Benign Lymphoma (Castleman)
Angionodular hyperplasia of cervical lymph node

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Castleman in 1954 described hyperplasia of mediastinal lymph gland, which roentgenologically and histopathologically resembled thymoma and which he simply called hyperplasia of the lymph gland. This lesion, the etiology of which is unknown, has subsequently been given various names, such as lymphonodal hamartoma (Abell, 1957), tumour-like proliferation of lymphoid tissue (Cohen, 1957), giant intrathoracic lymph node (Mason, 1959), follicular lymphoreticuloma (Zettergren, 1961), angiofollicular mediastinal lymph node hyperplasia (Harrison and Bernatz, 1963), and angiomatous lymphoid hamartoma (Tung and McCormack, 1967). The last study mentioned contains a comprehensive list of previous papers dealing with this topic. Altogether, sixty tumours, most of them located in the mediastinum, have been reported. Only seven tumours of the cervical region have previously been described.

Case report

The patient was a 32-year-old man who had otherwise been generally healthy. He was seen in the Outpatient Department of Oulu University Ear, Nose and Throat Clinic in June 1970 because of a lump at the left side of his neck, gradually growing over a period of ten years. A tumour of the size of a hen's egg bulging slightly into the pharynx and moving easily amid the surrounding organs was found at the anterior edge of the sternocleidomastoid muscle. Laboratory findings and thorax X-ray were normal. Roentgen examination of the paranasal sinuses revealed bilateral maxillary sinusitis which did not heal with antral irrigations and a bilateral Caldwell-Luc procedure was performed in October 1970.

The cervical tumour was operated in November 1970. The tumour was located between the common carotid artery and internal jugular vein and in gross examination it resembled a neurinoma (Fig. 1). Histologically it was diagnosed as lymphoma benignum Castleman. The patient has been symptom-free postoperatively and has been followed regularly, the most recent check-up being in December 1971.

Histopathological examination

Selected parts of the specimen were fixed in formalin and embedded in paraffin. The 5μ sections were stained with haematoxylin-eosin, Gomori and Azur-eosin stains and a study with serial sectioning was performed with Lillie allochrome stain. The lesion was enveloped in a thin connective tissue capsule with slight scattered lymphocytic infiltrations. The lymph node was not divided into a cortex and a medulla as normally; instead there was highly vascular stroma and small groups of follicle-like nodules dispersed around the lymphatic tissue (Fig. 2). Some nodules were composed exclusively of mature small

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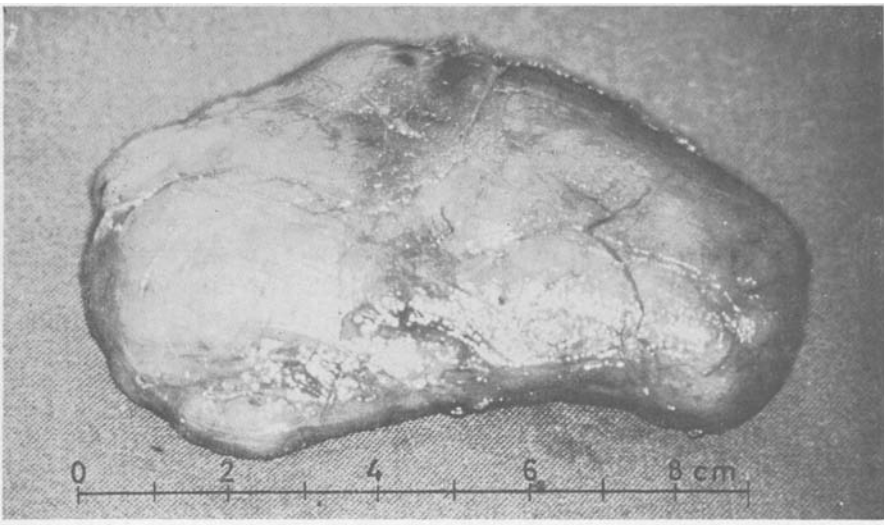


FIG. 1.
The removed tumour.

