REGULAR REVIEW

Recurrent parotitis

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Recurrent parotitis is defined as recurrent parotid inflammation, generally associated with non-obstructive sialectasis of the parotid gland.[1] Also known as juvenile recurrent parotitis,[2 3] this disease is characterised by recurring episodes of swelling and/or pain in the parotid gland, usually accompanied by fever and malaise. It usually affects children, but may persist into adulthood.(*Arch Dis Child* 1997;**77**:359–363)

It is a rare condition, and its aetiology remains an enigma. Its natural history is variable, and in adults more aggressive intervention is often needed. In addition, there is no satisfactory explanation for its usual tendency to resolve spontaneously after puberty. All this has resulted in considerable uncertainty concerning its appropriate management. This review was undertaken to collate all the information available on this uncommon and distressing condition. Diagnosis and management of this condition is also discussed.

Aetiology

Its cause remains unknown despite several studies. Though the affected glands demonstrate sialectasis of the distal ducts, there seems to be no element of obstruction in most cases. Several theories of causation have been put forward over the years.

Traditionally, ascending infection from the oral cavity has been considered the primary event, with sialectasis being a secondary change. Maynard proposed that the recurrent episodes of parotid swelling was the end result of a sequence of events[4]:

- There is first a low grade inflammation of the gland and duct epithelium, possibly caused by a low salivary flow rate due to dehydration and debility.
- This results in distortion and stricturing of the distal ducts, and metaplasia of the duct epithelium.
- The metaplasia results in excessive mucus secretion.

These changes, along with possibly a further reduction in salivary flow rate, then predispose to recurrent parotid inflammations.

A reduced salivary flow rate may result from glandular damage caused by the primary infection. However, it may be a primary factor as well. Several workers showed low salivary flow rates,[4–7] and the significant finding was that the flow rate was reduced in even the unaffected glands in patients with unilateral disease.[4] This suggests that those with low salivary flow rates might be predisposed to suffer from repeated ascending infections. This relation to salivary flow rates could also explain the familial tendency that has been reported.[3 8 9]

The histological picture includes lymphocytic infiltration around the intralobular ducts, and Patey and Thakray proposed that this lymphocytic infiltration damages the duct wall reticulum, allowing extravasation of secretions into the gland parenchyma, and thus exacerbating the inflammation.[10] This was supported by Hemenway and others.[4 11 12]

The fragmentation of connective tissue supporting the intralobular ducts was also implicated by these authors in the production of the characteristic punctate sialectasis. They proposed that the dye used for sialography ruptured the already weakened duct walls, producing the appearance of punctate sialectasis. This theory therefore neatly explained the presence of sialectasis in the absence of demonstrable distal obstruction.

The situation, however, is not as simple. Punctate sialectasis is seen in totally asymptomatic glands of affected individuals in up to 70% of cases.[13] Further, detailed histopathological studies have confirmed the presence of duct dilatation and cystic cavities associated with a chronic inflammatory process.[2 14] And more recently, ultrasonography consistently revealed hypoechoic areas that corresponded to the punctate sialectases demonstrated by sialography.[15] It therefore would appear that the sialectases are actually present, and are not merely artefacts produced by the radio-opaque dye.

As long ago as 1945, Hamilton Bailey proposed the presence of a congenital abnormality of the ductal system, and drew a parallel with bronchiectasis.[16] He pointed out that bronchiectasis could be congenital as well as acquired, and in both cases, the end result was secondary infection of the bronchioles and alveoli. He has been subsequently supported by several others.[1–3 14] According to this argument, punctate dilatation of the small distal ducts results in stasis and ascending infection, giving rise to the recurrent acute attacks. Though no evidence has so far emerged in favour of a congenital abnormality, it is still possible that genetic factors may prove important.

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Given this state of incomplete knowledge, the present consensus is towards a multifactorial approach. Thus Kono and Ito concluded that the sialectasis is both the cause and the result of recurrent parotitis.[14] Their histological studies detected dilated cavities consistent with true sialectasis, as well as a few areas of extravasated dye which mimicked sialectasis on the sialogram. Similarly, a detailed study of clinical, radiological, immunological, bacteriological, and histological findings in 20 affected children concluded that the cause was probably a combination of a congenital malformation of portions of the salivary ducts and infections ascending from the mouth after dehydration of the affected children.[2] However, it must also be acknowledged that juvenile recurrent parotitis can occasionally occur without sialectasis.[1 14 17] One child has suffered repeated attacks of parotitis secondary to repeated chewing of the Stenson's duct orifice.[18]

Many associations have been proposed in the past; these include immunodeficiency, allergy, upper respiratory infections, mumps, etc. None of these, however, has been conclusively shown to have any bearing on this disease.[2] Friis *et al*[19] and others[14 17] proposed an autoimmune origin, but the self limiting nature of recurrent parotitis and the absence of detectable autoantibodies makes this unlikely.[2 13 14 20] There have been reports of sensitivity to upper respiratory tract infections[2 14 21]; these infections may set off attacks of sialadenitis merely by causing dehydration in a child with sialectasis.[2]

The higher rate of secretion in the submandibular gland compared with the parotid gland may protect it from infections[2]; also, the submandibular gland secretion is relatively richer in mucus, which has antiseptic properties. In fact, no report was found in the literature implicating the submandibular gland.

