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Recurrent Parotitis in Children

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Abstract Recurrent parotitis of childhood is the second most common disease of salivary glands in children next to mumps. It is defined as recurrent parotid inflammation that is non-obstructive and non-suppurative. However, the aetiology and management of this disease remains controversial. We report a series of 5 children presenting with this condition who were investigated by ultrasonography and sialography. All of them were managed conservatively. The aetiology, diagnosis and treatment of this condition is reviewed. A management plan for this disease is formulated to prevent over-investigated and over-treated situations.

Key words Children; Parotitis; Recurrent

Introduction

Recurrent parotitis of childhood, or juvenile recurrent parotitis, is referred as repeated episodes of parotid gland inflammation that is non-obstructive and non-suppurative. It is the second most common disease of the salivary gland in children following mumps.[1]

The aetiology and pathogenesis of this disease however remains uncertain. It is most believed to have a multifactorial cause such as congenital malformation of the parotid glands leading to retrograde infection, allergy and the association with auto-immune diseases.[2,3] The diagnosis is mainly based on history taking and physical examination which is confirmed by imagings such as sialography and ultrasonography of the parotid glands. Management of recurrent parotitis is also controversial. While most tend to treat conservatively with antibiotics and analgesics,[2,4] aggressive therapies have been suggested for the more persistent symptoms, but results vary.[2]

Case Series

In this case study, five children presenting with this condition were identified in the hospital discharge database of Princess Margaret Hospital from 2006 to 2009. Their characteristics, investigation results and imaging findings were summarised in Tables 1, 2 and 3. The typical presentation and imaging findings of recurrent parotitis in one of the patients will be further described. Literature review of the etiology, diagnosis and treatment of this condition will also be discussed. Based on this case study and the literature review, we hope to formulate a management plan for this disease to prevent children from being over-investigated and over-treated.

Four of the 5 patients were admitted through the Accident and Emergency Department and 1 was referred from another hospital for further investigation of the recurrent parotid swelling. The age distribution is between 3 to 5 years old, and the number of recurrence ranges from 2 to 6 times (Table 1). Blood investigation showed a normal or elevated white cell count, and 2 out of the 5 patients had an

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elevated amylase level. Screening for auto-immune markers including IgA, IgG, IgM and rheumatoid factor, and saliva for mumps virus culture and serology were all negative (Table 2). Parotid sialography was performed in all of them and all showed evidence of dilated branches suggestive of sialectasis. These findings corresponded to the multiple hypoechoic areas in the parotid gland seen in the ultrasonography which was performed in 2 of the patients (Table 3). All 5 children were treated conservatively with analgesics and empirical antibiotics before exclusion of suppurative parotitis. They were followed up in our paediatric infectious disease clinic.

Case Report

A 5-year-old girl complained of acute onset of painful swelling at the right angle of her jaw for 1 day. Mastication was painful. She did not have fever, respiratory symptoms, history of local trauma to her face or recent contact with persons suspected of having had mumps parotitis. She was born in Hong Kong and had completed her primary immunisation including a dose of MMR vaccine. She had a history of right parotitis in 2005, which was confirmed not to be mumps by a negative mumps saliva serology and was treated with a course of amoxicillin. No cause of the

Table 1 Characteristics of children with recurrent parotitis hospitalised at Princess Margaret Hospital from 2006 to 2009

Sex	Date of 1st presentation	Age at onset (year)	Number of recurrence	Imaging study	Management
1) Female	2005	5	2	Sialography ultrasonography	Antibiotics + analgesics
2) Male	2006	4	5	Sialography	Antibiotics + analgesics
3) Female	2008	5	3	Sialography	Antibiotics + analgesics
4) Male	2008	5	6	Sialography ultrasonography	Antibiotics + analgesics
5) Female	2009	3	4	Sialography	Antibiotics + analgesics

Table 2 Investigation results of blood (white cell count, amylase and autoimmune markers) and saliva for the five children