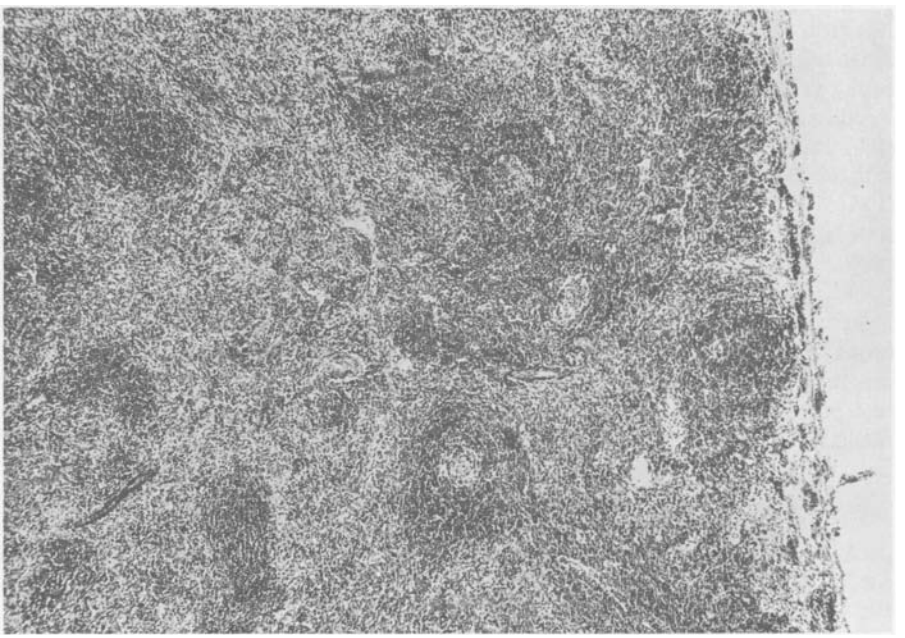


FIG. 2.
A typical view of the nodular lesion covered by a thin capsule. (Haematoxylin-eosin, × 40.)

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lymphocytes, others had a light central part which consisted of concentrically arranged large reticular cells with a large vesicular nucleus and a prominent nucleolus. Every reticular cell was surrounded by an argyrophilic fibre network, and an open blood vessel was often seen in the middle of the area. PAS-positive material was occasionally seen in the capillary walls. In the reticulocellular centres or just adjacent to it PAS-positive refractile, dense-looking particles with differently shaped angular configurations occupied the capillary walls. Nothing suggestive of Hassall's corpuscles of thymus was found in the sections (Fig. 3).

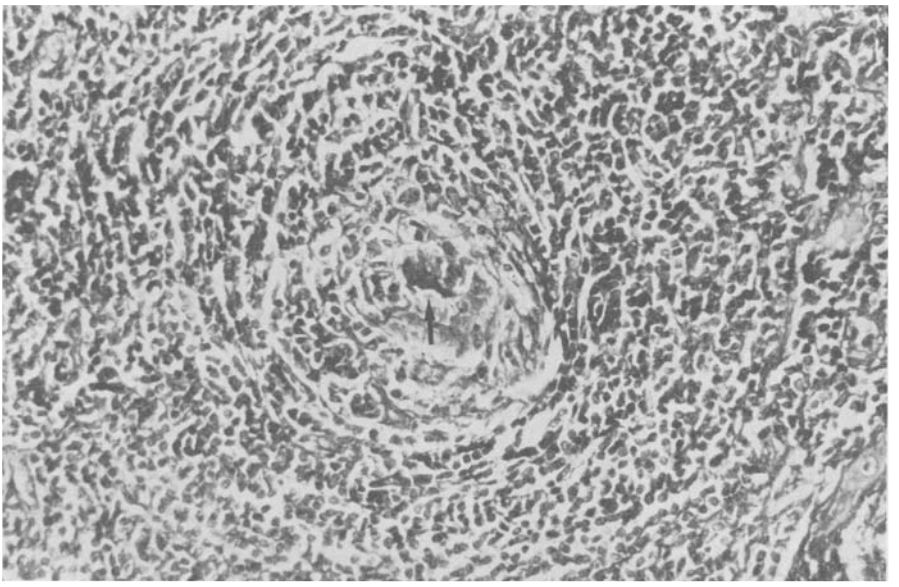


FIG. 3.

The centre of the nodule made up of concentrically arranged reticular cells. A PAS-positive dense particle can be seen (arrow). (Lillie allochrome, ×400.)

The internodular stroma consisted of highly vascular lymphoid tissue (Fig. 4). The endothelial tissue of blood vessels was hyperplastic and the vascular walls showed hyalinization, fibrosis, even here casual occurrences of PAS-positive material was noted. No plasma cells or calcific deposits were noted in the stroma.

