Sialendoscopy-Based Diagnosis and Classification of Parotid Duct Stenoses

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Objectives/Hypothesis: To assess stenoses of Stensen's duct considering findings provided by sialendoscopy-based direct visualization. A practical classification scheme based on different parameters is presented.

Methods: Ultrasonography and sialendoscopy were used to diagnose and analyze symptomatic stenoses of the parotid gland excretory duct in 93 patients. Sialendoscopy was performed with semirigid endoscopes that enabled direct evaluation and classification of those stenoses in terms of tissue quality, luminal narrowing, extend, number, location, and in consideration of possible etiologic diseases or conditions.

Results: A total of 111 stenoses were diagnosed in 93 patients. Three types could be distinguished. Stenoses were inflammation-dominated with various narrowing of the lumen (type I) in 16.1% of patients, fibrous and web-associated, predominant incomplete (luminal narrowing <50%, type II) in 18.3%, and fibrous, predominant high-grade (luminal narrowing >50%, type III) in 66.6%. A length of more than 1 cm or diffuse involvement of the duct system was observed in 12.9% of cases. Together 70.1% of all stenoses were found in middle and distal regions of the duct. Multiple stenoses were found in 12.9% of patients, bilateral in 6.5%. Of all patients, 45.2% had diseases possibly implicated in the etiogenesis. Type III stenoses were associated with these comorbidities in up to 100%.

Conclusions: Minimally invasive techniques play a central role in the diagnosis and treatment of Stensen's duct stenoses. Sialendoscopy is the diagnostic method of first choice. It enables an exact and direct classification of stenoses and provides additional information for planning effective treatment.

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INTRODUCTION

Stenoses of Stensen's duct are present in approximately 15% to 25% of salivary gland obstructions involving the parotid gland. This was shown in a large retrospective study that reviewed reports of over 1,000 sialograms undertaken to diagnose salivary obstruction.[1] In one study with selected cases of salivary gland swelling of unclear origin in which sialendoscopy had to be used for diagnosis, the frequency was even greater than 50%.[2]

Stenoses cause painful swellings of the parotid gland after or during eating and concomitant autonomic symptoms, which may result in a marked reduction in the quality of life.[3]

In the majority of cases, the cause of these stenoses is unclear.[1,4] Chronic parotitis, as well as allergic, infectious, granulomatous, or autoimmune diseases, and radiotherapy or radioiodine therapy, have been discussed.[1,4,5]

Ultrasound, sialography, and sialendoscopy play an important role in diagnosis and can contribute to a more precise characterization of stenoses. Sonography provides an overview of the entire ductal system. It enables stenoses to be located and shows the area of prestenotic dilatation. Typical sonographic findings with a stenosis are a hypoechoic band on the masseter muscle as evidence of ductal dilatation and additional hypoechoic parenchymal changes as signs of gland obstruction. Sialography provides precise information as to the site, number, and extent of stenoses. Typical findings are the narrowing of the duct lumen and/or the absence or discontinuation of contrast medium in the ductal system.[1,5,6] However, the detection of prestenotic areas or less marked stenoses is not so reliable, and no statements can be made about the nature of the stenosis. The development of sialendoscopy has enabled direct visualization of the ductal system and thus a more precise diagnosis can be made of unclear swellings of the salivary glands, particularly in the case of stenoses.[2,4–9]

Only a few authors have reported a more detailed characterization of stenoses in terms of structural changes of the ductal epithelium, and their location and extent in the ductal system.[1,4,7] Ngu et al. described the number and location of stenoses diagnosed by sialography.[1] Marchal et al. proposed a classification in terms of sialendoscopy-based location, which took account of the site, number, and severity of stenoses.[6] A sialendoscopy study in a smaller group of patients found marked differences in tissue appearance in the stenosis region itself.[4] This criterion has not received adequate consideration in any study to date.

A retrospective review was therefore carried out of sialendoscopy results in 93 patients with Stensen's duct stenosis, in which their tissue quality in the stenotic region, the narrowing of the ductal lumen, and the extent, number, and location were characterized. This paper presents a classification based on the inclusion of all of the previously mentioned factors.