White cell count/ neutrophils	Amylase	Autoimmune markers (IgA, IgG, IgM and rheumatoid factor)	Saliva for mumps culture and serology
1) 25/22.8	468	Normal	Negative
2) 7.8/3.9	1005	Not done	Negative
3) 9.6/2.4	266	Not done	Negative
4) 10.2/6.7	113	Not done	Negative
5) 18.9/15.9	274	Normal	Negative

Table 3 Sialography and ultrasonography findings for the five children

Sialography	Ultrasonography
1) Puntate sialectasis + poor emptying of contrast in delay film	Small hypoechoic areas in parotid gland
2) Dilated terminal side branches + poor emptying of contrast in delay film	Not done
3) Dilated terminal side branches + poor emptying of contrast in delay film	Not done
4) Globular sialectasis with parenchymal cystic changes + upholding of contrast in cystic areas in delay film	Multiple hypoechoic areas in parotid gland
5) Dilated terminal side branches + poor emptying of contrast in delay film	Not done

disease was identified at that time. Physical examination showed a 2-3 cm soft tender swelling over the right angle of jaw with ill-defined border, corresponding to the anatomical region of the right parotid gland. There were no dental caries and the right Stenson's duct opening was not inflammed. No stone or pus could be expressed through the opening on bimanual palpation and local massage of the enlarged right parotid gland. Blood tests showed a high amylase level of 468 U/L (normal range: 38-135 U/L) and elevated white cell count of 25.13 x 10^9/L (neutrophil count 22.8 x 10^9/L). Autoimmune markers including anti-nuclear antibody and rheumatoid factor, and immunoglobulin pattern were all normal. Saliva for virus culture and mumps serology were also negative. The patient was treated with analgesics and empirical antibiotic (Augmentin). The swelling resolved spontaneously within a week.

Right parotid sialogram was performed which showed multiple puntate glandular collections suggestive of puntate sialectasis and delayed images showed mild uphold of contrast medium in small ducts (Figures 1 and 2). Ultrasonography of right parotid gland showed multiple hypoechoic nodules corresponding to the puntate sialectasis in sialogram, and enlarged local lymph nodes (Figure 3).

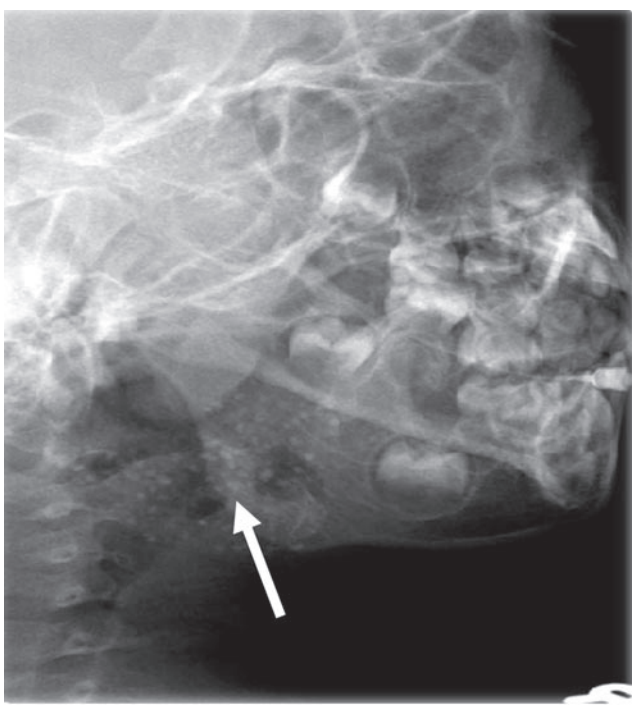

Figure 1 Sialogram of the right parotid gland showing multiple puntate glandular collections, 1 mm in diameter, suggestive of puntate sialectasis.

