Case Report

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Sonographic and CT Findings of Sialadenosis in a Child with Leukemia

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Sialadenosis is characterized by asymptomatic bilateral enlargement of the parotid glands. It is uncommon, especially in children. Diagnosis and analysis of sialadenosis based on imaging modalities have been rarely reported. Here, we report a case of sialadenosis in a child with leukemia, in which the diagnosis was made based on sonography and CT findings.
Index terms: *Sialadenosis; Sialosis; Sonography; Computed tomography; Children*

INTRODUCTION

Sialadenosis or sialosis is an uncommon disease of the salivary glands, characterized by asymptomatic, bilateral, diffuse swelling of the major salivary glands, particularly the parotid gland (1, 2). This condition usually does not affect salivary gland function. Since systemic diseases or conditions such as diabetes mellitus and alcoholism related to the development of sialadenosis are rare in children (1), almost all cases with sialadenosis have been reported in adults. Aside from the rare incidence of this disease, the imaging findings of sialadenosis are even further seldom described. We present a case of sialadenosis with sonographic (US) and CT appearances.

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CASE REPORT

A 7-year-old boy with acute lymphoblastic leukemia who had been treated with multiple cycles of chemotherapy consisting of vincristine sulfate, cytosine arabinoside, methotrexate, daunorubicin, and 6-mercaptopurine, presented with bilateral infra-auricular painless swelling that developed gradually after the administration of a steroid (dexamethasone). Laboratory findings showed mild pancytopenia, slightly hypocellular bone marrow with complete remission of acute lymphoblastic leukemia, no evidence of leukemic blast on CSF cytospin, and slightly increased blood levels of C-reactive protein, amylase, and lipase. No infectious organisms were documented by blood tests or culture. Upon physical examination, the swelling was found to be symmetric, hard, non-movable, not tender, and with no heat emitting from it. It was measured to be approximately 4 cm in diameter. The skin over the swelling was normal. The neck US performed with 12-5 MHz broadband linear and 5-2 MHz broadband convex transducers (iU22; Philips Healthcare, Andover, MA, USA) showed diffuse enlargement of both parotid glands without a focal lesion or increased blood flow (Fig. 1A, B). Because of posterior acoustic attenuation in the enlarged parotid gland with the linear probe, the convex probe was added to the US examination in order to assess

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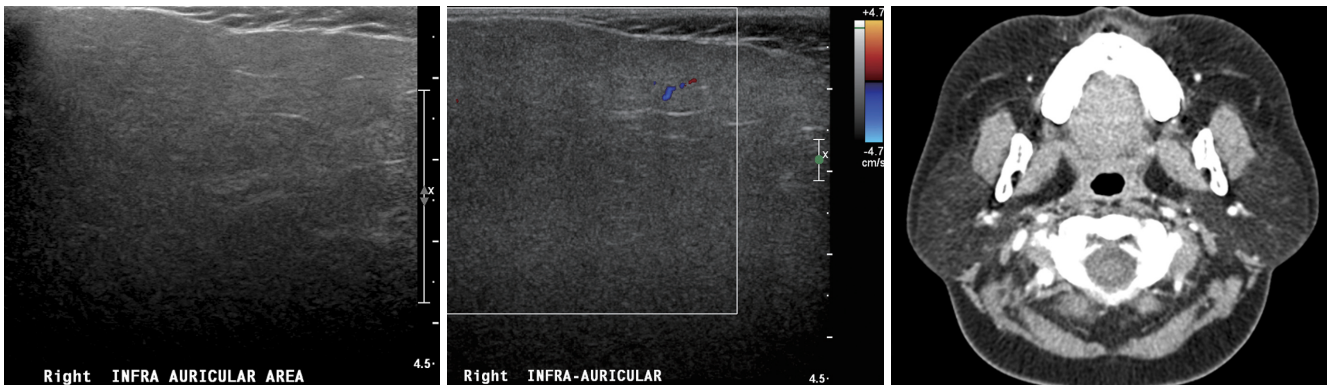

A B C

Fig. 1. Sialadenosis of parotid glands.
A. High-resolution neck sonographic image using 12-5 MHz broadband linear probe shows diffusely enlarged, hyperechoic parotid gland without focal lesions. Due to enlargement of gland and posterior acoustic attenuation, deep lobe of parotid gland could not be evaluated. **B.** Color Doppler Sonographic image using linear probe demonstrates no evidence of increased blood flow in enlarged parotid gland. **C.** Contrast-enhanced neck CT. Axial CT image demonstrates diffuse enlargement of bilateral parotid glands with diffuse hypodensity (-79 - -103 HU) indicateding fat infiltration and without focal lesions. Thin capsule and internal septa of gland are noted. Thick subcutaneous tissue may be secondary to steroid treatment.

