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INVITED REVIEW

CT and MR imaging findings of non-neoplastic cystic lesions of the parotid gland

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Abstract
A variety of neoplastic and non-neoplastic lesions of the parotid gland can present with a predominantly cystic architecture, and although radiologists frequently encounter cystic parotid tumors, other non-neoplastic lesions should also be included in the differential diagnoses of cystic parotid lesions. Non-neoplastic cystic lesions are usually classified as either congenital/acquired cystic lesions or inflammatory/infectious lesions. Adequate knowledge about these rare conditions is essential for appropriate diagnosis and optimal treatment strategy. This review article describes CT and MR imaging features of non-neoplastic cystic lesions of the parotid gland and provides helpful suggestions on the differential diagnoses for cystic parotid lesions.

Keywords Non-neoplastic · Cystic lesion · Parotid gland · CT · MRI

Introduction

CT and MR imaging can identify intralesional cystic components in 50% of benign parotid tumors and in 79% of malignant parotid tumors [1]; therefore, cystic degenerations are commonly observed imaging features in parotid gland tumors. In contrast, non-neoplastic cystic lesions in the parotid gland are relatively rare compared to cystic parotid tumors [2]. In this article, non-neoplastic cystic lesions of the parotid gland are described and classified into two categories, namely, congenital/acquired cystic lesions or inflammatory/infectious diseases. Congenital/acquired cystic lesions include first branchial cleft cyst (FBCC), lymphatic malformation (LM), benign lymphoepithelial lesion (BLEL) [sporadic/simple lymphoepithelial cyst (LEC), human immunodeficiency virus (HIV)-associated BLEL, and BLEL of Sjögren's syndrome], salivary duct cyst (SDC), pseudocyst (retention cyst, mucocele, and sialocele), and epidermoid/dermoid cyst, while inflammatory/infectious diseases include sialodochitis fibrinosa, hydatid cyst, intraglandular lymphadenitis, and abscess (Table 1). Correct and immediate diagnosis of these rare conditions will help in choosing an appropriate treatment strategy and can improve prognosis. Although a review article about neoplastic and non-neoplastic lesions of the parotid gland have been published, we found no review articles with emphasis on imaging features of non-neoplastic parotid lesions [2]. Hence, the aim of this review article is to describe CT and MR imaging features of common and uncommon non-neoplastic cystic lesions of the parotid gland and to provide helpful suggestions on the differential diagnoses for cystic parotid lesions.

Congenital/acquired cystic lesion

First branchial cleft cyst (FBCC)

FBCCs account for 5–8% of all brachial cleft defects and are most commonly seen in middle-aged women with characteristic clinical manifestations of swelling in the preauricular, parotid, or cervical region [3]. FBCCs can typically present as cysts, sinus, or fistulae at various locations, including between the submandibular region and the external auditory canal along the the first branchial arch tract, as extending from a cutaneous opening in the submandibular triangle, at a location superolateral to the hyoid bone, or as ascending to the region of the parotid gland that terminates

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Table 1 List of non-neoplastic cystic lesions of the parotid gland

Congenital/acquired cystic lesions
First branchial cleft cyst
Lymphatic malformation
BLEL
Sporadic/simple lymphoepithelial cyst
HIV-associated BLEL
BLEL of Sjögren's syndrome
Salivary duct cyst
Pseudocyst (retention cyst, mucocele, and sialocele)
Epidermoid/dermoid cyst
Inflammatory/infectious diseases
Sialodochitis fibrinosa
Hydatid cyst
Intraglandular lymphadenitis
Abscess

BLEL benign lymphoepithelial lesion, *HIV* human immunodeficiency virus

at the cartilaginous/bony junction of the external auditory canal [4]. FBCCs can be classified into two groups based on presumed pathogenesis as Type I or Type II; however, this classification has limited applicability from an imaging perspective [4]. Type I FBCCs usually present as cystic masses adjacent to the external auditory canal and are generally considered as a duplication anomaly of the membranous external auditory canal. Further, they are purely ectodermal in origin; hence, they are lined by stratified squamous epithelium. In contrast, Type II FBCCs may present as cysts, sinuses, or fistulae in the region of the mandibular angle and may communicate with the external auditory canal. They are ectodermal or mesodermal in origin, and hence, contain squamous epithelium, skin adnexa, and cartilage.

FBCCs on CT and MR imaging usually appear as well-defined, thin-walled, unilocular or multilocular, oval or round cystic masses that may be located superficial to, within, or deeper than the parotid gland [5]. As with other branchial cleft cysts, cyst wall thickness and contrast-enhancement are variable as they are proportional to the degree of inflammation with superimposed infection leading to increased CT attenuation of fluid content (Fig. 1). Fistulas and sinuses are not always obvious on CT and MR imaging.

Lymphatic malformation (LM)

Simple vascular malformations are classified into two categories based on their flow characteristics as, slow-flow (capillary malformation, venous malformation, LM) and fast-flow (arteriovenous malformation). LMs can be dilated lymphatic channels or cysts that are lined by endothelial cells with a lymphatic phenotype. LMs are classified according to cyst size as microcystic, macrocystic, or mixed subtypes.

