Sialographic and Ultrasonographic Features of Juvenile Recurrent Parotitis

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Abstract

The sialographic, and ultrasonographic features were studied to investigate the relationship between sialographic, and ultrasonographic features in this disease. Forty-five cases of juvenile recurrent parotitis were studied, age ranging from 2 to 13 years. Seventy-nine parotid glands were investigated both sialographically and ultrasonographically. Sialographic features were classified into 4 stages: delayed emptying, punctate, globular, and cavity sialectasia. Ultrasonographic observation included echo level, echo heterogenicity, and pseudomass lesions. Sialographically, 16 glands showed delayed emptying, or mild dilation of main duct; 16 punctate, 22 globular, and 25 cavity sialectasia. Ultrasonographic evaluation revealed normal echogenicity in 28 glands, heterogenicity in 32 glands, hypoechoic, heterogeneous internal echoes in 14 glands, and occupying lesions in 5 glands. Comparative study between the sialographic and ultrasonographic features showed that normal echogenicity was observed in those with delayed emptying or mild dilation of main duct. Hypoechoic, heterogeneous internal echoes could be detected in about two third of those with punctate sialectasia; and more often in those with globular or cavity sialectasia. Our results implied that ultrasonographic features reflected the extent of sialectasia in juvenile recurrent parotitis. Ultrasonography should be adjuvant in diagnosing the disease.

Key Words: Juvenile Recurrent Parotitis/Sialography/Ultrasonograph

Introduction

Juvenile recurrent parotitis (JRP) is, next to mumps, the most common inflammatory disease of the salivary glands in childhood. The pathogenesis of the disease is considered to be infections ascending from the mouth following dehydration from a respiratory
infection. Among other factors that have been considered are congenital malformation of the ducts and local manifestation of systemic immunologic diseases. The condition is characterized by periodic swelling of the parotid gland, sometimes accompanied by pain, fever and malaise. The disease starts early in life and the symptoms usually subside after puberty. The sialographic image of the diseased gland is quite characteristic and shows multiple punctate sialectasis. Ultrasonography has been widely used in the diagnosis of salivary gland diseases as a non-invasive, non-radiative imaging modality. However, there are few reports on comparative study between sialographic and ultrasonographic features of JRP. In this investigation, the sialographic, and ultrasonographic features were studied in order to elucidate the relationship between sialographic, and ultrasonographic features in this disease.

Patients and methods

The sample consisted of 45 cases of juvenile recurrent parotitis, 31 boys and 14 girls between 2 and 13 years of age at the time of investigation. The average age for the test group was 7 years. Seventy-nine parotid glands were investigated both sialographically and ultrasonographically during symptom-free intervals. Ultrasound scan was performed before sialography, with TOSHIBA SSA 220 A scanner and a 7.5 MHz transducer. Echo intensity level, echo heterogeneity, and pseudo-mass lesions were recorded. Water-soluble contrast medium was used for sialography, and exposures were made during the filling and emptying phases. Sialographic features were recorded as 4 stages, delayed emptying, punctate, globular, and cavity sialectasia.

Results

1. The results of the sialographic investigation are listed in Table 1 and illustrated in Fig. 1. The results showed that multiple sialectasis of varying order is the major feature of JRP. Apart from sialectasis, the main duct was intact or slightly to moderately dilated, and the secondary and tertiary ducts partly invisible.

2. The results of ultrasonographic evaluation are listed in Table 2 and illustrated in

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<tr>
<th>Table 1 Sialographic features of JRP</th>
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<tr>
<td>Delayed emptying</td>
</tr>
<tr>
<td>No. of glands</td>
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<tr>
<th>Table 2 Ultrasonographic features of JRP</th>
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<tr>
<td>Normal</td>
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<tr>
<td>No. of glands</td>
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Fig. 1 Sialographic image of a 4-year boy, showed punctate and globular sialectasis, accessory gland was also involved.

Fig. 2 Ultrasonographic image of a 5-year boy, showed heterogeneous pattern with decreased echo intensity.

Fig. 3 Ultrasonographic image of a 6-year boy, showed heterogeneous pattern with normal echo intensity.

Fig. 4 Sialographic image of a 4-year girl, showed globular and cavity sialectases.

Fig. 5 Ultrasonographic image of the same gland with Fig. 4, showed heterogeneous echo with decreased intensity.

Figs. 2 and 3. The Heterogeneity, with normal or decreased echo intensity, is the major ultrasonographic feature of JRP.