Clinical features

Recurrent parotitis presents as a recurrent painful swelling during mastication and/or swallowing.[22] The disease usually starts in a child between 3 and 6 years of age,[1 3 13 23] but earlier and later occurrence has been observed.[2 14 17 24 25] Ericson *et al* followed up 20 children and the age of onset in their series ranged from 3 months to 16 years.[2]

Most studies report a sex distribution favouring males.[1 23 24 26] Seventy two per cent of patients were male in the series of Geterud *et al* of 25 patients.[1] However, Watkin and Hobsley found from a study of 68 patients, of whom 26 were children, that the sex distribution was equal in childhood, and actually affected females more than males (7.5:1) in patients where the onset of symptoms was after the age of 16 years.[13]

The symptoms are usually unilateral; when bilateral, the symptoms are more prominent on one side.[2 3 23 27] In a series of 25 patients, symptoms were unilateral in 18 patients and bilateral in seven patients (28%).[1] Katzen and Du Plessis, in 1964, reported a higher incidence of bilateral presentations than that observed by more recent studies.[28]

The number of attacks vary individually, with attacks every three to four months being the commonest pattern.[1 3] Mandel and Kaynar state that attacks tend to occur one to five times a year.[23] The frequency rate peaks during the first year at school, but otherwise remains fairly constant for each individual until puberty. After puberty, the symptoms usually subside, and may disappear completely.[1–4 14 17 25] Geterud *et al* reported that 84% of their patients had recovered by the time they attained puberty.[1] Further, another 8% of their patients were considered cured by the time they reached the age of 22. Galili and Yitzhak proposed two possible ways by which this spontaneous recovery might occur: total atrophy with consequent lack of symptoms, or regeneration of the gland from surviving ductal system.[29] The authors favoured regeneration as the likely mechanism. However, there are also persistent cases.[1–3 13 20 23 29 30] The actual proportion of 'persisters' is debatable, though most researchers agree that the numbers are small.[1 3 4 11 14]

The painful swelling is usually associated with fever.[2] There is typically an absence of pus despite the pyrexia and malaise,[3] though Geterud *et al* noticed a few drops of mucopurulent secretion on palpating the parotid gland.[1] The swelling lasts from several days to two weeks,[23] and resolves spontaneously, independent of any treatment.

Investigations

(1) SIALOGRAPHY

The mainstay of diagnosis is sialography, though its role is now becoming secondary to ultrasonography. In 1971, Hemenway classified sialectasis into (a) *large duct sialectasis*, due to obstruction of the main duct, and (b) *small duct/punctate/terminal sialectasis*, in which there are multiple small round opacities at the termination of the smaller ducts.[11] Later, Gates[31] and Noyek *et al*[32] reviewed the classification of punctate sialectasis, and further classified this into (a) *pruned tree appearance*—in the early stages, the ducts are stretched, tapered, and decreased in number; (b) *punctate sialectasis*—the peripheral ducts demonstrate punctate dilatation <1 mm diameter, and the intraglandular ductal system is stretched and tapered; (c) *globular sialectasis*—the ducts are between 1 mm and 2 mm in size and they may be partially non-visualised or irregular, giving rise to a 'mulberry pattern' fruit laden tree appearance; (d) *cavitatory sialectasis*—coalescence of cystic lesions produces a cavitatory appearance; and (e) *destructive sialectasis*—there is a bizarre pattern of pooling, possibly with stones in the gland.[33]

The typical changes in recurrent parotitis are punctate and globular sialectasis, which are scattered throughout the gland; cavitatory and destructive sialectasis are not seen.[2]

These changes are usually bilateral even if the presentation is unilateral.[1 2 13 14 23] Ericson *et al* found eight out of 12 clinically symptomless glands demonstrated sialectasis.[2] The lesions were however smaller and fewer in the symptom-free glands. Watkin and Hobsley

found punctate sialectasis in 69% of asymptomatic glands.[13] On the other hand, sialographic changes are not always bilateral, as had been previously believed.[1] Several authors report that these changes tend to diminish and sometimes disappear after the disease becomes quiescent.[3 14 23 29] However, Geterud *et al* found that only two out of 16 patients demonstrated a significant reduction in the sialectasia and further, these two patients had only minimal primary changes.[1]