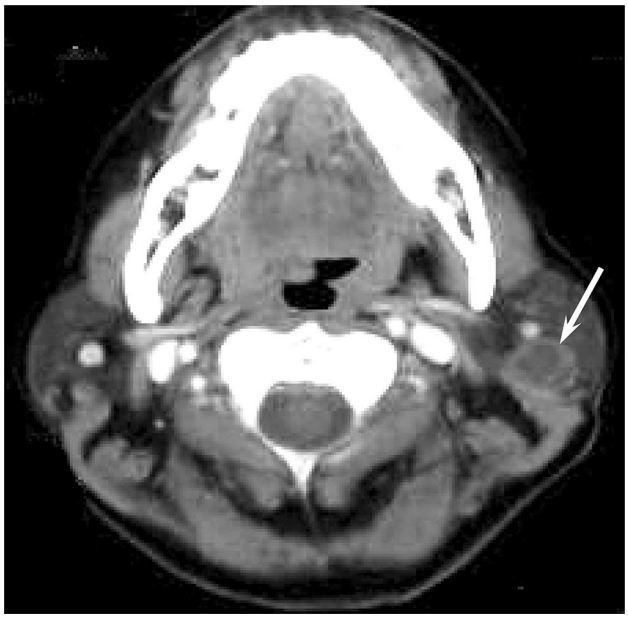

Fig. 1 A 54-year-old woman with first branchial cleft cyst. Contrast-enhanced CT image shows a thick-walled unilocular cystic lesion (arrow) in the left parotid gland

Parotid LMs occur frequently in children younger than 2 years of age and are rare in adults. Patients usually present with painless soft swelling and often experience symptoms for long durations. Complications associated with parotid LMs include rupture, infection, and nerve compression secondary to hemorrhage within the cyst; however, malignant transformation has never been reported. Parotid LMs can evolve in one of three ways: (1) spontaneous regression (2) slow progression, and (3) rapid enlargement due to hemorrhage or infection.

On CT imaging, uncomplicated LMs typically exhibit homogeneous fluid attenuation, whereas infected or hemorrhagic LMs exhibit higher attenuation. On MR imaging, the typical signal patterns of LM are low to intermediate intensity on T1-weighted images and hyperintensity on T2-weighted images. However, hyperintensity on T1-weighted images can be infrequently observed due to the presence of clotted blood or high lipid (chyle) content [6] (Fig. 2). In cases with hemorrhage, fluid–fluid formation may be observed. Compared to CT images, MR images can clearly demonstrate internal septa.

Benign lymphoepithelial lesion (BLEL)

Sporadic/simple lymphoepithelial cyst (LEC)

Sporadic/simple LECs of the parotid gland have no underlying disease and are not associated with HIV infection or Sjögren's syndrome. They usually occur in middle-aged men

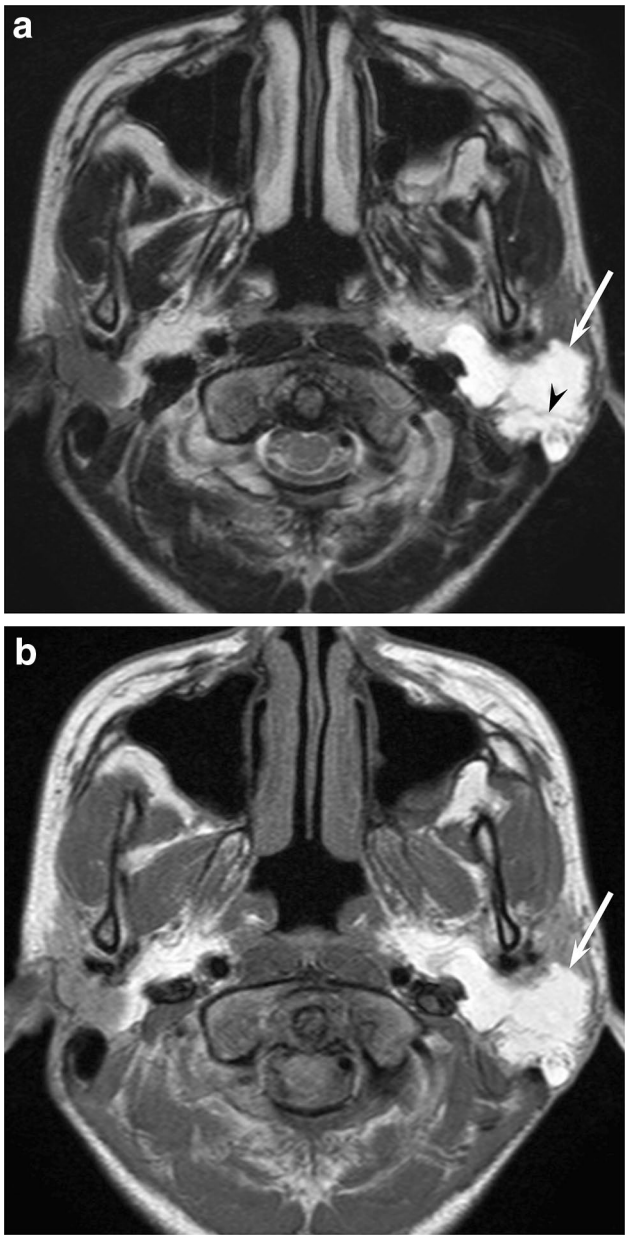

Fig. 2 A 12-year-old girl with lymphatic malformation. **a** T2-weighted image shows a lobulated cystic lesion (arrow) with internal septum (arrow head) in the left parotid gland. **b** T1-weighted image shows a hyperintense cystic lesion (arrow) due to hemorrhage

