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ORIGINAL RESEARCH

Sialoscopy in Cases of Unclear Swelling of the Major Salivary Glands

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OBJECTIVE: Sialoscopy has developed into an important diagnostic and therapeutic tool for diseases of the major salivary glands.

STUDY DESIGN AND SETTING: We evaluated 103 patients with chronic swelling of the major salivary glands. Routine diagnostic measures revealed no clear diagnosis. The findings of 109 sialoscopies are described. A semi-rigid endoscope (with a diameter of 1.1 mm and 2 integrated working channels) was used for sialoscopy, 51.5% of the cases in Warthon′s duct and 48.5% in Stensen's duct.

RESULTS: Pathologic findings resulted in 83.0% of the submandibular and in 96% of the parotid ducts. Obstruction neither due to sialolithiasis nor stenosis was observed in 56.3%, whereas sialolithiasis was observed in 20.3% of the patients. In 36 (35%) patients, an interventional sialoscopy was performed.

CONCLUSIONS: In cases of invisible salivary duct obstruction, especially in those with low mineralized calculi, strictures, stenoses, or postinflammatory changes, sialoscopy gives immediate and direct information about causal pathologies. Moreover, further therapy can be planned within the same procedure.

EBM RATING: C

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Swelling of the major salivary glands is one of the main symptoms that leads patients to our clinic. Although the diagnostic procedures and apparatus have continually developed to a higher standard, in some cases it is not possible to establish an accurate diagnosis. This is the case, for example, in small sialoliths that are responsible for obstructive non-neoplastic disorders of the major salivary glands in many cases.[1-4] The most widely used diagnostic tools are ultrasound, x-ray examination, and sialography.[2,3,5,6] Between 15% and 30% of all submandibular concrements and up to 40% to 60% of all parotideal stones are not detectable by plain x-ray or other conventional radiological means.[2,6-11] Sialography is the gold-standard examination in many centers and can reveal sialolithiasis with high sensitivity and specifity.[5,6] It is contraindicated in acute infection and sensitivity to contrast medium. In distinct cases in which it is not possible to distinguish between different causes of obstruction, when the duct is filled with air,[2,12] there may be misleading positive results. Ultrasound revealed calculi over 1.5 mm in size in nearly all cases in an experimental study,[11,13,14] but low mineralized or early-stage stones may not be detected.[2,6,10] Often, only indirect signs of obstruction with duct dilation or changes in the echogenicity of the gland parenchyma can be identified.[2,9,10] Other diagnostic tools that also indirectly visualize salivary gland diseases, such as CT-scan, MRI, MR-sialography or scintigraphy, are known to be expensive, time-consuming procedures with no advantages to be gained in respect of sensitivity or specifity in those cases; in the case of CT-scan, the risk of x-ray exposure should also be mentioned.[2]

In our experience, in up to 5% of all cases where the swelling of the major salivary glands was present, possible causes remain unclear. This diagnostic gap has been filled with the introduction of sialoscopy, which is able to directly visualize the underlying pathological condition.[9,10,15,16] After implementation in the early 90s,[15] sialoscopy rapidly developed into an important diagnostic and therapeutic technique in salivary gland disorders, especially in sialolithiasis.[3-6,9,10,16-20] In recent years, the development of smaller, semi-rigid sialoscopes, with high image quality, considerably improved both the diagnostic and particularly

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the therapeutic possibilities. Nowadays semi-rigid sialoscopes with a diameter below 1.5 mm and with 2 working channels are considered state of the art.[3-6,19-22]

In our department, ultrasound is the diagnostic examination of first choice. Between January 2001 and December 2003 over 2200 patients visited our department with diseases of the major salivary glands. Excluding patients with neoplastic disease, who amounted to 10% to 15% of the total, over 600 patients with inflammatory, non-neoplastic disease were treated per year. All patients were examined with ultrasound. In the cases of those with inconclusive diagnosis, ultrasound was supplemented with sialoscopy. Sialoscopy was performed in the case of 103 patients due to recurrent swelling of the major salivary glands of uncertain origin. This retrospective clinical study aims to describe our 3-year experience using sialoscopy in the diagnostic evaluation and therapy of these patients.