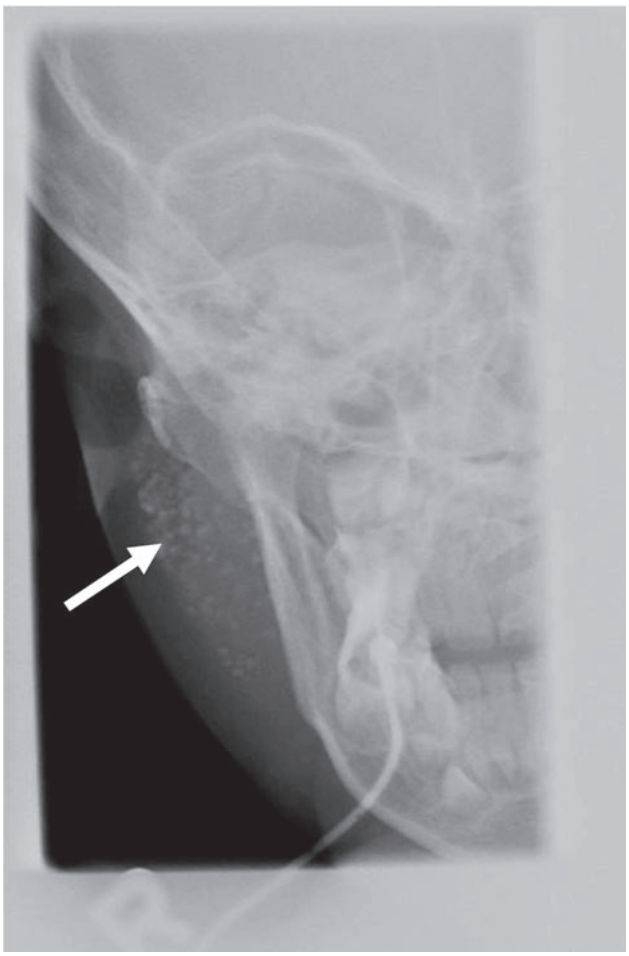

Figure 2 Delayed sialogram image of the right parotid gland showing mild uphold of contrast medium in the eccentric small ducts.

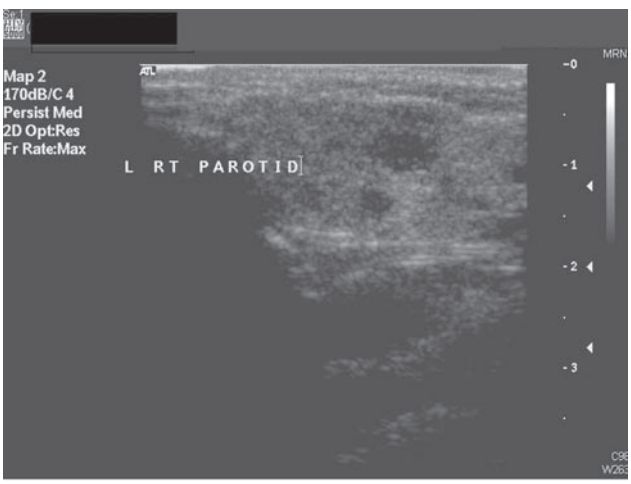

Figure 3 Ultrasound image of right parotid gland showing multiple hypoechoic nodules suggestive of sialectasis.

The girl had a few more recurrence during the next 2 years and were all managed conservatively by antibiotics and analgesics. She is now followed up as an outpatient in our paediatric infectious disease clinic and is managed conservatively with advice of maintaining good oral hygiene and adequate hydration. The number of her disease recurrences has markedly decreased with no recurrence this year.

Discussion

Recurrent parotitis is characterised by intermittent swelling of unilateral or bilateral parotid glands, often associated with fever, malaise and pain with mastication and swallowing. It is commoner in boys with peak age of onset between 3 to 6 years old. Leerdam et al have found a biphasic age distribution of 2 to 5 years old and 10 years old.[5] The exacerbation usually lasts for few days, and occurs every 3 to 4 months. There is, however, a wide variation in the frequency and severity of attacks. It is usually self-limiting and symptoms generally subside after puberty.[1, 2, 5] Differential diagnoses and associated conditions which need to be considered include Sjogren's syndrome in older children, which usually has elevated immunoglobulins; hypogammaglobulinaemia, and immunodeficiency such as common variable immunodeficiency or HIV/AIDS. In our case study, the number of patients is too small to show any significant pattern in gender and age, but they are all treated conservatively with gradual subsiding of symptoms, as they grow older. However, the time needed for the attacks to subside varies.