MATERIALS AND METHODS

On average, approximately 150 sialendoscopies are carried out annually in the Department of Otorhinolaryngology, Head and Neck Surgery of the Friedrich Alexander University, Erlangen-Nuremburg. Between 2001 and 2006, in 93 patients who presented with recurrent painful swellings of the parotid gland after or during eating, stenoses of Stensen's duct were diagnosed. Of the patients, 43% were men (40/93). The average age was 49 years (range, 15–75; median, 49 years).

All patients were examined using high-resolution sonography (Sonoline Elegra, 7.5 MHz, Siemens Medical Solutions, Inc., Malvern, PA). Sialendoscopy was performed after a tentative diagnosis had been made from a clinical examination and ultrasound. In addition to direct visualization, pretherapeutic assessment of stenoses was also possible. We used a variety of semirigid sialendoscopes.[4,10] Since 2002, in collaboration with Karl Storz GmbH & Co. KG (Tuttlingen, Germany) we have used newly developed prototypes, which from 2004 onwards formed the endoscopy set. This consists of three sialendoscopes with an external diameter of 0.8 to 1.6 mm. In addition to an irrigation channel (0.25 mm), an additional working channel (0.45 or 0.8 mm) is available for interventional treatment.[2] The shaft of the endoscope is marked in centimeters to enable better positioning in the ductal system (Fig. 1).

Fig. 1. Centimeter marking on the shaft of the sialendoscope for exact measurement of stenosis length. It belongs to the Erlanger sialendoscopy set with one diagnostic and two interventional endoscopes (Karl Storz GmbH & Co. KG, Tuttlingen, Germany). [Color figure can be viewed in the online issue, which is available at www.interscience.wiley.com.]

Tissue Characteristics

The stenoses were assessed in terms of macroscopic differences in tissue appearance that had been found during sialendoscopy in a smaller group of patients.[4] The stenoses of the current larger group of patients, which included our smaller series, were evaluated in terms of such criteria. Tissue changes in the stenotic area and in the remaining duct system were assessed under direct endoscopic view. The main criteria for distinguishing the appearance in the stenotic area were the presence of dominant inflammatory or fibrotic changes. The remaining duct system and obviously not involved duct areas were assessed in terms of any changes of the epithelium and the consistency of the duct wall.

Luminal Narrowing

Luminal narrowing was estimated using published anatomical data on the diameter of Stensen's duct (average, 1.5 mm) as reference.[11] An exact estimate of the residual lumen was based on the shaft diameter of the sialendoscope used (0.8, 1.1, or 1.6 mm).[2] Stenoses were described as low to medium grade if these were still passable with the 0.8 mm sialendoscope. If the stenosis could only be passed after an additional instrumental dilatation, they were described as high grade.

Extend

Stenoses were distinguished based on their extent as short, segmental stenoses (membranous stenosis, pinhole stenosis, length $\leq$1 cm), long, segmental stenoses (length >1–3 cm), and diffuse stenoses (>3 cm, including several parts of the main ductal system, main duct, and hilum region).

Number

The number of stenoses was recorded as single or multiple (two or more).

Location

An exact localization was possible based on the anatomical landmarks detectable during sialendoscopy and the centimeter marking of the sialendoscope. The main duct was divided into four segments: the papilla region, the distal ductal system (from approximately 0.5 cm proximal of the papilla up to approximately 1 cm before passage through the buccinator muscle), the mid-ductal system (buccinator area as far as approximately 2 cm behind the buccinator region), and the hilum region (from approximately 1 cm before the hilum up to the posthilar duct system).

Association With Operative Manipulation and/or Diseases

Patients records were reviewed to associate diseases and/or preceding operative procedures, which may be a possible cause for the development of the stenosis.

RESULTS

A total of 111 stenoses in 99 glands were diagnosed in our 93 patients. In 86.1% of all cases there was one isolated stenosis at the time, and 13.9% of these were combined with a sialolithiasis (13/93).

TABLE I.
Characteristics of the Tissue Reactions Typical in Different Stenoses.