In glands with sialectasis, peripheral intraglandular ducts are invisible,[1 2 5 13 27 30] indicating that the changes affect the peripheral parts of the ductal tree. The main ducts may be affected as well. Ericson *et al* found slight to moderate dilatation of the main duct in 25% of symptomatic glands,[2] while Geterud *et al* reported severe main duct changes in 8% of their patients.[1] However, the presence of main duct dilatation did not influence the clinical course of these patients, as had previously been proposed.[4] Other findings on sialography include acinar and ductal atrophy, with impaired glandular function.[3]

There is some controversy whether sialographic changes correlate with clinical symptoms. Ericson *et al* found a strong relationship between clinical symptoms and multiple sialectases.[2] However, Geterud *et al* reached exactly the opposite conclusion.[1]

(2) DIGITAL SUBTRACTION SIALOGRAPHY

This has been recommended as superior to conventional sialography for the recognition of inflammatory changes and chronic sialolithiasis.[34]

(3) ULTRASONOGRAPHY

Ultrasonography consistently revealed hypoechoic areas that corresponded to the punctate sialectases demonstrated by sialography.[15] The authors therefore recommended ultrasonography as the primary investigation for diagnosis, in addition to follow up. An earlier study had also found ultrasonography useful,[35] though a direct comparison between conventional sialography, digital subtraction sialography, and ultrasonography had found ultrasonography less sensitive than the other two for inflammatory changes and sialolithiasis.[34] It is possible that improvements in radiological skills and equipment may have now rendered ultrasonography as reliable as sialography.

Ultrasonography may completely replace sialography in the near future. Murrat *et al* have recently proposed a protocol for the investigation of investigating a case of intermittent pain and/or swelling of the salivary glands, and they recommend an initial ultrasound.[36] If this investigation reveals calculi/duct dilatation/cysts/gland enlargement, they then proceed to sialography. Duct dilatation having been already revealed by ultrasonography, the role of sialography would be to rule out duct stenosis or obstruction. If, on the other hand, ultrasound reveals a solid mass, computed tomography or magnetic resonance imaging is indicated. If the ultrasonography reveals no abnormality, sialography is performed only if symptoms recur.

(4) CYTOLOGY

Salivary smears of normal children are acellular. In contrast, saliva in the presence of sialectasis revealed large amounts of granulocytes, some lymphocytes, and in about 50% of cases, bacteria.[2] The bacteria were mixed, and included aerobic and anaerobic cocci.

(5) HISTOLOGY

Dilated interlobular ducts with lymphocyte infiltration in the surrounding tissues is seen[2]; the lymphocytes tend to form lymphoid follicles. The duct epithelium shows hyperplasia and metaplasia, with a pseudostratified cylindric pattern being common.

Treatment

Uncertainty about its aetiology has hampered the development of a universally accepted treatment strategy for this condition. The problem is compounded by the rarity of this disease and its uncertain natural history.

Treatment of the acute episode aims to deliver relief of symptoms and to prevent damage to the gland parenchyma. Analgesics and antibiotics have been found to be rapidly effective in relieving the pain and swelling.[2 13] Though co-amoxiclav may be used, penicillin is considered adequate, as the infecting agent is not usually a staphylococcus.[2] Most workers agree in practice, but many question whether antibiotics really change the natural course of the disease,[1 28] and wonder if resolution of symptoms could merely reflect the natural progression of this disease.[23] In addition to antibiotics and analgesics, other treatments include sialagogic agents to increase salivary flow, warmth and massage, and duct probing.[2 23] The logic behind probing is questionable[1] as duct dilatation and not duct stenosis is the underlying pathology. The treatment seems to be effective, however, and may help by clearing the plugs of mucus and cells that form in the acute phase. Bailey recommended duct cannulation and lavage with 1% mercurochrome.[16] Steroids may reduce swelling, but will not prevent recurrences.[13 23]

Prevention of recurrences is difficult, but preventing dehydration and prescribing a prophylactic course of penicillin/co-amoxiclav during winter may help; no studies are available, however, to substantiate this belief.

The treatment of repeated attacks is more difficult, and several different methods have been tried.