PATIENTS

This study was conducted in accordance with the guidelines of the Ethics Committees of the University of Erlangen-Nuremberg. All patients were informed of the possible discomfort and risks of the surgical treatment, and signed consent was collected prior to therapy. From 2001 to 2003 over 1900 patients were evaluated because of swelling of and/or pain in the major salivary glands, which was not caused by neoplasm. Ultrasound was performed in all patients (Sonoline Elegra, 7.5 MHz, Siemens Medical Solution, Germany). In 103 cases, adequate diagnosis was unable to be assessed. In these cases, a total of 109 sialoscopies were performed.

Prior symptoms had first appeared as recently as 3 weeks before and as long ago as 10 years before. Ages ranged from 16 to 82 years (average, 49.5; median, 49). Sex distribution was nearly equal with 52.4% female (54 of 103) and 47.6% male patients. In 51.5% (53 of 103) of the patients, sialoscopy of Warthon′s duct was performed, and in 48.5% (50 of 103) of patients, sialoscopy of Stensen′s duct was performed. Three patients had bilateral gland evaluation (2 submandibular, 1 parotideal). One patient had 2 and another 3 sialoscopies of the parotid gland due to recurrent stenoses.

SIALOSCOPY

All instruments used in this study are commercially available and certified medical products. Previous published data described a feasibility of all instruments used.[19] As favored nowadays by the most centers,[3-6,19-21] we used 2 different types of semi-rigid endoscopes. The first sialoscope had a diameter of 1.1.mm and 2 working channels, 1 for irrigation and 1 to introduce the instruments (0.4 mm). The optical resolution was 6000 pixels (Polydiagnost, Type PD ZS 2000). The second endoscope was coated in nitinol and therefore was extremely flexible (Polydiagnost Salivascope flex, Type PD ZS 2001, Fig 1). The instruments that can be inserted through the working channel had a diameter of 380-390 μm (basket, grasp, drill, and laser fiber). Published technical data has been described elsewhere.[19]

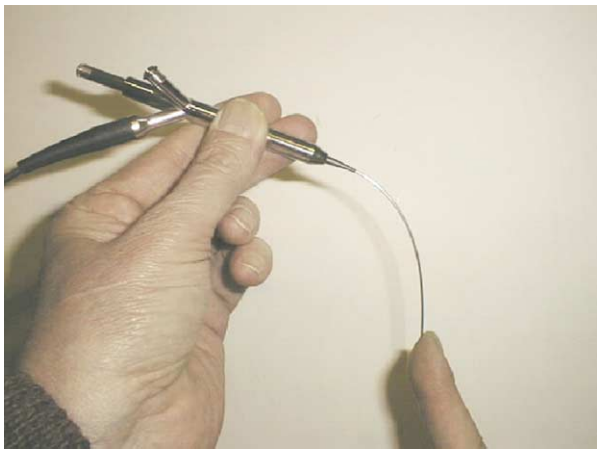

Figure 1 Sialoscope (Polydiagnost Salivascop flex, type PD ZS 2001).

After local anesthesia of the mucosa with lidocain spray (4%) and dilation of the papilla with custom-made conical shaped dilator, intraductal instillation of about 1 to 2 mL lidocain 2% with epinephrine 1:100000 via a venous cannula (22GA, 0.9 mm) was performed. After insertion of the sialoscope, dilation of the duct was achieved through continuous rinsing with solution of NaCl 0.9% or isotonic electrolyte-solution. Thus the ducts of the second and third order could be reached with the sialoscope. Interventional sialoscopy was performed under continual rinsing using various instruments (drill, grasp, or basket), and where necessary, in the case of endoscopic stone removal, in combination with a mini-papillotomy.

Postoperatively routine antibiotic and antiphlogistic medication was administered (ampicillin/sulbactam 2 × 1g/d, serrapeptase 10 mg 3 × 2/d for 3 weeks, piroxicame 2 × 20 mg/d for 3 days). Gland massage and sialagoga were recommended.

RESULTS

All sialoscopies were performed with the patient given a local anesthesia, and the procedure was well tolerated by the patients. The time taken for the diagnostic procedures ranged between 15 to 20 minutes, and between 15 to 45 minutes for interventional sialoscopies.

Insertion of the endoscope was successful in all but 3 procedures. In these 3 (3 of 103, 2.9%) cases, papillotomy had to be carried out due to a too narrow ostium (all submandibular gland).