Characteristic	Type I	Type II	Type III
Appearance in stenotic region	Marked inflammation, slight fibrotic remodeling	Fibrotic stenosis, often short segment	Fibrotic stenosis, massive fibrotic reaction of the duct wall
Luminal narrowing	Variable	slight to moderate, incomplete (<50%)	High grade (>50%)
Associated ductal changes	Obstructive plaques	Circular or web-like encroachments of the duct wall, megaduct	Duct wall segmental or diffuse with massive fibrotic reaction
Number of patients (%)	n = 15 (16.1)	n = 17 (18.3)	n = 61 (65.6)

Tissue Characteristics Appearance of Stenoses and Luminal Narrowing

Irrespective of the stenosed region, in all cases sialendoscopy showed marked local or diffuse changes of the ductal system that were not limited to stenotic regions. These changes regularly consisted of a thickening of the ductal epithelium with mucous and/or fibrinous exudates.

Three different types of stenosis in terms of their macroscopic nature and severity were seen in our group of patients (Table I). The first type showed, in addition to an incipient fibrotic remodeling, an inflammatory reaction with epithelial edema and marked fibrinous exudates into the ductal lumen, which dominated the overall picture. These patients often also had obstructive fibrin plaques in addition to the acute inflammation-accompanying reaction of the ductal system (Fig. 2A). The luminal narrowing was variable (majority, slight to moderate; high-grade, four cases). This type of stenosis (type I stenosis dominated by inflammation) was detectable in 16.1% of all patients (15/93) (Fig. 2B).

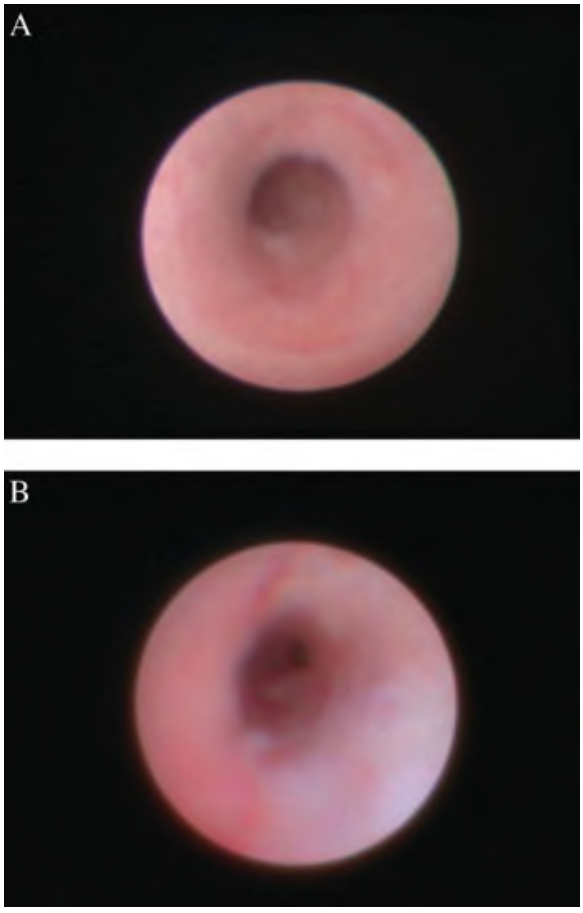

Fig. 2. Inflammatory segmental duct thickening in subacute sialodochitis as possible precursor for a predominantly inflammatory or fibrous stenosis (A). Stenosis with inflammation-dominated reaction (type I) (B).