the overall size and echotexture of the affected parotid gland. Contrast-enhanced neck CT was performed with a low-dose protocol (100 kV, 61 mAs, volume CT dose index of 3.09 mGy, dose length product of 65 mGy cm, and dose estimate of 0.21 mSv) to exclude possible abnormalities in deep neck structures that could not be evaluated by US. As demonstrated on US, diffuse enlargement of bilateral parotid glands with uniform parenchymal hypodensity (-79 - -103 HU) without a focal lesion was shown on CT (Fig. 1C). In addition, the fibrous capsule and internal septa of the gland were accentuated on CT (Fig. 1C). Cervical lymphadenopathy and secondary changes in the adjacent subcutaneous tissue were not present on either US or CT. The enlarged parotid glands gradually subsided approximately 1 week later at physical examination without any specific treatments. Neither follow-up imaging study nor pathologic examination was performed. Associated systemic disease such as diabetes mellitus, liver disease, or autoimmune disease, was not found.

DISCUSSION

Sialadenosis is a rare disease entity showing diffuse, non-inflammatory, non-neoplastic enlargement of the major salivary glands. Although elucidation of the detailed etiology and pathogenesis of sialadenosis is dependent on further research, it is thought that a loss and thinning of the myofilament component of the myoepithelial cells leads to a loss of mechanical support for the acini of the salivary gland (2). This alteration at the micro-histological level allows acinar cells to expand as secretary granules accumulate in the cells, which leads to the gross change of diffuse enlargement of the salivary gland. The functionally deficient myoepithelial cells in sialadenosis may result from an autonomic neuropathy secondary to severe and long-standing metabolic or hormonal disorders such as diabetes mellitus, alcoholism (particularly with liver disease), liver cirrhosis, chronic malnutrition, and medications (1-4). Almost all cases with sialadenosis have been reported in adult patients. This exceeding rarity in children may be related to underlying diseases common in adults and also due to the probable long latent period required for the development of this condition. In our case, longstanding medications and nutritional problems may be related to the development of sialadenosis but leukemia itself has not been reported to be associated with its development. In fact, chemotherapeutic agents, such as thiouracil and valproic acid, are known to be associated with the development of sialadenosis (1). In contrast, the association between steroid treatment and the development of sialadenosis has not yet been elucidated.

For the diagnosis of sialadenosis, other disease presenting with persistent bilateral parotid swellings, including infection, autoimmune disease, granulomatous disease, kimura disease, polycystic parotid disease, iatrogenic causes, and neoplastic disease, should be excluded (1, 5, 6). Salivary glands may be rarely involved by leukemia/lymphoma and may often manifest as a painless, progressive swelling. The involvement with leukemia/lymphoma is usually associated with autoimmune disease such as Sjögren

syndrome or rheumatoid arthritis (6). Although imaging findings are not pathognomonic, single or multiple salivary gland lesions with increased blood flow and sometimes with microcysts may be revealed on US, CT, or MRI (4).

For the imaging evaluation of the salivary glands, various imaging modalities including sialography, scintigraphy, US, CT, and MRI may be used. When the clinical diagnosis of sialadenosis is confident, it is rarely imaged (5). However, further evaluation with imaging methods may be required when other diseases need to be excluded. In sialadenosis, US reveals diffusely enlarged, hyperechoic salivary glands with posterior acoustic attenuation but without focal lesions or increased blood flow (4). Color Doppler US appears particularly useful to exclude infectious, inflammatory, and neoplastic infiltration as a cause of the diffuse enlargement by demonstrating no evidence of focal or diffuse hypervascularity in the salivary glands. In this case, the absence of focal lesions and the lack of increased blood flow or enhancing portions in the gland on US and CT were greatly helpful in differentiating this lesion from leukemic infiltration or infectious complications. The lack of cervical lymphadenopathy and secondary changes in the adjacent soft tissue on US and CT were additional clues to exclude leukemic infiltration or infectious complications. CT findings of sialadenosis were reported to be possibly biphasic (3). Initially, parenchymal hypertrophy may increase parenchymal density, approaching the density of the muscle (6). Then, fat infiltration in the salivary glands, proven by histologic examination, considerably decreases parenchymal density. CT numbers measured in the affected parotid glands in our case were -79 - -103 HU, confirming fat infiltration in the lesion. However, it remains to be clarified whether this biphasic phenomenon is a common feature of sialadenosis and the fat infiltration represents an advanced stage of sialadenosis (3). MRI may be advantageous over CT in soft tissue characterization of the salivary glands affected by sialadenosis.

We report a sialadenosis case presenting with bilateral painless parotid swellings in a child with leukemia. US and CT findings of this rare case are described.

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