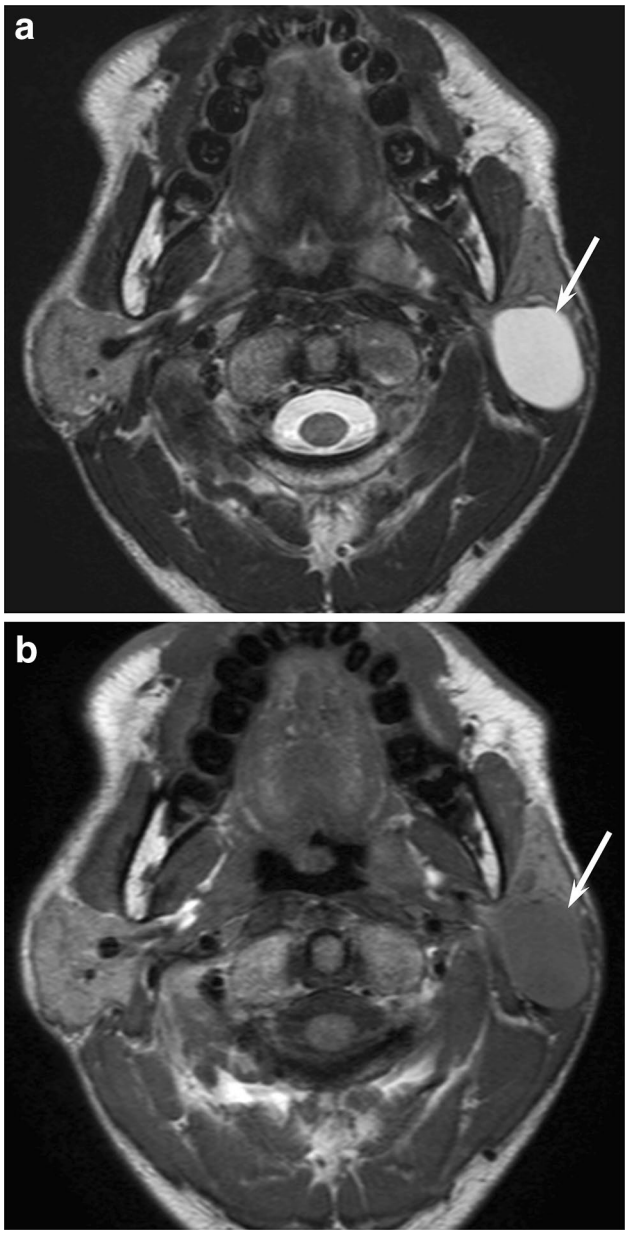

Fig. 3 A 38-year-old man with sporadic/simple lymphoepithelial cyst. **a** T2-weighted image shows a thin-walled unilocular cystic lesion (arrow) in the left parotid gland. **b** T1-weighted image shows a slightly hyperintense cystic lesion (arrow) compared with the muscles

and manifest as slowly enlarging painless swellings that affect a single gland with normal movable overlying skin [7]. LECs are typically unilateral, solitary, well-circumscribed, and predominantly unilocular cysts. The parotid gland is the preferred site for LECs due to the presence of intraparotid lymph nodes. Microscopically, typical lesions are lined by stratified squamous epithelium surrounded by abundant lymphoid tissue with prominent germinal centers [7].

On CT and MR images, sporadic/simple LECs are usually visible as thin-walled, unilateral, solitary, unilocular cysts (Fig. 3). Intracystic attenuation or signal intensity are due to viscosity or protein concentration of the fluid content, and cyst wall thickening and enhancement can be observed in infected lesions.

Human immunodeficiency virus (HIV)-associated BLEL

HIV-associated BLELs of the parotid gland usually present with painless, unilateral or bilateral enlargements of the parotid glands with or without xerostomia. Parotid gland

enlargement is sometimes the initial clinical sign of HIV infection as BLELs develop as an early manifestation of HIV infection before AIDS, with an incidence rate of 3% [8]. HIV-associated BLELs are histologically typified by lymphoid hyperplasia in combination with an epithelial component, along with cystic and proliferative changes, and formation of lymphoepithelial islands [8].

On CT and MR imaging, numerous entirely cystic, combined cystic and solid, or entirely solid lesions, which can be either bilateral or unilateral, are observed in the parotid gland (Fig. 4). Characteristic imaging features of HIV infection such as non-necrotic reactive cervical lymphadenopathy and hypertrophy of lymphoid tissue in the palatine, lingual, or adenoid regions help in differentiating between HIV-associated BLEL and BLEBs of Sjögren's syndrome [9, 10].

BLEL of Sjögren's syndrome

BLEL, also referred to as lymphoepithelial sialadenitis (LESA), lymphoepithelial lesion, myoepithelial sialadenitis (MESA), Sjögren's-type sialadenitis, or autoimmune sialadenitis of the salivary glands, is histologically characterized by extensive infiltration of chronic inflammatory cells into the salivary parenchymal space, which then replace the native acinar and ductal cells [11]. Sjögren's syndrome occurs predominantly in the fourth and fifth decades of life with a female: male ratio of 9:1. Clinical manifestations include keratoconjunctivitis sicca, arthritis, dry mouth, and parotid enlargement. The parotid glands are affected in about 90% of the cases and parotid enlargement is usually bilateral. The incidence of lymphoma has been reported to be 44 times higher in patients with Sjögren's syndrome than in the general population.

