Branchial Cleft Cyst with Xanthogranulomatous Inflammation

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Abstract Xanthogranulomatous inflammation (XGI) is a special type of inflammation presenting as mass lesions and mimicking malignant tumors. It is rarely described in the head and neck region, and to the best of our knowledge, there are no cases associated with branchial cleft cyst. A 39-year-old woman with an infiltrative cystic neck mass that radiologically mimicks a malignant tumor is herein presented. The histopathologic diagnosis was a branchial cleft cyst with XGI. XGI may be associated with branchial cleft cysts. Patients may present with radiologic findings mimicking an invasive malignant tumor, and should be taken into account during the differential diagnosis with cystic squamous cell carcinoma.

Keywords Xanthogranulomatous inflammation · Branchial cleft cyst · Neck · Cystic squamous cell carcinoma · Differential diagnosis

Introduction

The most common cause of a cystic neck mass in patients under the age of 40 is a branchial cleft cyst (BCC) [1].

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The mechanism for the development of BCC is unclear. The most favoured hypothesis is that these cysts are caused by the failure to obliterate the cervical sinus from the second branchial cleft remnant [2, 3].

There are many case reports of carcinomas arising from BCC, the so-called “branchiogenic carcinoma”. However their existence is controversial since these might be mimicked by metastatic cystic carcinomas, which are frequent lesions of the lateral neck in adults [1–4]. A malignant tumor, especially metastatic cystic squamous cell carcinoma, should always be considered in the differential diagnosis of a cystic lateral neck mass, particularly in patients older than 40 [1, 2].

XGI is a mass-forming lesion, sometimes presenting with clinical and radiologic findings mimicking malignant tumors. It is recognised by foamy histiocytes, Touton type giant cells and granulomas. XGI is diagnosed by histopathologic examination and generally the histopathologic features of the lesion are not mistaken for an epithelial malignancy at different anatomic sites. Xanthogranulomatous pyelonephritis and cholecystitis are the most recognized locations of this special inflammation [5–7]. To the best of our knowledge, this type of inflammation is very rarely described in the head and neck region and association of XGI and BCC was not described previously. Herein, such a case is presented, as this lesion may cause confusion in the clinical distinction from other malignant entities and, for the pathologist, XGI at such a setting should be kept in mind.

Case Presentation

A 39-year-old woman presented with a mass lesion in the lateral neck. She had a history of a similar mass in this

Fig. 1 Fine needle aspiration cytology from the mass. Squamous cells with atypical features and inflammatory cells (Papanicolaou stain, original magnification X40)

region 3 years prior. At that time, sonographic examination revealed a 30 mm mass and the fine needle aspiration diagnosis was an epidermal cyst with abscess formation. Following treatment with antibiotics, she did not have any other complaints until 1 year ago. Otherwise her personal history was unremarkable.

Head and neck examination, including upper aerodigestive tract endoscopy, conducted after her recent presentation was unremarkable, except for the 30 × 20 mm firm, semi-mobile mass in the anterior sternocleidomastoid muscle (SCM) inferior to the angulus mandibula. The recent fine needle aspiration revealed squamous cells with mild atypia and inflammatory cells (Fig. 1).

Computerized Tomography (CT) examination revealed a 31 × 20 × 33 mm cystic mass postero-inferior to the right mandibular angle. The mass was medial to the carotid

sheath, dorsal to the submandibular gland, and ventro-medial to the SCM. The wall thickness was variable, with the thickest and thinnest portions measuring 5 and 1 mm, respectively. Furthermore, the cyst had a solid component that occupied its inferior anterior region. The fat plane between the mass and the SCM was effaced, suggesting muscle invasion (Fig. 2a and b).

The mass, which was partially adherent to SCM, was excised under general anesthesia.

Macroscopic examination revealed a 40 × 35 × 15 mm soft, partially cystic lesion. The cut surface revealed a 25 × 15 × 10 mm cystic mass filled with purulent material; adjacent to the cyst, a 20 × 10 × 10 mm yellow to grey-white mass with irregular contours, suspicious for an invasive lesion, was observed. (Fig. 3).