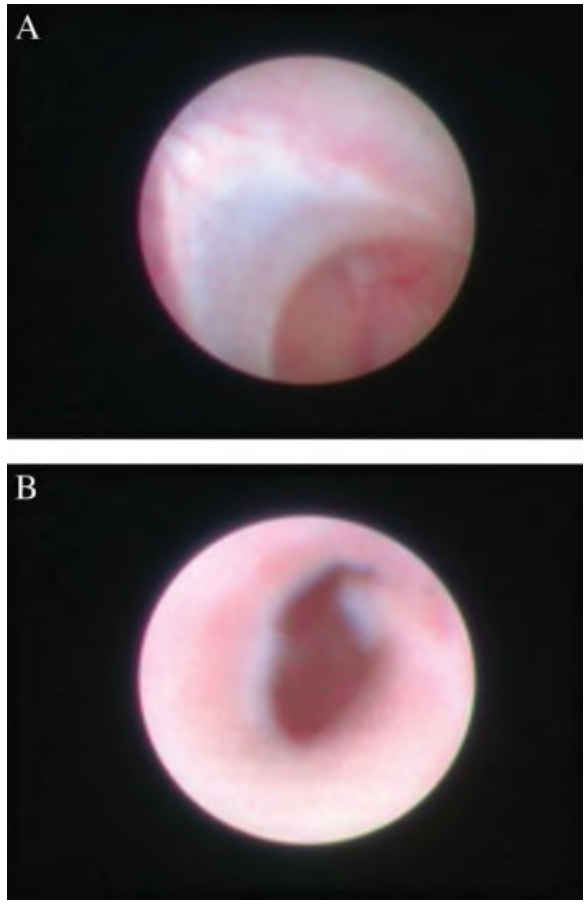

Fig. 3. Web-like encroachment on the ductal system as possible precursor of a membranous or short segment fibrous stenosis (A). Moderately severe fibrous stenosis through short segment ductal encroachments that were passable after dilatation with the endoscope (type II) (B).