Imaging features of the parotid gland in Sjögren's syndrome depend on the stage of the disease. In the early stages, the parotid glands may appear normal or diffusely enlarged with a normal parenchymal pattern, while in the intermediate stage, bilateral, diffusely enlarged, parotid glands show multiple, scattered, small cysts and solid masses [9]. In the late stages, parenchymal heterogeneity, abnormal diffuse fat tissue deposition, diffuse punctate calcification, and large cystic and solid masses are observed bilaterally [12] (Fig. 5). These cystic components represent areas of destroyed gland or collections of saliva, while the solid masses represent lymphoid aggregates that actively destroy the gland.

Salivary duct cyst (SDC)

SDCs are true cysts and are also referred to as mucus retention cyst, mucus duct cyst, sialocyst, or simple cysts that can either be congenital or acquired. SDCs represent 10% of all salivary gland cysts and frequently occur in individuals aged between 30 and 40 years without gender predilection [13]. They are unilateral, painless, compressible growths that range in size from 0.8 to 10 cm (predominantly 1–3 cm)

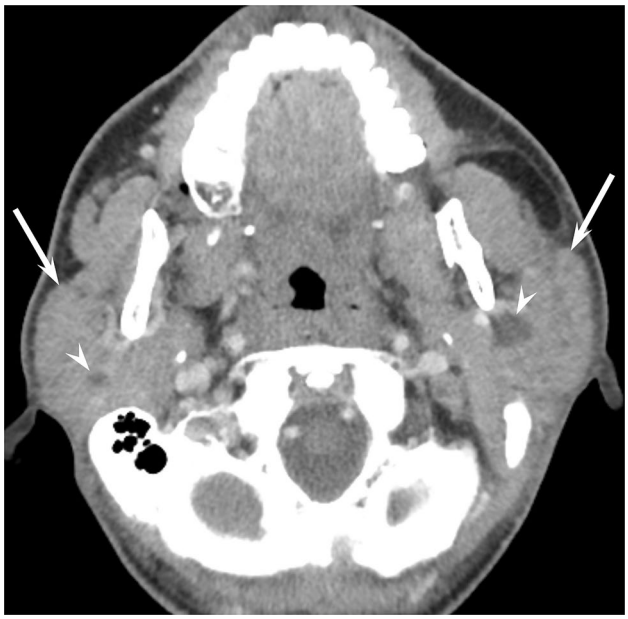

Fig. 4 A 48-year-old man with human immunodeficiency virus-associated benign lymphoepithelial lesion. Contrast-enhanced CT image shows cystic lesions (arrow heads) within the bilateral enlarged parotid glands (arrows)

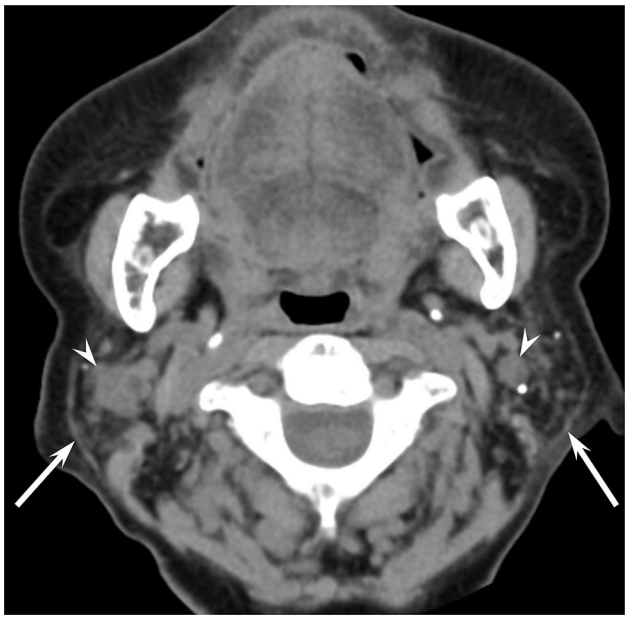

Fig. 5 A 73-year-old woman with benign lymphoepithelial lesion of Sjögren's syndrome. Unenhanced CT image shows combined solid and cystic lesions (arrow heads) within the bilateral atrophied parotid glands (arrows)

[13]. The common sites of SDCs are the minor oral salivary glands; they rarely occur in the major salivary glands but show a marked predilection for the superficial lobe of the parotid gland [13]. The cyst wall of SDCs are lined with ductal epithelium that may have undergone squamous or oncocytic metaplasia; therefore, SDCs are classified as true cysts based on histopathologic features [14]. Possible causes of SDCs include obstruction of the salivary duct due to calculi, mucus plugs, postoperative damage, or post-inflammatory strictures [14].

On CT and MR images, SDCs are usually visible as unilateral, solitary, well-defined, thin-walled, unilocular cysts (Fig. 6). Fluid–fluid formation and mural nodules were reported as uncommon MR findings [13].