3. Comparative study between the sialographic and ultrasonographic features of JRP was shown in Table 3. It is shown that normal echogenicity was observed in those with delayed emptying. Heterogeneous internal echo, normal or decreased intensity, could be detected in 69% of those
Table 3 Comparative study between the sialographic and ultrasonographic features

<table>
<thead>
<tr>
<th>Sialographic</th>
<th>Ultrasonographic</th>
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<tr>
<td></td>
<td>Normal</td>
</tr>
<tr>
<td>Delayed emptying</td>
<td>16</td>
</tr>
<tr>
<td>Punctate</td>
<td>5</td>
</tr>
<tr>
<td>Globular</td>
<td>5</td>
</tr>
<tr>
<td>Cavity</td>
<td>2</td>
</tr>
<tr>
<td>Total</td>
<td>28</td>
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with punctate, and 68 % globular sialectasia; and in 80 % of cavity one (Figs. 4 and 5). Five glands with solitary or multiple mass lesions showed globular or cavity sialectasis, without occupying features seen sialographically (Figs. 6 and 7).

Discussion

Juvenile recurrent parotitis is an unusual childhood inflammatory disease of the parotid salivary glands, characterized by intermittent episodes of parotid swelling over a period of years. The sialographic image of the diseased gland is multiple punctate sialectasis of varying order appearing in the peripheral parts of the duct tree, which is quite characteristic for JRP. Whether there is a high relationship between clinical symptoms and multiple sialectases is still under discussion. Our study showed that 80 % of the diseased glands showed sialectasis. Sialectases were also found in clinically uninvolved glands. Therefore, sialography is still an unsurpassable procedure in visualizing ductal changes, which are actually the characteristic of JRP. This is also thought to be the reason for the low
sensitivity of ultrasonographic investigation in this study.

Although sialography is considered the most reliable for diagnosing JRP, it is not a desirable method because of ionizing radiation, some contraindications, and difficulty in cannulation. Ultrasonographic investigation is a non-invasive, non-radiation modality for diagnosing salivary diseases. The result from the present study showed that the major ultrasonographic feature of JRP is heterogeneous pattern, with normal or decreased echo intensity. The results are in agreement with earlier studies. Unfortunately, ultrasonography is not so sensitive as sialography. In our report, it is positively detected in only 65% of the affected glands. This study lends no support to the suggestion that sialography could be replaced by ultrasonographic evaluation in diagnosis of JRP. It has been reported in previous study that the incidence of heterogeneous glands was up to 80% of the diseased glands. The inconsistency might be due to different inclusion criterion, because only the children with typical sialographic features of JRP were collected in their study.2

The comparative study between sialography and ultrasonography indicated that those with more advanced sialographic stages of globular and cavity sialectasia tend to show heterogeneous in echogram, implying the change in echogeneity is well associated with sialectasis shown on sialogram.

Histologically, these sialectases correspond to dilatations in intralobular ducts and appear in areas with massive, chronic, lymphatic infiltration.2,3 It is speculated that some internal interfaces were produced and the acoustic characteristic of the diseased parotid gland was altered by peripheral sialectasis, resulting in abnormal echogeneity. The previous report also revealed that larger the sialographic shadows were, the more the internal echoes became hypoechoic and heterogeneous.2 Our study also suggested that there might also be other factors involved in the formation of the altered echogenicity, apart from sialectasis, because some glands with advanced sialectasis showed normal echo feature. Further study is needed to verify the histopathologic basis of heterogeneous pattern of the disease.

There is little histopathologic information about occupying lesions in juvenile recurrent parotitis, it needs further study to verify whether it represent lymphocytic infiltration in the gland involved, as shown in some Sjogren syndrome cases. They were also hypothesized as lymphadenitis in some case because some solitary lesions disappeared in the follow up. Rubaltelli et al10 reported an enlarged lymph node in one of their JRP cases.

Conclusion

Sialography is still the most important and most reliable diagnostic method of JRP. The major ultrasonographic feature of JRP is heterogeneous pattern. Our results implied that ultrasonographic features reflected the extent of sialectasia in juvenile recurrent parotitis. Therefore, ultrasonography could be an adjuvant and useful test in diagnosing the disease.

References

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小児の反復性耳下腺炎の唾液腺造影並びに
超音波検査による臨床的研究

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小児の反復性耳下腺炎（JRP）の唾液腺造影所見と超音波検査所見を比較検討した。2 歳から13 歳までのJRPに罹患している45 人（男児31 例、女児14 例）を対象にして、79 例の耳下腺造影ならびに超音波による検査を行った。

超音波検査所見：（1）形態的には正常であるが造影剤の排出が遅い、（2）微小導管の点状拡張、（3）微小導管の球状拡張、（4）微小導管の管状拡張などである。超音波検査所見：（1）エコーは正常、（2）エコーは不均一である、（3）不均一で低エコーを示す、（4）エコー像の消失。

造影所見並びに超音波検査を比較すると、形態的には正常な造影像を示している耳下腺はエコー像においても正常な像が得られた。微小導管の点状拡張を示す所見の約半分はエコー像においても正常な像が得られた一方、微小導管の球状拡張、管状拡張所見を示すものは、不均一な低エコー像やエコー像の消失を呈した。

以上の結果から、耳下腺のエコー像は導管の形態を精密にみることは難しいが、耳下腺全体の形態を把握することが可能である。したがって超音波検査は小児の反復性耳下腺炎診断の補助的手段として応用できることが示唆された。