Findings in Sialoscopy

Normal findings were noted in 10.7% (11 of 103) of all cases; pathological findings were noted in 89.3% (92 of

Table 1
Results of diagnostic evaluation and treatment of obstructive diseases of the salivary glands of unclear origin in 103 patients

	Parotid ducts		Submandibular ducts	
	n	%	n	%
∑	50	48.5	53	51.5
Normal findings	2	1.9	9	8.7
Sialolithiasis	11*	10.7	10	9.7
Stenosis/foreign bodies	28*	27.2	30	29.1
Sialodochitis	8*	7.8	2	1.9
Variation/malformation	2*	1.9	2	1.9
Gland preserving therapy	27	26.2	30	29.1
Interventional sialoscopies	28	27.2	13	12.6
Gland resection	1	0.9	1	0.9

*Combined pathology in 1 salivary gland each.

103) of the patients (Table 1). Anatomic variations of the ducts (such as kinking or accessory ducts) were thought to be the reason for unclear gland swelling in 4 (3.9%) cases. Sialolithiasis was diagnosed in 20.3% (21 of 103) of the patients, 18.9% (10 of 53) of submandibular and 22% (11 of 50) of parotid gland. All but one sialolith had a smooth consistency due to low mineralization (Fig 2). One of these patients showed combined stenosis and sialolithiasis of Stensen′s duct (Fig 3). The diameter of the calculi was between 3 and 5 mm in submandibular (average, 3.85 mm) and 2.5 and 5 mm (average, 3.85 mm) in parotid glands. This may be one important factor, as to why sialoliths were not clearly detectable by ultrasound (Fig 3).

Obstruction due to inflammatory fibrotic stenoses or due to a foreign body or fibrotic plugs was seen in 56.3% (58 of 103) of all patients, and obstruction affecting the parotid ducts was seen in 56% (28 of 50) of all patients (Fig 3). Submandibular duct stenosis was diagnosed in 56.6% (30 of 53) of cases. It should be noted that 14 of these 58 patients had a history of sialolithiasis. In 1 case, obstruction was caused through a fibrinous plug combined with an inflammatory stenosis whereas in another case, a hair could be found in Warthon′s duct (Fig 4). Sialodochitis, as the single cause of obstruction, was found in 10.7% (11 of 103) of all patients, and in over 80% (9 of 11) of these cases the parotideal gland was affected (Fig 5). One of these patients showed an infection of a blind ending in Stensen′s duct after prior parotidectomy, and another patient had had prior ESWL.

Therapeutic Consequences of Sialoscopy

All patients, who showed pathologic findings, were given conservative treatment, which consisted of antiphlogistic and antibiotic medication (as described earlier), sialagoga and gland massage. After extended treatment, dilation of stenoses or, if chronic inflammatory disease or autoimmune cause was suspected, a single dose of hydrocortisone (50 mg) was applied into the duct.

Overall, in 35% (36 of 103) of all cases or 37.6% of all sialoscopies, interventional therapy was performed (41 of 109). Bilateral therapy was performed in 2 cases (first patient, submandibular stenosis of 1 side and sialolithiasis of the other; second patient, bilateral stenosis of Stensen′s duct). 86.1% (31 of 36) of the patients were treated successfully with interventional sialoscopy and showed no more symptoms.

Therapy of sialolithiasis. Removal of sialoliths with interventional sialoscopy was achieved by extraction, using basket or grasp in 52.4% (11/21: in 5 submandibular and in 6 parotid gland cases) of cases. When stones could not be treated successfully by interventional sialoscopy, they were either impacted or not accessible by the sialoscope. Partial or extended duct slitting was performed in 5 cases of submandibular stones. Extracorporeal shockwave therapy (ESWL) was carried out in 6 cases of parotid gland calculi (including 1 case of combined stenosis, where opening was required before performing ESWL).