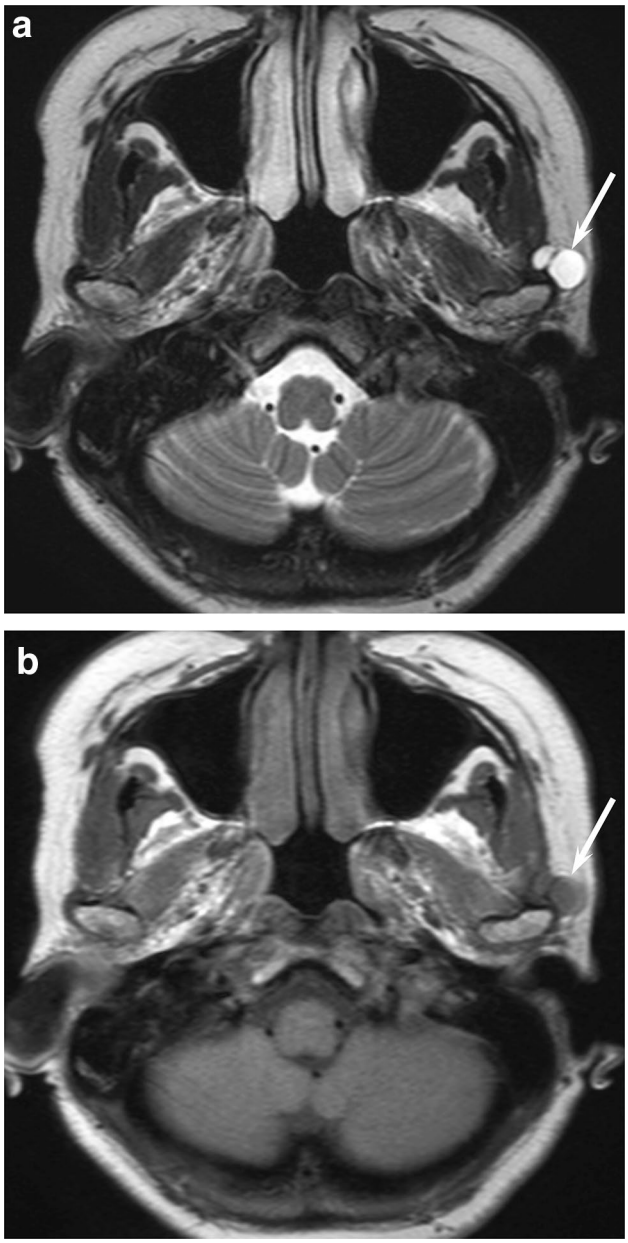

Fig. 6 A 54-year-old woman with salivary duct cyst. **a** T2-weighted image shows a thin-walled bilocular cystic lesion (arrow) in the left parotid gland. **b** T1-weighted image shows a hypointense cystic lesion (arrow)

Pseudocyst (retention cyst, mucocele, sialocele)

Pseudocysts of the parotid gland, such as retention cyst, mucocele, and sialocele, are devoid of a distinct epithelial lining and are secondary to causes such as obstruction, stenosis, or laceration of the salivary duct. Retention cysts are typically present as ranulae arising from the sublingual gland as a result of ductal obstruction and fluid retention. Mucoceles most frequently occur in the lower lip and the most common cause is trauma or the habit of lip biting [15]. Therefore, retention cysts and mucoceles of the parotid gland are extremely rare, while congenital and other acquired cysts, including sialocele, commonly occur in the parotid gland. A sialocele is an accumulation of extravasated saliva into the glandular or the peri-glandular tissues and develops secondary to excretory duct blockage due to complications after facial trauma or surgery.

CT and MR images demonstrate well-defined unilocular or multilocular cystic lesions that are typically filled with fluids that exhibit homogenous water attenuation or signal intensity. Further, CT attenuation and MR signal intensity of fluid content vary according to mucin or protein concentration (Fig. 7), and MR sialography is a non-invasive method for investigating iatrogenic sialocele to assess the ductal system, site of ductal injury, and focal cystic lesions [16].

Epidermoid/dermoid cyst

Epidermoid cysts are composed of epidermal-type squamous epithelium without any adnexal structures derived from the lining epithelium, whereas dermoid cyst have identical squamous epithelium with pilosebaceous units and sweat glands [17]. Although the head and neck region is a common site of occurrence for these cysts, their localization in the parotid gland is an extremely rare occurrence. Patients usually present with a slow-growing, mobile, non-tender, painless mass with or without fluctuation that requires surgical intervention and these cysts are common in young to middle-aged adults.

Epidermoid cysts typically exhibit well-defined cystic masses that are almost isoattenuating compared to cerebrospinal fluid on CT, hypointense on T1-weighted images, and hyperintense on T2-weighted images [6]. On diffusion-weighted images (DWI), epidermoid cysts show marked diffusion restriction due to the presence of white

