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Dynamic CT Findings of Bilateral Castleman Disease in the Neck : A Case Report[1]

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We present a case of hyaline vascular type Castleman disease involving the bilateral cervical lymph nodes. To our knowledge, no previous case of this localized form of the disease has been reported. Dynamic CT demonstrated a hypervascular pattern of enhancement, with central less enhanced areas that corresponded histologically to fibrosis. For the diagnosis of this uncommon lymph node disease, these findings might be helpful.

Index Words : Neck, CT
Lymphatic system, hyperplasia
Lymphatic system, neoplasms

Castleman disease is an uncommon lymphoproliferative disorder most commonly found in the mediastinum (1). It may, however, occur in any area where lymphatic tissues are found such as the lung, neck, axilla, mesentery, pelvis, and retroperitoneum (2-6). It is estimated that 71% of cases occur in the thorax and less than 10% in the neck (2, 3). Although the literature contains several reports of isolated cervical Castleman disease (3, 6), bilateral disease confined to the neck has not, to the best of our knowledge, been described. We report a case of Castleman disease involving the bilateral cervical lymph nodes with emphasis on the dynamic CT findings.

Case report

A 57-year-old man was referred to our hospital on account of painless bilateral neck masses. He had first noticed a mass on the left side two months previously, and another mass was subsequently palpated on the right. The size of both masses had progressively increased. Antibiotics were administered, but the patient did not respond, and incisional biopsy performed at that time revealed only inflammation. Physical examination and laboratory tests showed no remarkable findings except for two palpable masses on both sides of the upper cervical neck with the left one larger than the right. He denied any mass-related symptoms, and chest radiography and abdominal ultrasonography were unremarkable.

CT of the neck was performed using a Somatom Plus S (Siemens Medical System, Erlangen, Germany). CT scans obtained before the use of contrast material showed a 6×4cm lobulated soft tissue mass on the left of the neck and a smaller 4×3cm ovoid soft tissue mass on the right. No intra-mass calcification was demonstrated; single-slice dynamic scanning performed after intravenous administration of contrast material demonstrated prominent enhancement of both masses. Less enhanced central portions radiated peripherally, and were more conspicuous in the mass on the left (Fig. 1A). While the masses were not enhanced to the same degree as the vessels, a time-density curve revealed that peak enhancement of the masses occurred almost simultaneously with that of the vessels. It also demonstrated that contrast material was retained much longer in the masses than in the vessels (Fig. 1B). The patient underwent excision of the left neck mass, and histologic examination revealed hyaline vascular type Castleman disease. The central, less

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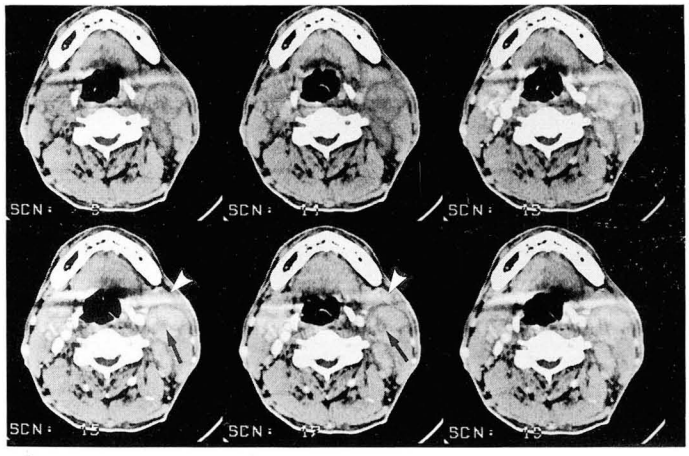
enhanced areas seen on dynamic CT corresponded to fibrosis (Fig. 1C). The mass on the right of the neck also showed the same histopathology, and was subsequently excised.

