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CASE REPORT

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A case of neck xanthogranulomatous inflammation-suspected malignant tumor

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Abstract

Xanthogranulomatous inflammation (XGI) is an uncommon chronic inflammatory disease. A 59-year-old male presented with a neck mass which had been diagnosed as an undifferentiated carcinoma. From CD68 staining, XGI was confirmed. It is important to consider the possibility of XGI for a neck mass mimicking a malignant tumor.

KEYWORDS

CD68, neck mass, xanthogranulomatous inflammation

1 | INTRODUCTION

Xanthogranulomatous inflammation (XGI) is an uncommon chronic inflammatory disease mainly occurring in the kidney and gallbladder.[1,2] Its characteristic histology includes formy histiocytes and giant cells. The head and neck region is an uncommon site for XGI, with only three cases having been reported to date.[3-5] Two of these cases were cystic in origin. Here, we report a case of a neck tumor lesion that was mistaken for a malignant tumor at a previous hospital and ultimately diagnosed as XGI without cystic lesion.

2 | CASE PRESENTATION

A 59-year-old male presented with a mass lesion in the left lateral neck. He had noticed neck swelling 3 months before visiting our hospital. One month before visiting our hospital, he had a neck biopsy at another hospital that indicated undifferentiated carcinoma. He was subsequently referred to our hospital. He had no medical history or family history. During the head and neck examination, a roughly 5-cm elastic hard mass with about a 10 mm ulcer in the center of the mass was found, but there were no cystic findings (Figure 1A). CT scan

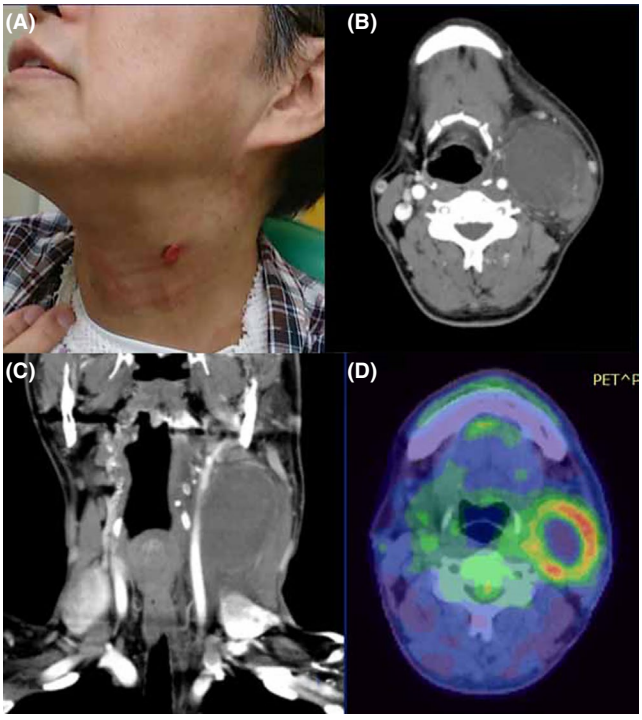

FIGURE 1 A, Ulcerated neck mass was seen. B, C, CT scan showed low-density mass with slight ring enhancement. D, PET scan showed the ring-like accumulation with no other abnormal lesions

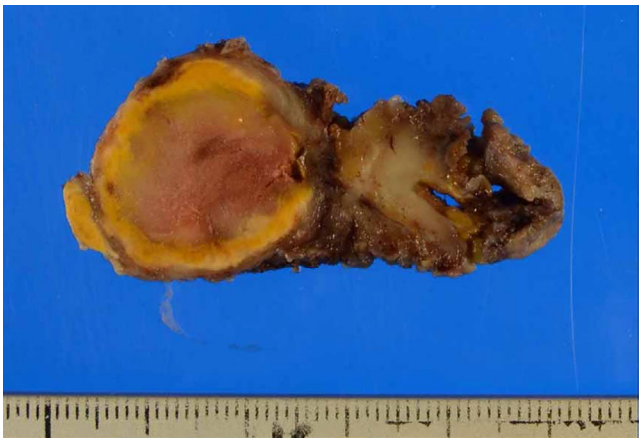

FIGURE 2 Ring-like yellowish mass with central necrosis was seen

revealed a low-density mass with slight ring enhancement (Figure 1B,C).