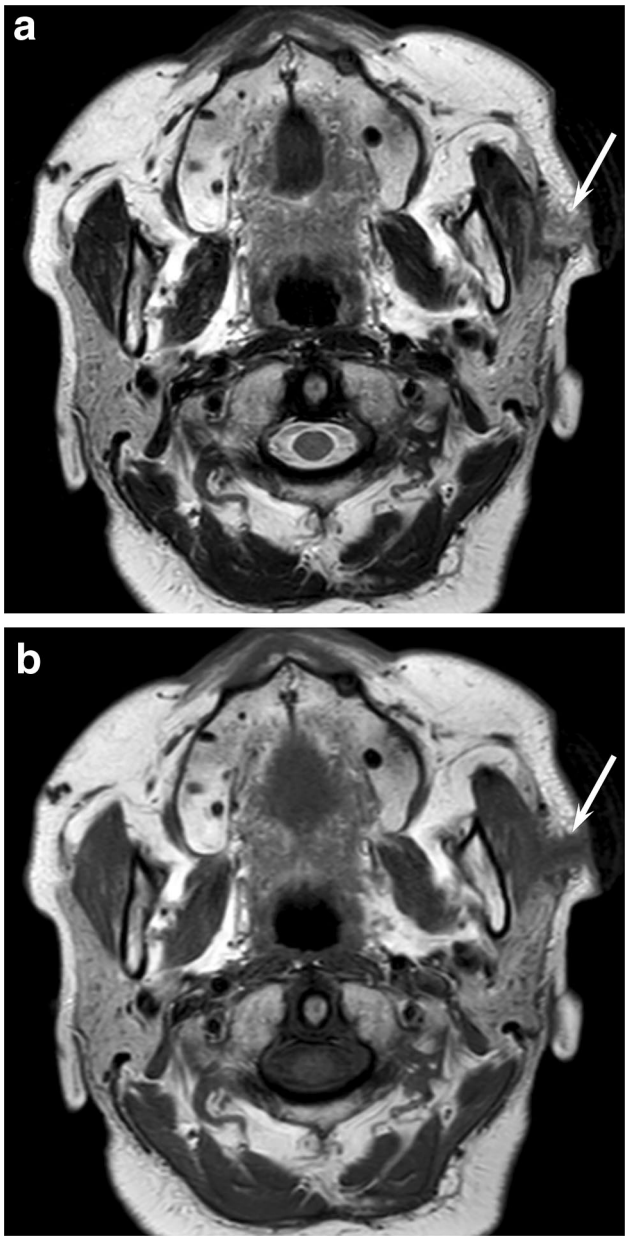

Fig. 7 A 69-year-old woman with sialocele occurring after surgery for removal of pleomorphic adenoma of the left parotid gland. **a** T2-weighted image shows fluid collection (arrow) corresponding to the site of operation. **b** T1-weighted image shows hypointense fluid collection (arrow)

waxy material rich in cholesterol crystals mixed with cellular debris derived from the progressive desquamation and breakdown of keratin from the epithelial lining (Fig. 8). On the other hand, dermoid cysts typically appear as well-defined, fat-containing cystic masses. Fat suppression or phase-shift MR sequences are useful for detecting fatty tissue and dermoid cysts with a small amount fatty tissue also show diffusion restriction on DWI.

Inflammatory/infectious disease

Sialodochitis fibrinosa

Sialodochitis fibrinosa is a rare disease characterized by recurring episodes of pain and swelling of the parotid or submandibular glands due to formation of mucofibrinous plugs [18]. The most accepted etiology is an allergic process related to the consumption of certain foods, namely tuna fish, wheat, lettuce, and legumes such as lima beans [18]. Patients with a medical history of allergic processes typically present with elevated serum eosinophils and immunoglobulin E (IgE) levels, and approximately two-thirds of all patients are female with a median age of 47 years [19]. Further, the parotid glands were more frequently involved than the submandibular glands and bilateral affliction was more common, as only approximately one-third of all cases were unilateral [19].

On CT and MR imaging, cylindrical dilation of the main drainage duct with or without multiple cystic dilations of the intraglandular ducts in the parotid and/or submandibular glands are the characteristic imaging features [18] (Fig. 9).

Hydatid cyst

Hydatid disease is a parasitosis that is also known as hydatidosis or echinococcosis commonly caused by the larval stage of the *Echinococcus granulosus*. *Echinococcus multilocularis* is less common but more invasive and mimics a malignancy. Hydatid cysts commonly involve the liver (70%) and lungs (25%), and primary hydatid cyst of the parotid gland is extremely uncommon. Although most hydatid cysts are slow-growing and asymptomatic, its mass effect may lead to manifestation of clinical symptoms. As spontaneous rupture of the cyst or rupture due to trauma or surgery can cause anaphylactic reactions and life-threatening conditions, preoperative diagnosis is essential [20].

Based on imaging appearance, hydatid cysts are classified into four types: Type I, simple cyst with no internal architecture; Type II, cyst with daughter cyst(s) and/or matrix; Type III, calcified cyst; and Type IV, complicated hydatid cyst [21]. According to previously reported cases, unilocular, well-defined cystic lesions with no internal architecture (Type I) may be the most common imaging feature of parotid hydatid cysts [22, 23].

Intraglandular lymphadenitis

The parotid gland is different from other salivary glands in that the lymph nodes are enclosed within its parenchyma and these lymph nodes are frequently the sentinel nodes