Treatment of obstructions. Drill, grasp, basket, and the sialoscope itself were used in interventional sialoscopy to dilate stenoses or remove foreign bodies or obstructive

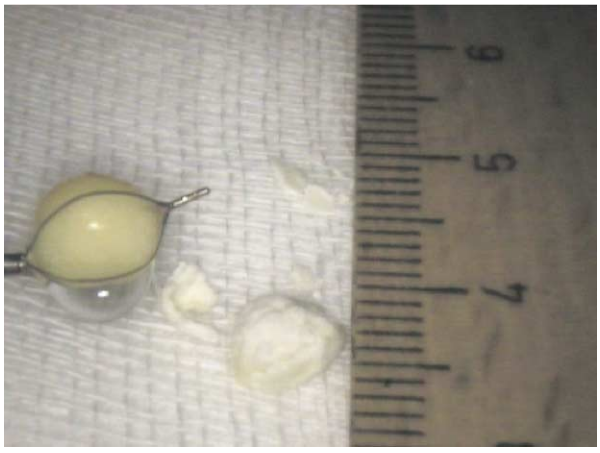

Figure 2 Stones with high (in the basket) and low mineralization. The latter was not detectable by ultrasound.

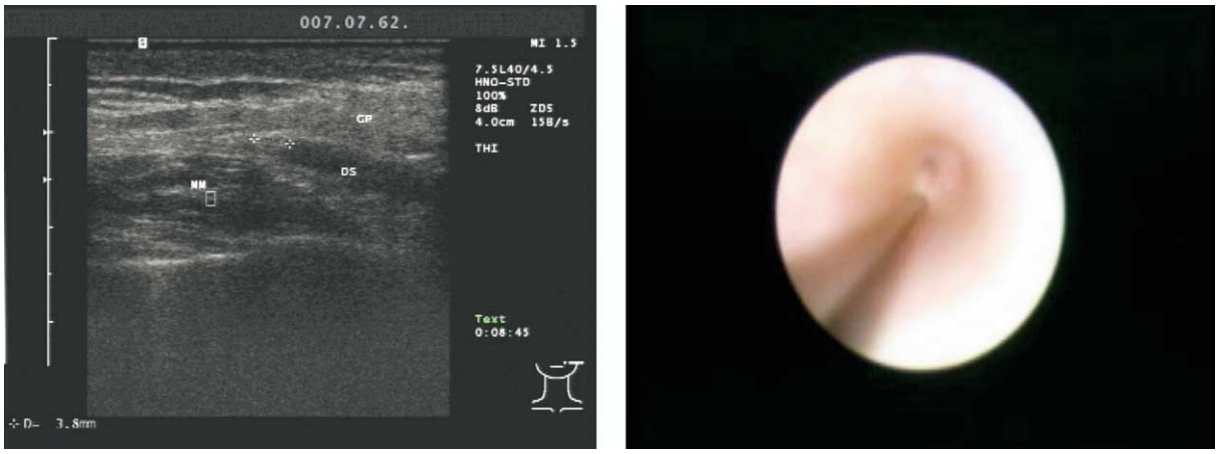

Figure 3 A, Ultrasound shows duct dilation and unclear formation region of obstruction. Differentiation between sialolith and stenosis was not possible. B, Sialoscopy revealed stenosis distal to a sialolith that was dilated by basket. The sialolith was treated with extracorporeal shock wave lithotripsy.

plugs in 41.4% (24 of 58, in 7 submandibular and in 17 parotid glands) of cases. Partial or extended duct reconstruction or slitting was performed in 15 cases (in 14 submandibular and in 1 parotid gland). In 2 cases of parotid duct stenosis, endoscopic guided stent implantation was performed. Sialodochitis was treated in 1 case by interventional sialoscopy, through irrigation of a blind ending in Stensen's duct, which was then filled with detritus after parotidectomy.

Altogether, gland preserving treatment was performed in 55.3% (57 of 103) of all cases (in 27 parotid gland and in 30 submandibular gland cases). Gland extirpation was necessary in only 1.9% of all cases (2 of 103). One patient showed diffuse stenosis of Warthon′s duct whereas another patient desired resection due to of therapy resistant sialodochitis of parotid gland with obstructive symptoms enduring over a 10-year period.

In summary, 89.3% (93 of 103) of the patients reported no further symptoms, postsialoscopy, or therapeutic intervention. The procedure of diagnostic and interventional sialoscopy was well tolerated by all patients. Postoperative pain sensation and gland swelling due to irrigation, was observed for a period of 2 to 3 hours. In 1 (0.9%) case of distal duct stenosis, perforation of the Warthon′s duct occurred while introducing the endoscope. However, no other major complications were observed.