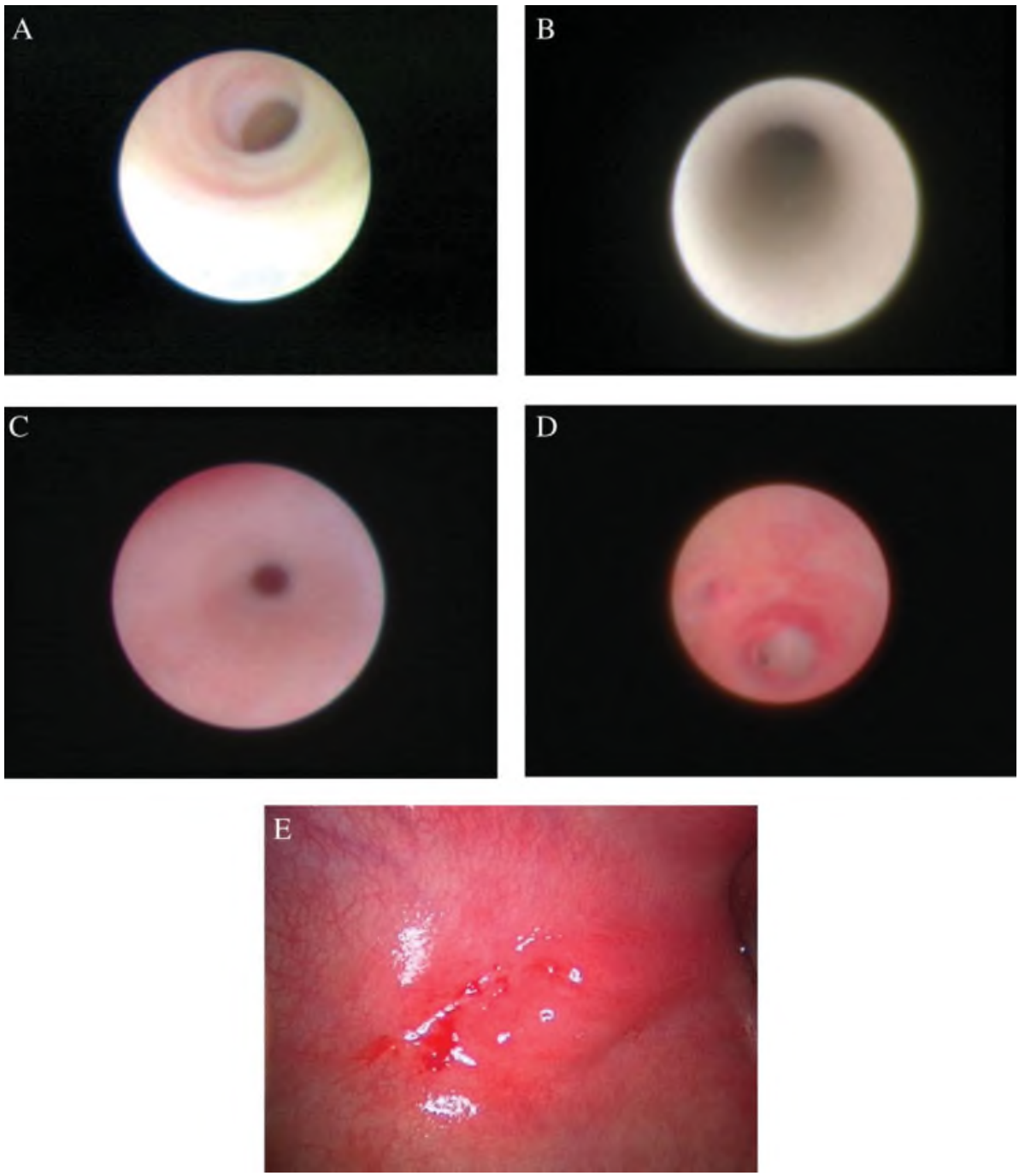

Fig. 4. Segmental (A) and diffuse (B) ductal fibrosis as possible precursor of a short or long segment severe fibrous stenosis. Filiform fibrous stenosis that required additional instrumental, endoscope-controlled dilatation (type III) (C). Stenosis with almost complete obliteration (type III) (D). Scarred papillary stenosis with complete displacement of the ostium after previous surgical manipulation (type III) (E).

The second type was frequently associated with circular and especially sickle-shaped web-like encroachments of the remaining ductal system, with an almost normal appearance of the epithelium between the encroachments (Fig. 3A). Almost invariably, these stenoses showed an incomplete luminal narrowing of up to approximately 50%. In some cases a massive duct dilation was visible, which resulted in the formation of a megaduct. In these cases the duct wall appeared to be thin between the encroachments. This type of fibrous stenosis (Fig. 3B) was present in 18.3% of patients (17/93, fibrous, web-associated, typically low- or medium-grade stenosis type II).

The third type showed fibrous, sometimes conically tapering areas, which almost always resulted in high-grade luminal narrowing of >50%, and was associated with a massive fibrotic thickening of the ductal wall. In

TABLE II.
Stenoses of Stensen's Duct: Extend and Number in the Different Types of Stenoses.

	Inflammation-Dominated (Type I)	Fibrous, Web-Associated (Type II)	Fibrous (Type III)
Patients, n = 93	16.1% (15/93)	18.3% (17/93)	65.6% (61/93)
Extend/length			
≤1 cm	73.3% (11/15)	82.4% (14/17)	91.8% (56/61)
>1–3 cm	26.7% (4/15)	5.9% (1/17)*	4.9% (3/61)†
Diffuse	0% (0/15)		
		11.7% (2/17)	3.3% (2/61)
Number			
n = 1	93.3% (14/15)	82.4% (14/17)	86.9% (53/61)
n ≥ 2	6.7% (1/15)	17.6% (3/17)	13.1% (8/61)
Bilateral (n = 1)	0% (0/15)	17.6% (3/17)	4.9% (3/61)

*One patient with bilateral stenosis; 1 side ≤1 cm.
†One patient with bilateral stenosis each side; >1–3 cm.

these cases there was also often a segmental or diffuse pallor of the entire ductal epithelium (Fig. 4A, B). These ductal changes typically were associated with filiform or complete obstruction of the duct (Fig. 4C, D) or papillary stenoses (Fig. 4E), and were generally only passable after additional instrumental measures (65.6%, 61/93, typically high-grade stenosis type III).

Extent and Number of the Different Types of Stenoses

Of the stenoses, 73.3% to 91.8% had a length <1 cm. A length of 1 to 3 cm was observed in 8.6% of all stenoses, but in 26.7% of the type I stenoses. In 4.3% the ductal system was diffusely stenosed. Depending of the type, between 82.4% and 93.3% of the stenoses were singular. Of the patients with type II stenosis, 17.6% had multiple and/or bilateral findings (further data, Table II).