PET scan showed the ring-like accumulation with no other abnormal lesions (Figure 1D). According to the laboratory data, tumor markers (SCC, CES, and IL-2) were negative, T-SPOT for tuberculosis was negative, and bacteria examination was negative. Also, gastrointestinal fiberscope (GIF) was negative for tumor.

In our clinic, re-biopsy was performed. The result was granulation tissue with no malignancy. To make the diagnosis, tumor extirpation was planned, and the surgery was performed under general anesthesia. The mass was partially adherent to the sternocleidomastoid muscle (SCM) and internal jugular vein (IJV). Ulcerated skin, SCM, and IJV were partially transected, and the cutting slice showed the yellow mass (Figure 2).

Microscopic examination revealed a fibrous, encapsulated mass with many formy cells and giant cells in the peripheral area. The inner space was necrotic (Figure 3A). In the peripheral area, a lymphoid nodule was seen (Figure 3B). Coagulative necrosis and hemosiderin deposits were abundant (Figure 3C). Some lymphoma-like, large nuclear cells were seen in the peripheral area (Figure 3D). In immunostainings, there were CD3- and CD20-positive cells in the peripheral area, but they were inflammation cells (Figure 4A,B). There were no epithelial cells. The main cells of the tumor were CD3, CD20, CD30, and AE1/3 negative. In the sections of the yellow mass, the cells of the infiltrate were CD68 positive (Figure 4C), while in the necrotic area, the cells were CD68 negative (Figure 4D).

From those findings, xanthogranulomatous inflammation was diagnosed. No recurrence was found after 1 year.

3 | DISCUSSION

Xanthogranulomatous inflammation is histopathologically characterized by a marked proliferative fibrosis, parenchymal destruction, and infiltration of foamy histiocytes intermixed with other inflammatory cells.[1,2] There have been only a few cases of XGI reported in the head and neck,[3-5] and periocular xanthogranuloma has also been reported.[6] This disease is very difficult to diagnose and is sometimes mistaken for malignant tumor.[7-9]

The precise pathogenesis of XGI is not well understood. The progression of this specific type of inflammation is presumed to be through benign cystic or cyst-like lesions, such as neuroenteric cysts,[9] pancreatic parenchymal cysts,[10] or Rathke's cleft cysts.[11]

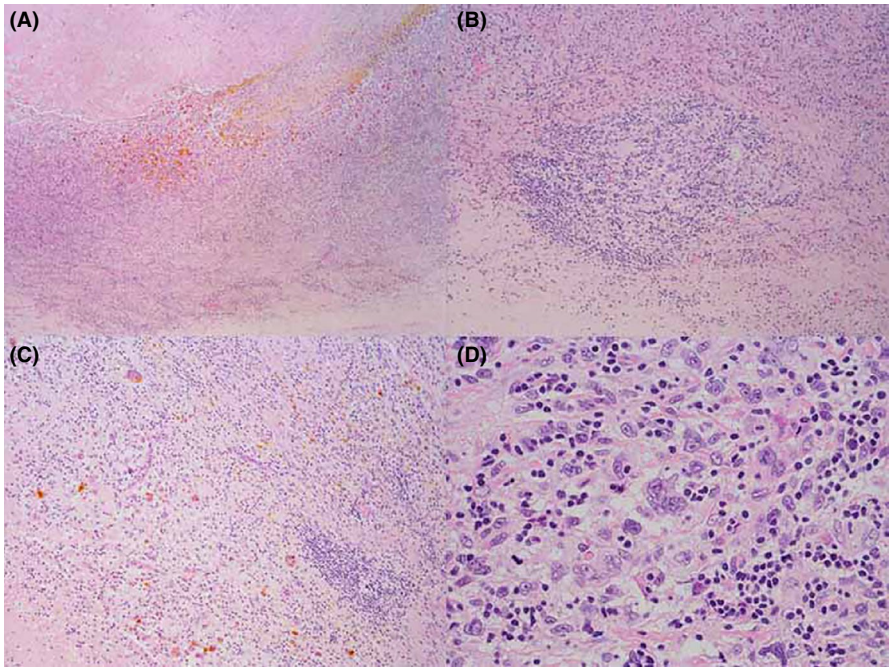

FIGURE 3 HE staining. A, Central necrotic area. B, Peripheral lymphoid nodule. C, Coagulative necrosis and hemosiderin deposits are seen. D, Lymphoma-like large nuclear cells are seen