Discussion

Since Castleman et al. (1) reported 13 patients with the characteristic histological features of a localized mediastinal lymph node hyperplasia in 1956, there have been numerous reports of this uncommon lymphoproliferative disorder involving various sites of the body (2—6). Although there have been many theories about the etiology and pathogenesis of Castleman disease (2), none satisfactorily explains the various spectrums of this disease, to which a plethora of names such as lymph nodal hamartoma, follicular lymphoreticuloma, angiofollicular lymph node hyperplasia, angiomatous lymphoid hamartoma, benign giant lymphoma, and giant lymph node hyperplasia have been applied.

Histologically and clinically, there are two types of Castleman disease (2). The hyaline vascular type is more common, and accounts for 90% of cases. It is usually a solitary lesion and the patient is usually asymptomatic unless adjacent structures are impinged. Histologically it is characterized by small hyaline-vascular follicles and interfollicular capillary proliferation which enhances considerably on CT, MR, or angiography. The hyalinized capillaries are surrounded by sheets of lymphocytes with an onion-skin appearance (2). The majority of cervical lesions are of this type, as are the lesions in the present case. Less common plasma cell type can be either solitary or multicentric and in approximately half of all cases is associated with systemic manifestations such as fever, anemia, elevated erythrocyte sedimentation rate, hyperglobulinemia,



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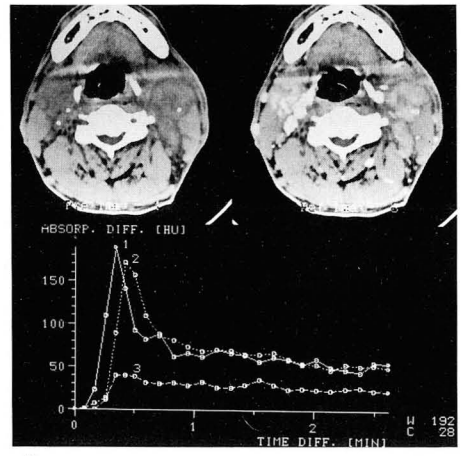

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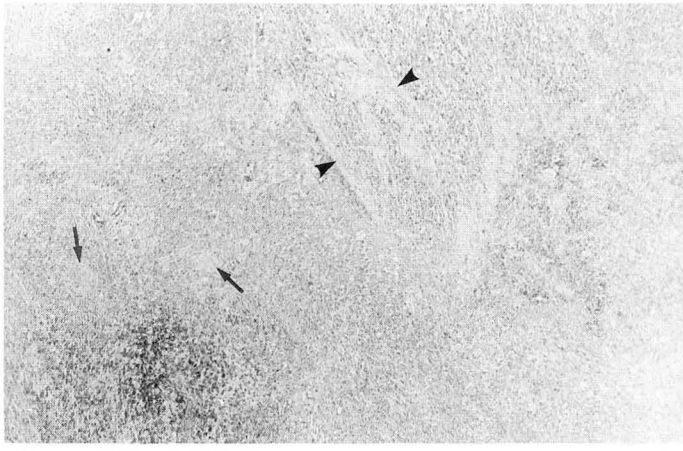

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Fig. 1. Bilateral cervical Castleman disease in a 57-year-old man.

A. The selected serial dynamic CT scans 10 seconds after IV administration of 60 ml of iodine contrast material with injection rate of 2 cc/s and demonstrate bilateral cervical lymphadenopathies with enhancement. Note the central and radial less enhanced area in the left-sided mass (arrows). The left submandibular gland that enhances to the same degree as lymphadenopathies is displaced anteriorly (arrowheads).

B. The time-density curve shows that peak enhancement of the left-sided mass (3) occurs nearly same time as that of the carotid artery (1) and the jugular vein (2). However, the wash-out of the contrast material from the mass is much more delayed. Histogram of the right-sided mass revealed exactly same appearance (not shown).