Location of the Different Types of Stenoses

Of the total of 111 stenoses, 9.1% (10/111) were located at the papilla, 36.9% (41/111) in the distal, 34.2% (38/111) in the middle, and 19.8% (22/111) in the hilum region. Several stenoses in the same duct system were seen in 12.9% (12/93) of patients (5.4% in the middle and distal parts of the duct, 6.5% near the hilum and in the middle of the duct, and in one patient combined papillary and distal stenosis). In 6.5% of patients (6/93) a bilateral stenosis was found (5 cases, same location: 2 distal and 3 hilum; 1 case, distal and middle part). Of type I stenosis, 87.7% were in the middle and distal region, and 96% of type II were in the distal and middle region and near the hilum. Of all type III stenosis, 74.6% were found in the middle and distal region. Eighty percent of papillary stenosis and 70.7% of those in the distal duct were type III (further data, Table III).

Association With Possible Causative Diseases

In 45.2% of patients (42/93), the stenoses were associated with diseases that might also be their cause (Table IV).[1,4,5] Sialolithiasis was present in 25.8% of patients (24/93), and in one third of these (8/24) surgical manipulations on the ductal system were therefore carried out. Of all of the patients, 11.8% had an allergic disease (11/93), 5.4% an autoimmune disease (5/93), and 4.3% a bruxism with discernible damage to the cheek tissue (4/93). A lateral parotidectomy was undertaken previously in 3.2% of patients (3/93) and radiotherapy in 1.1% (1/93). Type III stenoses were associated with all concurrent diseases with a frequency of 50% to 100% (further data, Table IV).

All the parameters had been included in a classification of parotid stenoses (Fig. 5).

DISCUSSION

Obstructive disorders of the parotid gland cause recurrent painful swellings of the main salivary glands. Stenoses of the excretory duct of the parotid gland are the second commonest cause and are present in at least

TABLE III.
Stenoses of Stensen's Duct: Type of Stenosis By Location.

	Inflammation-Dominated (Type I)	Fibrous Web-Associated (Type II)	Fibrous (Type III)
Number of stenoses*	n = 16	n = 24	n = 71
Location (4 segments)			
Papilla, n = 10	6.3% (1/16)	4.2% (1/24)	11.3% (8/71)
Distal, n = 41	37.7% (6/16)	25% (6/24)	40.8% (29/71)
Middle region, n = 38	50% (8/16)	45,8% (11/24)	33.8% (24/71)
Proximal duct/hilum, n = 22	6.3% (1/16)	25% (6/24)	14.1% (10/71)

*Total of 93 patients, 111 stenoses.

TABLE IV.
Stenoses of Stensen's Duct: Stenosis Type in Association With Pretreatment and/or Associated Diseases.

Pretreatment/Associated Disease	Inflammation-Dominated (Type I)	Fibrous Web-Associated (Type II)	Fibrous (Type III)
Sialolithiasis, n = 24			
Papillary surgery, n = 8	12.5% (1/8)		87.5% (7/8)
Abscess incision, n = 1			100% (1/1)
ESWL, n = 15	20% (3/15)	13.3% (2/15)	67% (10/15)
No therapy, n = 1			100% (1/1)
Parotidectomy, n = 3			100% (3/3)
Autoimmune disease, n = 5		40% (2/5)	60% (3/5)
Allergy, n = 11	27.3% (3/11)	18.2% (2/11)	54.5% (6/11)
Bruxism, n = 4	25% (1/4)	25% (1/4)	50% (2/4)
Radiotherapy, n = 1			100% (1/1)

Numbers represent 38 out of 93 patients, seven patients with multiple diseases.
ESWL = extracorporal shock wave lithotripsy.

20% of cases. They present a challenge in everyday practice.[1,2] Ultrasound, sialography, and sialendoscopy are currently the most important diagnostic techniques, all of which can contribute to a more precise characterization of stenoses.[1,2,4–6]

In the largest evaluation of sialograms to date, the number and location of 150 findings of Stensen's duct stenoses were investigated. To classify the stenosis sites, the ductal system was divided into an anterior, middle, and posterior third including the hilum. Single stenoses of Stensen's duct were most frequently found (41%) in the middle third of the duct. Multiple stenoses were present in 30% of cases, and these were found in the middle and posterior thirds of the duct. Bilateral stenoses were diagnosed in 16.1% of cases.[1] Because the diagnosis was made by sialography, no direct