The aetiology and pathogenesis of recurrent parotitis of childhood remain uncertain. The possibility of congenital malformation of the parotid glands resulting in low salivation rate leading to dehydration and thus recurrent retrograde infection has been suggested.[2] Alternatively, the production of puntate sialectasis is proposed to be due to the damage of the duct reticulum by lymphocytes, as suggested by the typical histological picture of lymphocytic infiltration of the intralobular ducts. However, most of the authors favour a multifactorial cause. Several have associated the disease with viral or bacterial infection, allergy and auto-immune diseases. Fazekas et al have reported a correlation between selective IgA deficiency and recurrent parotitis.[3]

The diagnosis of recurrent parotitis is usually made on a clinical basis as suggested by detailed history taking and adequate physical examination. It is distinguished from suppurative parotitis by the inability to express pus from the parotid duct. Parotid sialogram was the conventional tool for confirming sialectasis, which is a characteristic feature of recurrent parotitis. And in our case study, all of the patients had sialogram performed, which showed evidence of sialectasis, and only 2 had ultrasonography of parotid glands done as well. However, sialogram has been superseded by ultrasonography, which is non-invasive and has been shown to be equally sensitive as conventional sialography.[5] Ultrasonography also provides extra information such as the presence of stones (sialoliths), abscesses or mass lesions. As a result, many authors recommend using ultrasound as the investigation of choice.[2,5] The typical features of recurrent parotitis are the formation of puntate or globular sialectasis scattered throughout the gland without any stones or destructive changes (Figure 1).[4] Sialogram will show multiple round pools of contrast medium about 2-3 mm in size which persist in delayed films (Figure 2). These correspond to the multiple hypoechoic areas seen in ultrasound images (Figure 3). Computed tomography, magnetic resonance imaging and sialendoscopy are also used to assess the parotid gland but they are not widely used due to the relatively high irradiation risk in the former, and the lack of ready availability for the latter.[1,6]

The management of recurrent parotitis in children is controversial. Most authors tend to treat conservatively with analgesics and antibiotics. Although the disease is self-limiting and the use of antibiotics does not shorten the length of the disease, it is believed that antibiotics may prevent additional damage to glandular parenchyma.[2,4] Chitre et al. propose the use of prophylactic antibiotics for preventing recurrence.[2] The evidence for bacterial involvement in the pathogenesis is poor, and no studies compare outcome with and without antibiotics. In our case study, 2 patients were treated with analgesics only in their subsequent attacks after having their diagnoses confirmed by sialogram. Their symptoms resolved in 3 to 4 days which are of the same length of time compared with antibiotics treatment. Unproven interventions such as sialogogic agents (e.g. lemon juice), warmth, massage and duct probing to stimulate saliva flow have also been mentioned.[7] More aggressive therapy such as radiotherapy, parotid duct ligation and parotidectomy have been suggested for disease persisting to adulthood, but results vary and potential permanent damage to the facial nerve is a serious concern.[2] In general, most children need little intervention except reassurance and analgesia, but they are at risk of

over-investigation and over-treatment.[7]

In summary, five children with recurrent parotitis of childhood are reported. Diagnosis was established by history and characteristic sialogram and ultrasound scan findings. All of them were managed conservatively with analgesics and antibiotics.

We recommend a more conservative management for recurrent parotitis in children; the common aim is to alleviate symptoms as most of them will resolve after puberty. Although analgesics alone can alleviate the symptoms, antibiotics should be given before supperative parotitis is excluded. Ultrasonography of the parotid glands should be done to confirm the diagnosis, as it is non-invasive but equally sensitive as sialogram. Other appropriate investigations such as blood tests for white cell count, amylase and auto-immune markers should be done to exclude other differential diagnoses such as Sjogren's syndrome and immunodeficiency.

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