C. Photomicrograph of the resected left-sided lymph node demonstrates lymphoid follicular hyperplasia with small germinal centers containing hyalinized vessels (arrows). Note prominent fibrosis adjacent to those lymphoid follicles (arrowheads). Hematoxylin & eosin, original magnification ×40.

and hypoalbuminemia. Histologically, it is characterized by large follicles with sheets of mature plasma cells in the interfollicular tissue. No particular vascularization or hyalinization is evident in these follicular centers (2), which on imaging show a lesser degree of enhancement than do the hyaline vascular type. It has been reported that the multicentric form of Castleman disease is frequently associated with various immunodeficiency states (7). Its clinical course is usually aggressive, and the prognosis is poor. While complete surgical excision is the treatment of choice for localized Castleman disease, the most effective treatment for the multicentric form has not yet been established.

Although bilateral, the present case showed no evidence of systemic disease either clinically or radiologically. To the best of our knowledge, there has been no reported case of the localized form of bilateral cervical Castleman disease. Differential diagnoses for bilateral neck masses include lymphoma, tuberculous lymphadenitis, nonspecific reactive lymph node hyperplasia related to infections, and metastatic lymphadenopathy.

The pattern of enhancement seen on CT, particularly dynamic CT, can provide valuable information for differential diagnosis. Although dynamic CT has proved to be useful for the evaluation of hypervascular lesions in the head and neck (8), we are not aware of any published report which systematically described the results of dynamic CT of various lymph node diseases. Lymphoma usually produces homogeneous nodes which on CT rarely show enhancement (9). To date, we have visualized on dynamic CT six cases of lymphoma involving the cervical lymph nodes. In all cases, the time-density curve during scanning was rather haphazard, and peak enhancement was usually less than that of the present case. Even though our experience is so far limited, dynamic CT might be a useful adjunctive method for the diagnosis of lymphoma. Tuberculous lymphadenitis was also unlikely, since no central necrosis was demonstrated on CT, and the node was large. Although tuberculous adenitis can be manifested in the early stage as a significantly enhanced solid mass, central necrosis tends to develop as the disease progresses (10). In the CT study of mediastinal tuberculous lymphadenitis, Im et al. (11) reported that in nodes larger than 2 cm in diameter the central area was invariably of low density. Although acute infection with reactive regional adenopathy could also account for the enhanced lymph nodes seen on CT (12), the patient's lack of signs of infection and the large masses are unusual for hyperplastic lymph nodes, the greatest diameter of which is normally less than 2 cm (9). Because there was no history of primary tumor and no central necrosis within such a large mass, metastatic lymphadenopathy also seemed to be unlikely. A hypervascular metastatic tumor such as papillary thyroid carcinoma, hypernephroma, or hepatocellular carcinoma can, however, also be seen on CT as a significantly enhanced mass (12). Other uncommon diseases to be differentiated include angioimmunoblastic lymphadenopathy (with dysproteinemia), angiolymphoid hyperplasia with eosinophilia (Kimura disease), histiocytic necrotizing lymphadenitis (Kikuchi disease), Kaposi's sarcoma, sarcoidosis, and sinus histiocytosis with massive lymphadenopathy. The first four of these can show a significant degree of enhancement on CT.

In the present case, the centrally located less enhanced areas seen on dynamic CT were histologically correlated with fibrosis, which is reported to have been shown by CT or MR imaging (4, 6). All reported cases were the hyaline vascular type.

On the basis of our reports and those of others, dynamic CT might be helpful in diagnosing the hyaline vascular type of Castleman disease.

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양측 경부에 발생한 Castleman씨병의 역동적 CT 소견 : 1예보고[1]

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이 증례는 양측 경부의 임파절에 발생한 초자질 혈관성 Castleman 씨병에 관한 것이다. 역동적 CT는 병변내 과혈관에 의한 조영 증강 소견뿐 아니라 병변 중심부의 섬유화에 의한 조영 결핍의 소견을 잘 보여 주었다. 이러한 역동적 CT 소견은 임상적 또는 방사선학적으로 비슷한 양상을 보이는 병변의 감별 진단에 유용하리라 생각된다.