DISCUSSION

In more than 60% of obstructive non-neoplastic disorders of the major salivary glands, sialolithiasis is found to be the most common cause.[1-4] In some patients with swelling of major salivary glands, diagnosis cannot be made by conventional radiological means or even with high-resolution

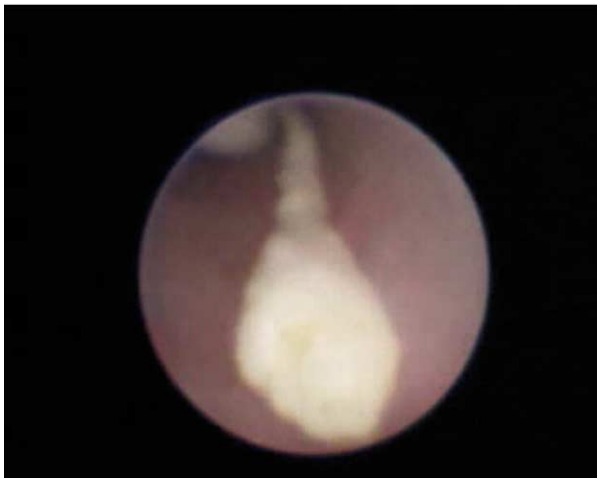

Figure 4 Obstruction of Warthon′s duct by a hair that developed superficial mineralization.

Figure 5 Sialodochitis as the cause of obstructive gland disease.

ultrasound.[1,2,11] This was the case in 5.4% of our patients with non-neoplastic inflammatory disease. These problems are well known in patients with sialolithiasis or other obstructive diseases. This is especially true in stenoses, which may not be readily distinguished from sialolithiasis. Sialoliths with a smooth consistency can often not be differentiated from fibrinous plugs, fibrotic changes, such as stenosis, chronic sialadenitis, calcifications in the gland or lymph nodes, and plugs in blood vessels. Moreover, obstructive diseases can be caused by other conditions, such as organic foreign bodies, intraductal tumors or anatomic variations/malformations.[2,6,9,10] Many of the aforementioned conditions of salivary glands can be directly visualized through sialoscopy, and it has, therefore, gained importance and now appears to have developed into a routine diagnostic tool for this purpose.[3-6,9,10,15,19,20] The technical advances and improvements in new endoscopes with respect to size, optical and material features had consequently led to a widened spectrum of indication for diagnostic and interventional sialoscopy.[3,4,6,19-21]

Sialolithiasis, which is widely presumed to be the most important cause of obstructive gland disease,[1,3,4] was found to be either the primary or secondary cause of gland swelling of unclear origin in 33% of all patients. In these patients, calculi, which had a smooth consistency and could not be clearly defined by conventional diagnostic means (Fig 2), were now observable by sialoscopy. Moreover, patients' histories of sialolithiasis in the past were positive in 13.6% of cases with duct stenosis.

Obstruction due to inflammatory or fibrotic stenosis or foreign bodies was the most commonly observed pathological finding and occurred in 56.3% of the patients. In a few cases, cylindrical shaped mucous or fibrinous plugs of various sizes (Fig 6) were observed and removed. Mucous plugs or other organic matrix are suspected to be involved in the development of sialoliths.[1,2] Intraductal fibrinous plugs, which can be identified through sialoscopy, may be regarded as organic matrix before mineralization and stone formation. In further cases, stenosis combined with fibrinous plugs and, in 1 patient, stenosis combined with proximal sialolithiasis, was observed. This points in the same direction. This theory is also supported by observation of a superficial, mineralized, organic foreign intraductal body (Fig 4). Thus, sialoscopy may be able to support efforts in explaining the stages of stone formation. Beyond that, it may also be useful in establishing early diagnosis and prophylaxis of sialolith formation through repeated irrigation of the duct or removal of mucous or fibrinous plugs.

Figure 6 Fibrinous plug causes obstruction in a parotid duct. (Hematoxylin and eosin stain; original magnification × 40.)

Figure 7 Thickening of the epithelial lining in the main and in second and third generation ducts of the parotid gland.

