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Case Report

Multimodality Diagnostic Features and Treatment by Sialography of Juvenile Recurrent Parotitis: A Case Report

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Abstract

Juvenile recurrent parotitis (JRP) is a rare disorder characterized by recurrent painful enlargement of the parotid gland on either one or both sides in children without suppuration or ductal obstruction. The etiology is unknown. The severity and frequency of the attacks vary and are characterized by enlargement of the gland, pain, fever, and decreased function of the gland. The diagnosis is usually made clinically and confirmed by imaging. We report a case of JRP in a 12-year-old girl and describe the imaging features of JRP in various modalities.

Key words: Juvenile recurrent parotitis, Magnetic resonance imaging, Ultrasound

INTRODUCTION

Juvenile recurrent parotitis (JRP) is recurrent inflammation of parotid gland occurring in children between 4 months and 15 years of age, is of unknown etiology without suppuration or ductal obstruction and subsides at puberty.[1-7] The inflammation episodes are characterized by enlargement of unilateral or bilateral parotid glands with pain, erythema, sometimes fever, and decreased function of the gland.[1-7] The disease is more common in boys, commonly unilateral than bilateral, and if bilateral it is more severe on one side.[1,2,4,5] Diagnosis is made by clinical examination and parental history of recurrent swelling in the region of parotid gland and confirmation is usually done by imaging techniques.[1-7]

CASE REPORT

A 12-year-old female child presented with complaints of swelling around both ears, more on the left side since the last 4 days. There was history of multiple previous such episodes during the last three years. There was no history of fever, joint pain, rashes or dryness of mouth or eyes. There was no family history of similar episodes of swelling around the ear. Examination revealed a diffuse, firm, mildly tender swelling at the left parotid region and a milder nontender swelling over the right parotid gland. There was serous discharge at the Stensen's duct orifice on applying pressure over the gland and there was no erythema surrounding the orifice. Blood counts were normal and the erythrocyte sedimentation rate was mildly raised (45 mm). Serology for human immunodeficiency virus, rheumatoid factor, anti-Ro, anti-La, and antinuclear antibodies were negative. The child was diagnosed to have JRP. Ultrasound showed multiple round hypoechoic areas interspersed on a background of heterogeneous glandular echotexture, and no vascularity on color Doppler (Figure 1). Magnetic resonance T2 weighted thin slice images of the parotid gland showed enlarged left parotid gland and small cystic areas scattered within the glands bilaterally (Figure 2). There were also mildly enlarged bilateral jugulodigastric nodes. Symptom relief was suboptimal on conservative management with analgesics and intravenous antibiotics for three days, and a sialography was planned. Sialogram showed multifocal sialectasis in the form of multiple small contrast filling cysts communicating with the ductal system (Figure 3). Further copious irrigation of the parotid

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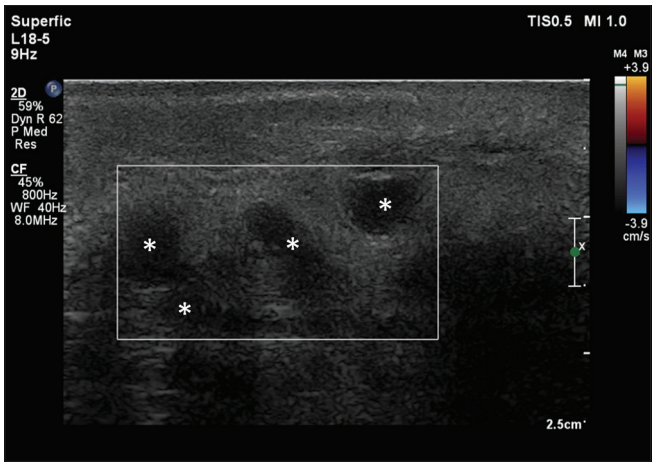

Figure 1: 12-year-old child with juvenile recurrent parotitis. Ultrasound image with Doppler showing multiple round hypoechoic areas (*) without any Doppler signal representing multifocal sialectasia