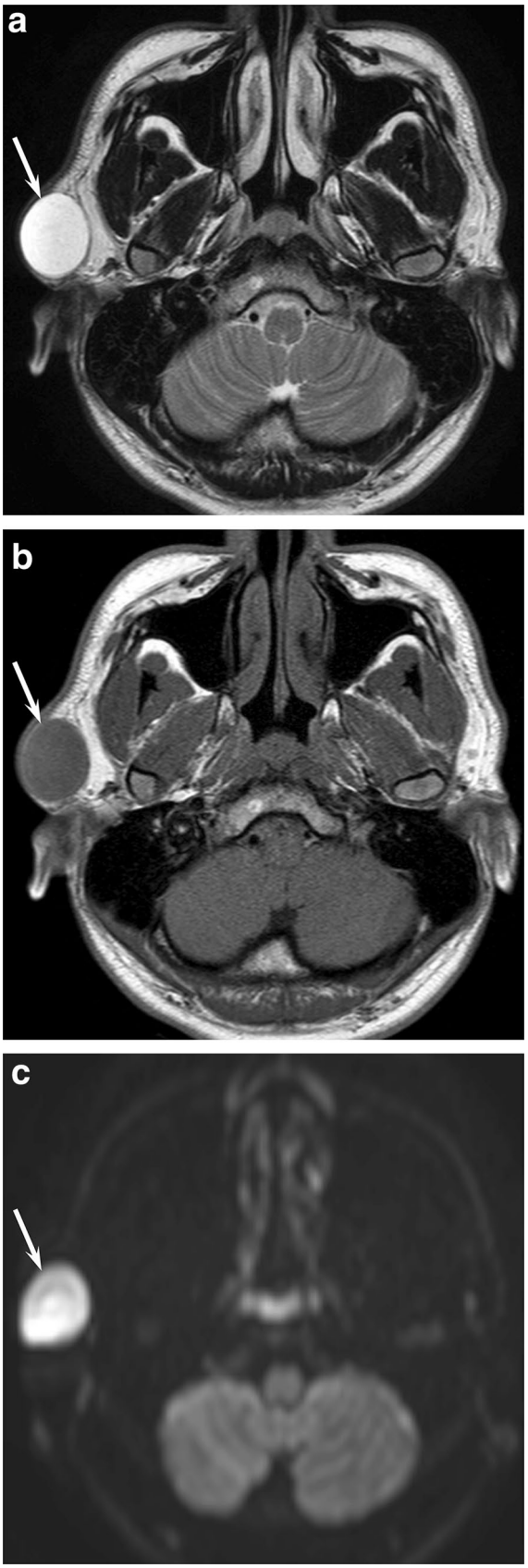

Fig. 8 A 25-year-old man with extraparotid subcutaneous epidermoid cyst. **a** T2-weighted image shows a subcutaneous unilocular cystic lesion (arrow), which compresses the right parotid gland medially. **b** T1-weighted image shows a slightly hyperintense cystic lesion (arrow) compared with the muscles. **c** Diffusion-weighted image shows a markedly hyperintense cystic lesion (arrow)

for the region encompassing the skin of the ear, cheeks, temple, forehead, and anterior scalp. Thus, cutaneous malignancies or infection of these areas can easily spread to the parotid lymph nodes. Even though various pathogens cause parotid lymphadenitis, its presence with cystic necrosis is commonly caused by bacterial or mycobacterial infection. Parotitis caused by non-tuberculous mycobacteria usually affects young children, whereas patients suffering from parotitis caused by *Mycobacterium tuberculosis* tend to be predominantly adults [24].

In suppurative lymphadenitis, central necrosis with thick, irregular, enhancing walls can be usually observed in the advanced stage (Fig. 10), similarly in mycobacterial lymphadenitis. Nonetheless, inflammatory changes surrounding the parotid lymph nodes are generally more prominent in suppurative lymphadenitis than in mycobacterial lymphadenitis. On DWI, central necrosis in suppurative or mycobacterial lymphadenitis usually shows marked diffusion restriction due to the presence of purulent fluid [25].

Abscess

Parotid abscess is rare disease in children and adults, and is most commonly observed in immunocompromized, dehydrated, or postoperative patients [26]. Parotid abscesses can arise from infection ascending via the Stensen duct or may be secondary to bacteremia. The most common pathogen associated with parotid abscess is *Staphylococcus aureus* [26] and the symptoms include spontaneous onset of warm, indurated, erythematous swelling of the angle of the jaw, pain due to stretching of the dense parotid capsule, erythema of the preauricular area associated with marked fever, and leukocytosis. Importantly, the infection can spread through the fascia and progress inferiorly to the deep neck spaces.

CT and MR images demonstrate multi-loculated, central necrotic components with contrast enhancement of the thick and irregular abscess wall. Inflammatory changes surrounding the abscess cavity on CT or fat-suppressed T2-weighted imaging are particularly prominent (Fig. 11). On DWI, marked diffusion restriction can be clearly seen due to the presence of viscous fluid containing inflammatory cells, bacteria, mucoid proteins, and cell debris.