In a few cases, remarkable diffuse ductal changes with signs of thickened epithelial layer, which also included nonstenotic regions, could be observed (Fig 7). Autoimmune disease, as a cause of the obstruction, has to be excluded in these cases. If no underlying immunologic disease could be found, existence of a disease limited to the gland should be considered. In 1 case, in which Sjögren's syndrome was excluded, the female patient continued to have repeated exacerbations of obstructive parotideal gland disease due to diffuse duct stenosis. Analysis of a sample of gland secretion showed a marked number of eosinophilic cells (Fig 8). This finding may point to isolated gland disease dominated by eosinophilic infiltration.

In addition to this, anatomic variations or malformations (such as kinking or bending of the duct) as well as other rare conditions (for example, pneumoparotitis) or postoperative complications (such as blind sack infection in the residual duct after parotidectomy) and their impact on gland morphology and function may be better assessed by sialoscopy and appropriate therapy, therefore, planned.

In 55.3% of all cases, sialoscopy was followed by an operative but gland-preserving therapy. Interventional sialoscopy was performed in 37.6% of all sialoscopies. These facts show that sialoscopy, in the treatment of disorders of the major salivary glands of unclear origin, is of importance as both a diagnostic and a therapeutic tool.

Our therapeutic success rates (89.3% of the patients free from further complaint) compare well with other results.[3,4,6,20] Beyond this, they underline the importance of

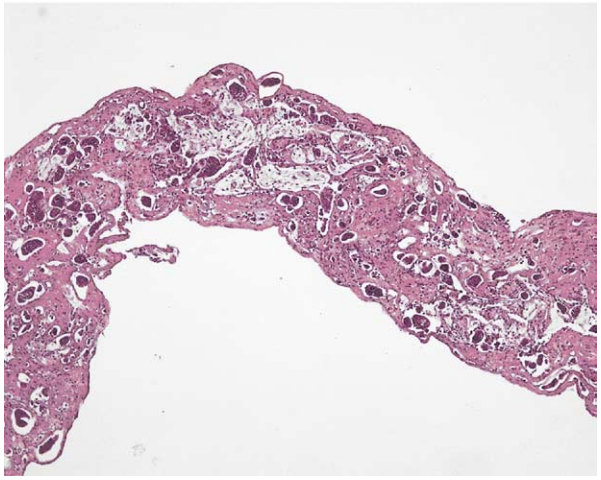

Figure 8 Sample of secretion, dominated by eosinophilic cells, of a patient who had repeated episodes of obstruction of the parotid gland and showed diffuse stenosis of Stensen′s duct in sialoscopy. (Hematoxylin and eosin stain; original magnification × 40.)

the concept of embedding minimal invasive techniques in a gland-preserving therapeutic regimen, especially when one considers the low rate of gland resection that was found necessary in only 1.9% of our patients.

Sialoscopy has been proven to be a safe procedure, and it has been well tolerated by all patients. The low rate of complications, 0.9%, was comparable with rates given in other reports.[3-6,20]

In the evaluation of diseases and disorders of unclear origin, sialoscopy proved to be a safe and effective procedure and able to fill the gap in conventional diagnostic means. In the concept of gland preserving therapy, sialoscopy has become an important, routine diagnostic tool. When focusing on the specific conditions of unclear swelling of the major salivary glands in of our patient population, the existing spectrum of indication owing to sialoscopy[3-6,20] can be confirmed and extended: (1) detection of occult sialoliths; (2) detection of early formation of sialoliths (mucous or fibrinous plugs) and prophylaxis of stone formation; (3) treatment of postinflammatory stenoses and other obstructive conditions; (4) detection and therapy of anatomic variations or malformations; (5) diagnosis and new insights into causes of autoimmune disorders that may involve salivary glands leading eventually to therapeutic consequences; and (6) follow-up and control of therapy success rates (therapy of sialolithiasis or stenosis, etc).

Beyond this, the low rate of gland resection in our patient cohort underlines the role to be played by sialoscopy as a therapeutic tool, which should now be routinely included in the concept of gland preserving treatment.

The further development of sialoscopic techniques and the publication of experience with sialoscopy will improve the diagnostic and therapeutic possibilities and widen the spectrum of applications.

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(References that are classified as Grade D were mainly experimental in character and related to the testing of new diagnostic and therapeutic tools.)