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Castleman's disease presenting as a midline neck mass

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SUMMARY. We report the case of a 13-year-old girl who presented with a painless midline submental mass. Excision biopsy confirmed Castleman's disease of the hyaline-vascular type. This unusual condition needs to be considered in the differential diagnosis of masses arising in the neck. © 2003 The British Association of Plastics Surgeons. Published by Elsevier Science Ltd. All rights reserved.

Keywords: castleman's disease, midline neck mass.

Castleman's disease is a rare benign condition of unknown aetiology that can present as a localised mass or in a more aggressive multicentric form. The localised form of the disease tends to present as progressive painless slow-growing lymph-node enlargement. The majority of cases involve the mediastinum, but the head and neck is the second most common site of occurrence, where it can pose a diagnostic challenge. Diagnosis is possible only by histological examination. Complete excision is the treatment of choice and tends to be regarded as curative for the localised form of the disease.

Case report

A 13-year-old girl was referred to the Department of Oral and Maxillofacial Surgery by her general medical practitioner because of an unsightly midline neck mass. The mass had been present for 2 years, and was first noticed by the patient after a minor trauma to her chin. Examination revealed a 2 cm firm painless mobile mass in the midline of the submental triangle. The mass did not rise on swallowing. The patient was euthyroid and in good health.

MRI demonstrated a 2 cm well-defined lesion in the subcutaneous fat immediately posterior to the mental symphysis. Coronal images confirmed that the lesion was lying between the anterior bellies of the digastric muscle and anterior to the mylohyoid muscle. The lesion was of intermediate T1 signal intensity (Fig. 1) and elevated T2 signal intensity, with a pattern suggesting soft tissue. The MRI appearances were not consistent with either a lipoma or a ranula, and we decided to carry out an excisional biopsy of the mass under general anaesthesia. At operation, the lesion was found to be solid and adherent to the digastric and mylohyoid muscle bellies.

Histology of the specimen showed the typical features of the hyaline-vascular type of Castleman's disease (Fig. 2). The lymph node was enlarged and contained numerous germinal centres, many of which were small and hyalinised, containing many follicular dendritic cells. The mantle zone was expanded and composed of tightly packed lymphocytes in a concentric arrangement around the germinal centres. The interfollicular area contained many small hyalinised vessels.

Discussion

Castleman's disease was first described in 1954 as a form of localised mediastinal lymph-node hyperplasia characterised by germinal centre and capillary proliferation.[1] Clinically and histologically, two forms of the disease are now recognised. One is localised and usually of the hyaline-vascular type, the other is more aggressive, multicentric and always of the plasma cell type. The aetiology is uncertain, but interleukin-6 is thought to be responsible for the clinical and biological signs[2]—fever, weight loss, elevated C-reactive protein, hypergammaglobulinaemia and microcytic anaemia. There is no sex predilection and young adults are particularly affected; only 94 cases, which have generally been of the localised form of the disease, have been described in children.[3–6]

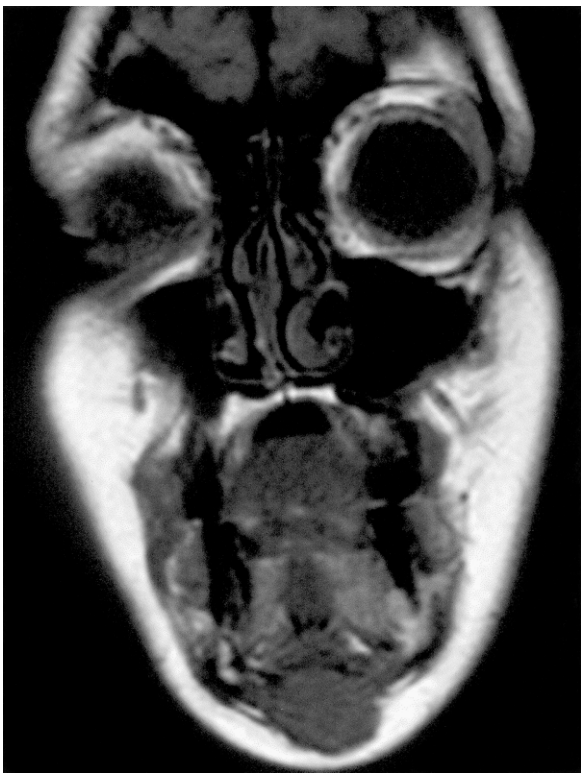

Figure 1—Coronal T1-weighted MRI showing a midline lesion of low signal.