Microscopic examination revealed a cyst lined by squamous epithelium with reactive changes and surrounded by chronic inflammation and reactive lymphoid tissue. In the sections of the yellow mass lesion and cyst, adjacent to the previous cyst, a diffuse infiltration of histiocytes with Touton type giant cells and granuloma formation were observed. The inflammatory lesion extended into a neighbouring lymph node and striated muscle. The cells of the infiltrate were CD68 positive; and S100 and CD1a negative (Fig. 4a–d). Ziehl Neelsen and periodic-acid Schiff (PAS) stains were negative for any specific microorganisms.

The final diagnosis was BCC with XGI. No further treatment was administered.

Discussion

XGI is a mass forming lesion, sometimes mimicking malignant tumors. Histologically, it is characterized by

Fig. 2 **a** Contrast-enhanced CT image through the mandible shows the cystic mass (*) posterior to the mandible (m), medial to the sternocleidomastoid muscle (SCM), and lateral to the carotid artery (arrow) and jugular vein (arrow head). The fat plane between the mass and SCM is effaced, suggesting muscle invasion. Note the normal fat plane (hollow arrow) medial to the contralateral sternocleidomastoid muscle. **b** Coronal reformatted view shows the mass medial to the sternocleidomastoid muscle (SCM) and lateral to the jugular vein (arrow)

Fig. 3 Yellow mass adjacent to cystic lesion

foamy histiocytes, Touton type giant cells and granulomas. The macroscopic appearance typically shows a yellow, granular surface with irregular contours. Infiltration into the adjacent tissues and organs is a frequent finding of this specific type of inflammation, which may be identified in many regions, such as pancreas, testis, kidney, and gallbladder [5–11].

There are few cases of XGI in the head and neck including periorbital region, Rathke cleft cyst and neuroenteric cyst at the cerebellopontine angle, presenting with pathologically analogue lesions [12–14].

The progression of this specific type of inflammation is related to infection and obstruction. The association with benign cystic or cyst-like lesions is described, such as neuroenteric cyst, colloid, epidermoid, renal, and pancreatic paranchymal cysts as well as cholesterol granuloma [14].

Histologically, BCC are lined by squamous or respiratory epithelium, frequently associated with subepithelial lymphocytes [4]. Fibroinflammatory reaction surrounding BCC is a frequent finding, especially if the cyst is ruptured or infected [2]. However, extensive infiltration by foamy histiocytes, Touton type giant cells and granulomas are characteristics of XGI, which are consistent with our findings in the case presented. Considering that our patient had a cystic lesion and infection, an association with XGI is, therefore, not unexpected, although, it has not been described previously.

The most important clinical differential diagnosis for our case included BCC and metastatic cystic squamous cell carcinoma; although not always the case, the latter is more frequently observed in an older age group compared with the present patient [1–4].

The radiologic examination of a typical branchial cleft cyst shows attenuation values suggesting mucoid content (10–25 Hounsfield Units) with a uniformly thin, smooth wall. Due to infection, the cyst wall is irregularly thickened and shows contrast enhancement [15]. An infected cyst with such findings may resemble a suppurative or metastatic lymph node. The findings, especially the solid component in our case, suggested that a malignant process was more likely rather than an infected branchial cyst.

Fig. 4 **a** Cystic lesion lined by stratified squamous epithelium (*lower part*) and adjacent to muscle tissue infiltrated with diffuse histiocytes (*upper right*) (H&E, original magnification x4). **b** Diffuse histiocytic infiltration extending to a follicular hyperplastic lymph node (H&E, original magnification x4). **c** Granulomata formation with Touton type giant cells (H&E, original magnification x20). **d** Diffuse CD68 positive histiocytic cells (antiCD68, original magnification x40)

The macroscopic examination of the surgical specimen was suggestive of an infiltrative lesion and the yellow color at the solid region was a clue for XGI. The microscopic findings were typical for BCC and XGI.

The histopathological diagnosis of XGI associated with BCC, suggests that this diagnosis should be considered in the differential diagnosis of complex neck cysts.

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