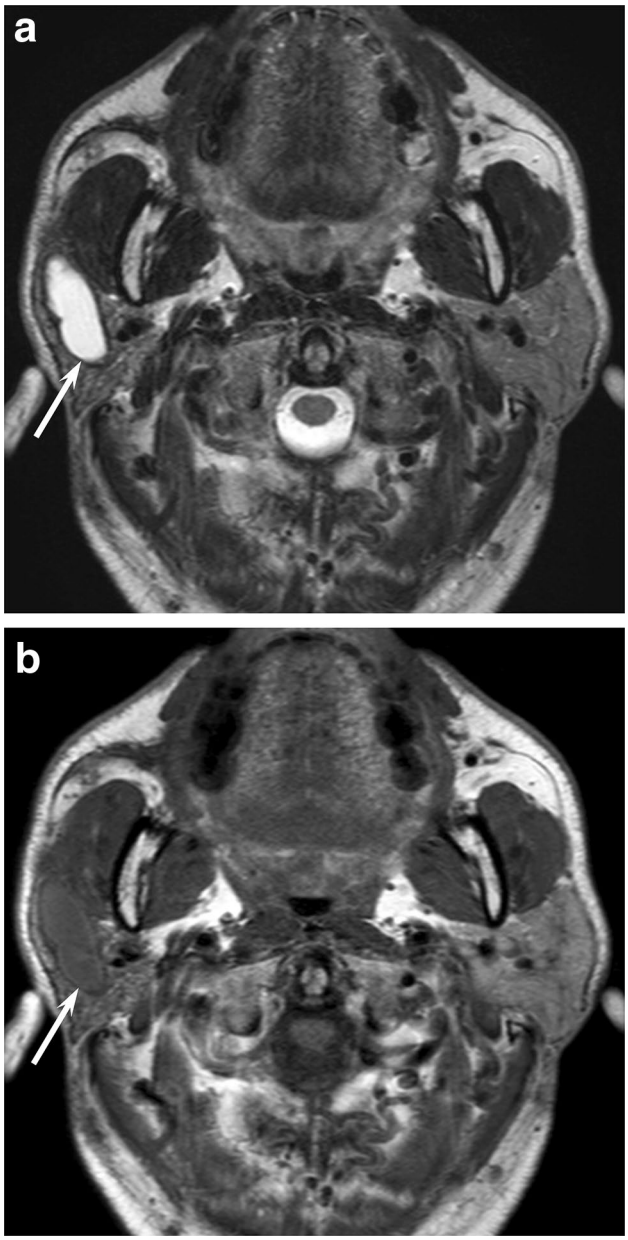

Fig. 9 A 55-year-old man with sialodochitis fibrinosa. **a** T2-weighted image shows an elongated cystic lesion (arrow) in the right parotid gland. **b** T1-weighted image shows a slightly hyperintense cystic lesion (arrow) compared with the muscles

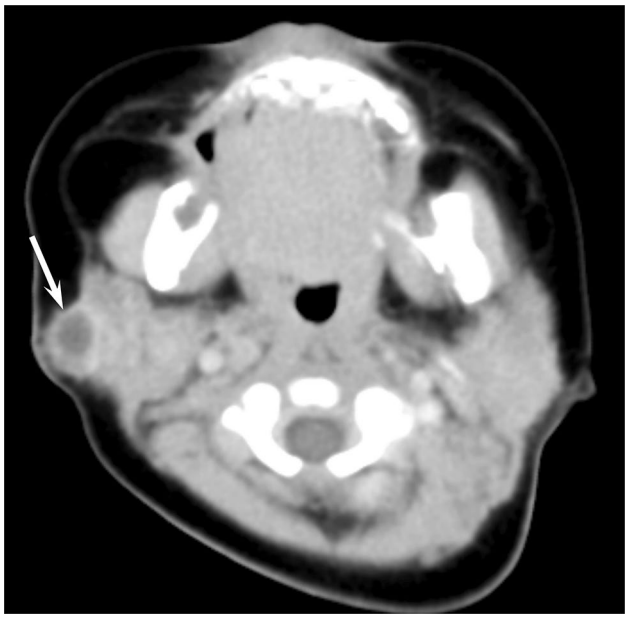

Fig. 10 A 6-week-old girl with intraglandular lymphadenitis. Contrast-enhanced CT image shows a thick-walled unilocular cystic lesion (arrow) with ring-like enhancement in the right parotid gland

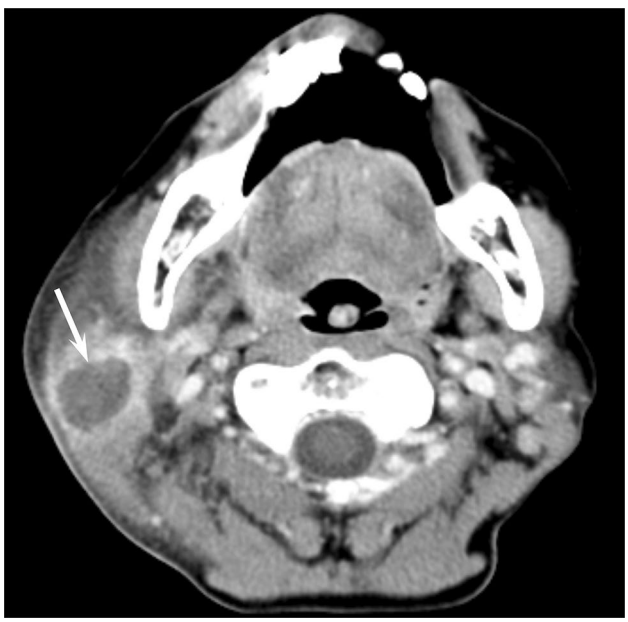

Fig. 11 A 64-year-old woman with abscess of the parotid gland. Contrast-enhanced CT image shows a cystic lesion (arrow) with enhancement of surrounding soft tissue in the right parotid gland

Conclusions

Although a majority of non-neoplastic cystic lesions of the parotid gland can be conservatively managed, life-threatening emergency, such as intraglandular lymphadenitis and abscess, need to be considered as differential diagnoses. As parotid lesions can often be the initial presentation of an underlying condition such as HIV infection, Sjögren's syndrome, and allergic disease, radiologists should be aware of their imaging characteristics. The differential diagnoses of thin-walled unilocular parotid cysts include FBCC, LM, sporadic/simple LEC, SDC, pseudocyst, epidermoid/dermoid cyst, or hydatid cyst.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical standards The authors declare that they preserve ethical standards.

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