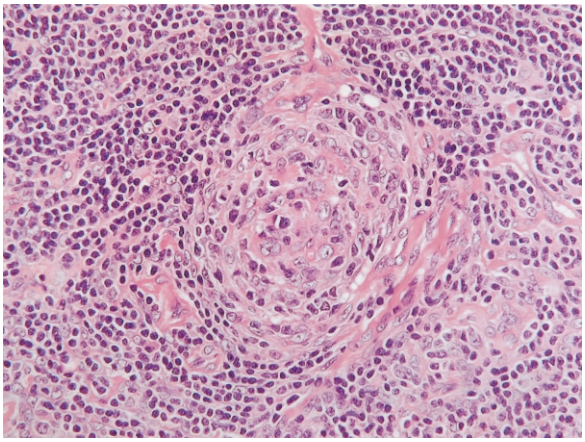

Figure 2—Photomicrograph showing a hyalinised germinal centre and a concentric arrangement of small lymphocytes (H&E × 120).

The aetiology of the disease is still unclear; suggested explanations include a hamartomatous lymphoid origin and a hyperplastic response to infection, although serological and tissue-culture studies have been inconclusive.[7] The neck is involved in only 14% of all cases, with the mediastinum being the most common site, accounting for 60% of cases.[8] When Castleman's disease occurs in the neck the lesions have been described as lateral rather than midline, with 52 out of the 63 reported cervical cases occurring in nodes under the sternocleidomastoid muscle. Other reported sites include the parotid gland (six cases), and there have been individual reports of cases in the floor of the mouth, the submandibular gland, the larynx, the palate and the parapharyngeal space.[9,10] To our knowledge, Castleman's disease has not previously been reported in the midline of the neck. The diagnosis is made by excision biopsy, and MRI may be helpful, particularly in showing the presence of stellate hypointensities on T2-weighted images, which may represent fibrosis in the lymph nodes.[4,10] These findings were not present in our case. Other diagnostic tests that may be useful when systemic symptoms are present include serum interleukin-6 assay, C-reactive protein and gammaglobulin assay. Fine-needle aspiration cytology can be considered where simple excision is not possible, but may lead to conflicting information.[7]

The plasma cell type of the disease is usually multicentric, may be associated with the systemic symptoms

mentioned and can occasionally progress to lymphoma. All patients with the plasma cell variant need full investigation, including CT of the chest.

In summary, although the majority of cervical Castleman's disease occurs laterally, this case demonstrates that Castleman's disease should be considered in the differential diagnosis of a solitary midline neck mass. The treatment for localised Castleman's disease is excision, which is regarded as curative.

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Pregnancy as an autologous tissue expander for closure of an abdominal-wall defect

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SUMMARY. We report the reconstruction of a complex abdominal-wall defect using expanded skin from pregnancy. Wound closure was achieved using a vertical abdominoplasty. © 2003 The British Association of Plastics Surgeons. Published by Elsevier Science Ltd. All rights reserved.

Keywords: tissue expansion, pregnancy, abdominal wall.

The use of tissue expansion for the reconstruction of defects in the abdominal wall is well described.[1–3] We report an unusual case of abdominal-wall reconstruction making use of pregnancy as an autologous tissue expander.

Case report

A 33-year-old woman was admitted with an abdominal-wall desmoid tumour measuring 15 cm × 8 cm. After tumour resection, the abdominal-wall defect was closed using a free latissimus dorsi myocutaneous flap over a mesh repair. The flap did not survive and was removed 24 h postoperatively. Twelve days later there was sufficient granulation tissue over the mesh to apply a skin graft and thereby close the wound. She was discharged for outpatient management with an abdominal support.

Seven months after this surgery, she presented to the clinic 2 months pregnant. There was a small discharging wound overlying the distal end of the mesh. It was decided