin, as would be expected from the congenital theories.

Careful interpretation of the embryological facts, as defined by Frazer (1923 and 1926), has revealed very little to support the congenital theories. Frazer described the relationships of the pharyngeal clefts, pouches and the cervical sinus to the nerves and vessels in the neck. He pointed out that if any of these embryological structures were to enlarge to form vestigial structures in the adult, these anatomical relationships would remain fixed. Stud-

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ies since that time, as in this review, have failed to demonstrate these constant anatomical relationships.

The principal support for the 'Lymph Node Theory' of aetiology comes from the histological studies. In most cases the cyst is surrounded by lymphoid tissue (Figure 3). The characteristics of this lymphoid wall are identical to those of a lymph node (Hirota *et al.*, 1989). Lymphoid follicles, marginal sinuses, immunoglobulins and T-lymphocytes are present (Crocker and Jenkins, 1985). The distribution of cells corresponds to that of a reactive lymph node (Figure 7).

While it appears clear that cervical cysts develop from cervical lymph nodes, the mechanism by which this occurs is less well explained. Bhaskar and Bernier (1959) suggest that the cystic alteration of cervical lymph nodes is stimulated by trapped epithelium. This hypothesis has become known as the 'Inclusion Theory'. They originally suggested three possible sources for these epithelial inclusions: branchial cleft, pharyngeal pouch and parotid gland. Since that time a number of other possible sites have been put forward. However, only two sites have any evidence to support them. Firstly the parotid and submandibular salivary glands, the source favoured by Bhaskar and Bernier (1959). They emphasize that the parotid gland develops at the same time as the cervical lymph nodes in the embryo, and that a number of cervical cysts present associated with salivary glands (Table II).

We favour a different source for the included epithelium, i.e., the palatine tonsil. Wild *et al.* (1987) studied the keratin content of the epithelium lining cervical cysts. They found it to be homologous to upper digestive tract epithelium, in particular palatine tonsil. Studies of normal cervical lymph nodes have shown squamous epithelium lining marginal sinuses, which may be distended or adjacent to definitive cysts (King, 1949). This may represent an early stage in the development of a lateral cervical cyst.

We propose that epithelium from the palatine tonsil enters cervical lymph nodes during adult or infant life and causes cystic transformation resulting in a lateral cervical cyst (see Figure 8).

Conclusion

There is no evidence to support the theory that lateral cervical cysts result from abnormalities of the branchial apparatus. Instead these cysts appear to be cystic lymph nodes. The mechanism which stimulates this cystic transformation of cervical lymph nodes is uncertain. However, it is possible that epithelium from the palatine tonsil enters a cervical lymph node, lines a lymphoid sinusoid and encourages its expansion into a lateral cervical cyst.

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