(1) RADIOTHERAPY

This was used for several years,[1] despite it being considered useless by several researchers more than 30 years ago.[37] In fact, the literature contains virtually no evidence in favour of this method of treatment, and yet radiotherapy was being used as the sole treatment at several centres.[13] Its popularity probably rested on its perceived efficacy in reducing the mortality from acute bacterial parotitis by about 50% in

the 1930s,[11] though the author rightly pointed out that the improved prognosis of that condition was probably due to improved medical care.

(2) DUCT LIGATION

This was popularised by Diamant and Enfors.[38] It has been used successfully by others,[1] though at least one study had described varying results.[12] Geterud *et al* recommend it as a simple and effective treatment.

(3) PAROTIDECTOMY

Parotidectomy has always been the gold standard for obtaining permanent relief. With this operation, however, one is faced with the risk of facial nerve injury; this is especially relevant in recurrent parotitis, where repeated infections result in fibrosis of the gland.

(4) TYMPANIC NEURECTOMY

This procedure has recently been recommended as an effective procedure,[22] with good results in 70% cases. Its aim is to destroy secretomotor fibres to the parotid gland thus abolishing/reducing its secretion. The authors recommend extensive interruption of the secretomotor fibres by thoroughly drilling into the hypotympanum and below the basal turn of the cochlea. Several others have also reported similar results.[39 40] Nerve regeneration and incomplete sectioning are probably responsible for the early and late failures; it is therefore necessary that the procedure is performed by an experienced otologist.[22]

(5) OTHER METHODS

Various other treatment options have been studied. Bowling *et al* proposed intraductal tetracycline instillation as an effective, low risk treatment.[41] They hypothesised that tetracycline produced acinar atrophy and demonstrated encouraging results in rabbits. No studies have been conducted on human subjects.

Some studies found that performing a sialographic study itself resulted in significant improvement of symptoms.[3 34 42] The hypothesis was that improvement resulted as a result of the flushing, dilating, and antiseptic actions of the iodine containing dye.[23]

Finally, the fact that many patients seem to recover spontaneously has led researchers to support a conservative approach. Watkin and Hobsley found that 56% adults and 64% children recovered with only symptomatic treatment, over a five year period.[13] Similarly, Geterud *et al* reported that symptoms disappeared by the age of 22 in 23 of 25 patients.[1] A conservative approach therefore is recommended for children, with more aggressive treatment being reserved for the 40% adults and 4% of children[7] whose symptoms persist or worsen.

A suggested management plan

INVESTIGATIONS

The investigations to perform if there is a clinical suspicion of recurrent parotitis are shown in fig 1.

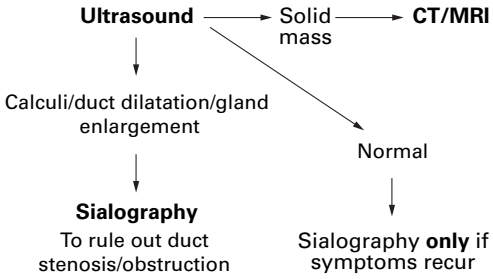

Figure 1 Investigations (proposed by Murrat et al[36]); CT = computed tomography, MRI = magnetic resonance imaging.

TREATMENT OF ACUTE EPISODES

Aim: to relieve symptoms and prevent damage to gland parenchyma.

- Analgesics
- Antibiotics—penicillin/co-amoxiclav
- Sialogogic agents
- Warmth and massage
- (? Probing of duct and ? steroids for severe attacks).

PREVENTING REPEATED ATTACKS

(Preventing repeated attacks is usually unsuccessful.)

- Prophylactic antibiotics—penicillin/co-amoxiclav
- Prevent dehydration.

RECURRENT ATTACKS CONTINUING INTO ADULTHOOD

- Duct ligation—simple operation, but with variable results
- Parotidectomy—good results, but with a small risk of facial nerve damage
- Tympanic neurectomy—good results in the hands of experienced otologists.

Summary

(1) Recurrent parotitis is probably caused by a congenital abnormality of the salivary gland ducts with recurrent attacks of ascending infection, perhaps aided by dehydration. The parotid gland is predominantly affected probably because of its lower rate of secretion compared with the submandibular gland.

(2) The condition mainly affects children between the ages of 3 and 6, with males being more commonly affected. The symptoms peak in the first year of school, and usually, but not invariably, begin to subside at puberty. By the age of 22, most patients are completely symptom-free. When the disease starts after puberty, females are predominantly affected.

(3) Ultrasound is the appropriate initial investigation, and is usually supplemented by sialography. The sialography may itself cause a resolution of symptoms.

(4) Treatment is conservative in the first instance, and an expectant policy is indicated. More aggressive treatment is justified only for those adults with persistent problems. This may be parotid duct ligation, parotidectomy, or tympanic neurectomy, depending upon the preference and experience of the treating physician.

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