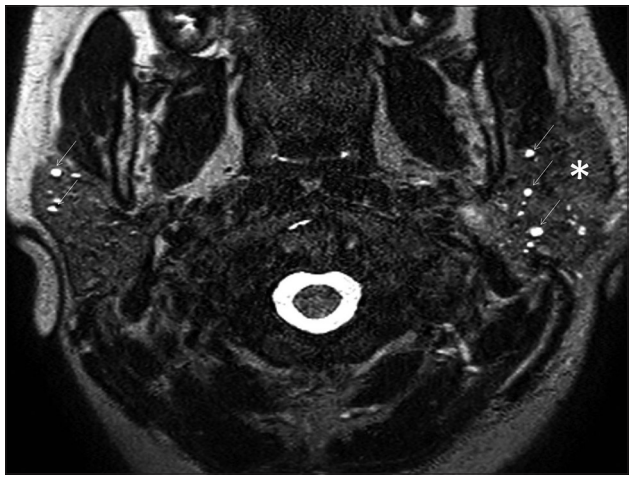

Figure 2: 12-year-old child with juvenile recurrent parotitis. Magnetic resonance T2-weighted thin slice image at the level of parotid glands showing enlarged left parotid gland (*) and multiple small cysts (some of them marked with white arrows) scattered within the glands bilaterally

ductal system with normal saline was performed and the symptoms drastically improved within a day and she was discharged the next day. Parents were counseled that the disease would wear out as the child grows.

DISCUSSION

JRP is usually misdiagnosed as mumps, ear or pharyngeal disease but the history of recurrent inflammation usually points toward proper diagnosis.[1,3] Etiology is unknown and disputed but the disease is considered multifactorial, factors including a congenital ectatic malformation of the salivary ducts, autoimmune factors, viral or bacterial infection, and altered salivary enzyme activity.[2,3,5,8-11] Another hypothesis based on endoscopic picture is decrease in glandular

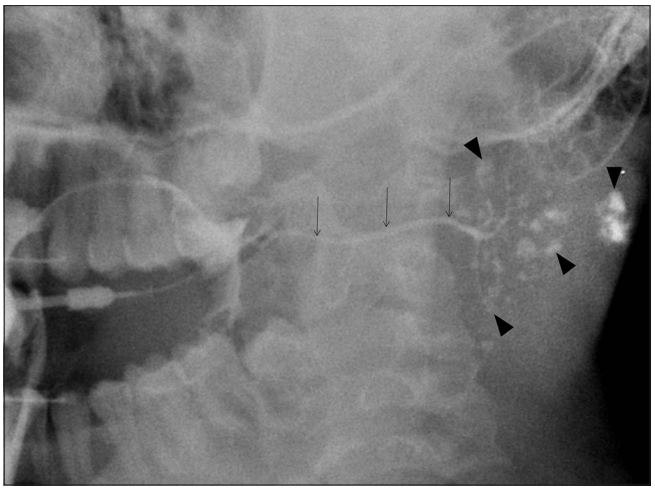

Figure 3: 12-year-old child with juvenile recurrent parotitis. Sialogram showed multifocal sialectasis in the form of multiple small contrast filling cysts (some of them marked with black arrowheads) communicating with the ductal system. Stensen's duct is shown by black arrows

vasculature causing hyposecretion and resultant recurrent infection.[12]

Although the diagnosis is suggested clinically, it is usually confirmed by ultrasound. Ultrasound shows a heterogeneous glandular echotexture representing lymphocytic infiltrate and interspersed small cysts representing multifocal sialectasia.[1-3,6,7,9] The cysts may sometimes show debris within. Magnetic resonance imaging (MRI) of salivary glands with heavy T2-weighting is called MR sialography, which shows gland enlargement, heterogeneous signal intensity of the gland, and multiple interspersed cysts.[2,13] MRI performed during an active phase of inflammation shows enhancement of the gland and cysts are seen in children with previous recurrent parotitis episodes.[8,13] Sjogren's syndrome and sarcoidosis can have similar appearance but have different clinical features.[2]

Sialography performed by injecting contrast medium through a cannula placed in the parotid duct shows intraparenchymal multifocal areas of sialectasis with contrast stasis which is the hallmark of the disease.[1,2,11] Sialendoscopy is another diagnostic modality where the duct walls appear whitish with the absence of ductal wall blood vessels.[1,2,4,5,14] Sialography and sialendoscopy is seldom used currently for diagnosis unless they are used as a therapeutic measure in cases where conservative treatment which consists of analgesics, antibiotics, and plenty of oral fluids fails.[5,15] After sialography or sialendoscopy, the parotid ductal system is irrigated either with saline, steroid or antibiotic solution to clear off viscous saliva, debris, and mucus plugs.[2,4,12,16,17] Our patient showed significant improvement after sialography. Tympanic neurectomy and

parotidectomy have been tried in adult patients with severe parotitis and are not recommended for children.[18,19]

CONCLUSION

Juvenile recurrent parotitis is a rare pediatric non-suppurative recurrent inflammatory disorder of the parotid gland diagnosed clinically, confirmed by imaging, and treated conservatively. Sialography and sialendoscopy have a therapeutic role in patients failing conservative management.

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