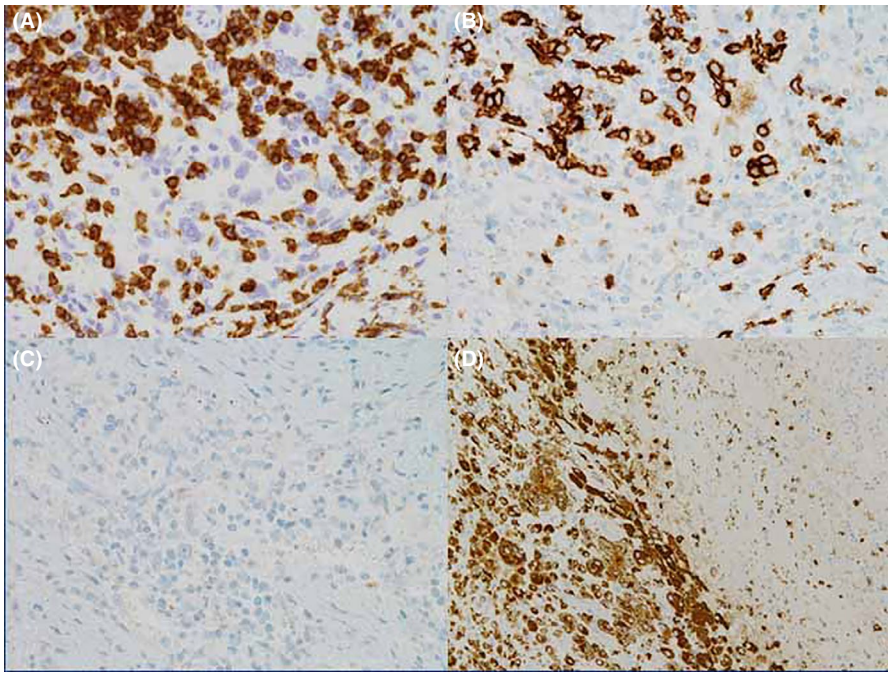

FIGURE 4 A, CD3-positive cells are seen around the tumor mass. B, CD20-positive cells are seen around the tumor mass. C, CD68 was negative for cells in the central area. D, CD68 was positive for cells outside the necrotic area

Several possible hypotheses have been proposed including defective lipid transport, immunological disorders, reaction to a specific infectious agent of low virulence, or lymphatic obstruction.[1,2] It is assumed that xanthogranulomatous cholecystitis is caused by the rupture of Rokitansky-Aschoff sinuses and mucosal ulceration.[1,2]

In the head and neck area, two cases were suspected to be of cystic origin, and the other case was a warthin tumor with XGI raised by fine-needle aspiration (FNA) biopsy.[5]

In our case, there was central necrosis, but there were no cystic findings. Based on the pathology, the cause of our lesion was not thought to be cystic, but instead lymph node in origin. We were, however, unable to conclusively determine the origin.

Immunohistochemical staining was the key to diagnosis since CD68 showed the monocyte/macrophage series and giant cells. Other epidermal tumor markers and lymphocyte markers are important for making a definitive diagnosis.

Histiocytes could be diagnosed only through HE staining, but it is very difficult to distinguish them from malignant cells. In these cases, CD68 is very useful for histiocytic identification. In our case, CT and PET scan showed the possibility of malignant tumor. Matsuoka et al[12] reported that low intensity in T1 and high intensity in T2 and diffusion is characteristic of TGI, but this is not the decisive factor. Open biopsy or extirpation is needed for final diagnosis.

In terms of treatment, FNA biopsy is useful in some cases, but a final diagnosis cannot always be made. In many cases, diagnosis has been made after surgery. If XGI is diagnosed beforehand, surgery can be avoided. We need to consider XCI for a neck mass mimicking malignant tumor.

CONFLICT OF INTEREST

None declared.

AUTHOR CONTRIBUTION

TN: treated this case and have full responsibility in this paper. MT: co-operated the case and cheeked the all literature. SK: is a chief of our Department and has a responsibility of all patients' outcome and paper publication. SM: is pathologist and helped us for histology part.

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