Comment

The etiology of this disease remains unknown. Castleman (1954) suggested that the condition is due to local, non-neoplastic hyperplasia of the lymph gland in which capillary proliferation, follicular hyperplasia, and infiltration of both plasma cells and eosinophils take place. He thought that these findings, characteristic of chronic non-specific lymphadenitis, were due to a slow inflammatory process of several years duration. The chronic bilateral maxillary sinusitis noted in the present case would fit this assumption, but it is doubtful whether its

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duration would have lasted for more than ten years. Zettergren (1961), on the other hand, regarded the lesion as a benign neoplastic process and suggested that it should be called follicular lymphoreticuloma, because the germinal centres were different from the corresponding germinal centres of chronic infection. He further deemed it impossible that processes initiated by infection would be restricted to one single lymph gland.

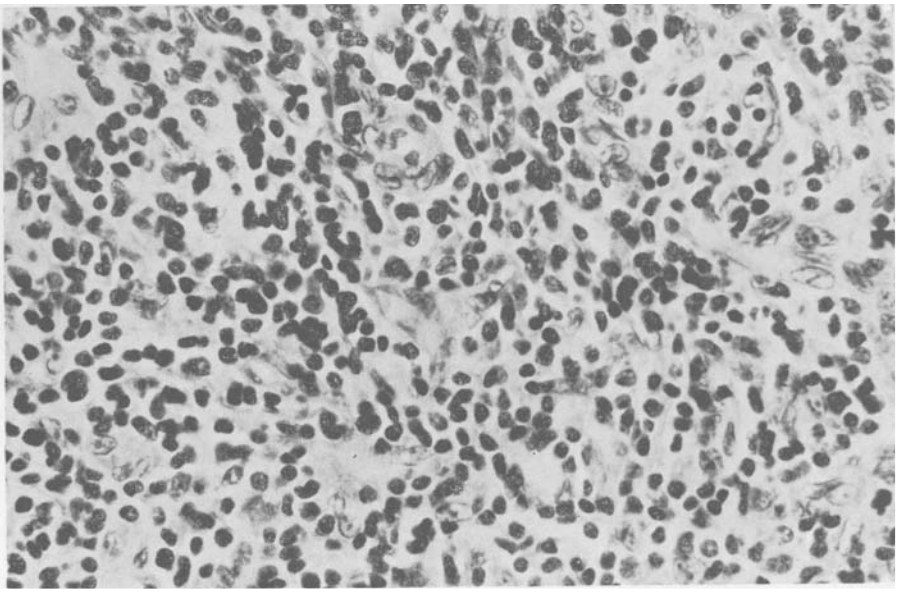


FIG. 4.
Internodular tissue consisting mostly of lymphocytes with abundant number of capillaries. (Haematoxylin-eosin, ×400.)

According to Harrison and Bernatz (1963), a primary defect in a hypertrophic gland may be developmental, possibly even mechanical. The relationship between mediastinal lymph gland enlargement and thymic tissue is not known, but some patients have shown myasthenic symptoms after the resection of the gland. The process may therefore be associated with genuine thymoma.

According to the mechanical theory, elevated mediastinal pressure, for example, may lead to disturbances of the normal architecture of the lymph gland. Additional factors, such as chronic inflammation, may bring about reactive hyperplasia, thereby leading to final malformation and enlargement of the lymph gland.

Tung and McCormack (1967) thought that vascular proliferation leads to distortion of initially normal follicles. In the present case the follicles were abnormal, and we therefore prefer to call them nodules. In our case such a nodule always had a central blood vessel with lightly staining pericapillary reticular cells which were concentrically covered by a lymphocytic coating. The process could most accurately be described by the name 'angionodular hyperplasia'.

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The origin of the PAS-positive material noted in the capillary walls remain obscure. It might be due to precipitation of plasma and perhaps γ-globulins on the vascular walls, which then might lead to the formation of the material observed. There is no doubt that the process in question is always a benign one and the reported observations suggest that the disease does not recur even after an incomplete resection.

Summary

A tumour which had been growing slowly over a period of ten years was removed from the neck of a 32-year-old male. The tumour was histologically diagnosed as a benign lymphoma of Castleman. It is a rare disease, only seven such tumours having previously been found in the neck. Histologically the tumour was characterized by atypical richly vascularized nodular lymphoid tissue, and the process could be termed as angionodular hyperplasia. Hyaline PAS-positive material was noted particularly in the nodule. The tumour is considered to be a benign disease with unknown etiology.

Acknowledgements

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