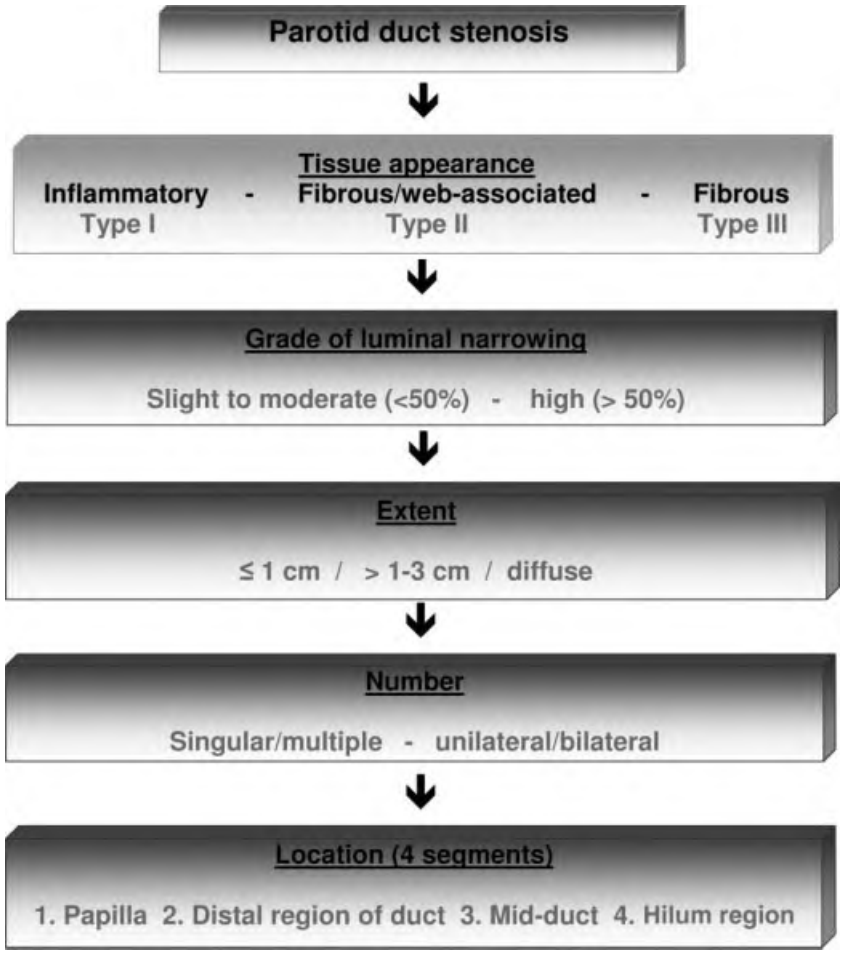

Fig. 5. Sialendoscopy-based classification of Stensen's duct stenoses.

visualization of the stenoses was possible. However, this is essential for an exact assessment of the tissue conditions in the stenosis area, the resulting luminal narrowing, and the length of a stenosis. A direct visualization also allows an assessment of the entire remaining ductal system, including possible precursor stages such as segmental or diffuse inflammatory or fibrous changes of the ductal epithelium, or the formation of web-like ductal encroachments (Figs. 2A, 3A, and 4A, B).

Sialendoscopy enables a more detailed and direct assessment of stenoses concerning all these parameters and offers the possibility of conducting effective treatment at the same time.

Several groups describe stenoses of the excretory duct as the cause of obstructive salivary gland disorders in adults and children,[2,4–9] but only a few papers classify the stenoses in more detail. Our own group and that of Qi et al. described inflammatory changes in the duct that might be a possible precursor stage to stenosis and were indeed observed in our group of patients (Fig. 2A).[2,7] Marchal et al. proposed a classification of salivary gland stenoses based on their site (main or accessory duct), extent, and number (single, multiple, diffuse). Assessment of the tissue appearance in the stenotic region and the narrowing of the lumen was not included, and the number of patients was not disclosed.[6] In an earlier paper we reported studies in a smaller group of patients in whom differences in the tissue quality of stenoses were observed. Some stenoses had marked inflammatory components whereas others were fibrous. The former type was easier to treat by conservative measures, whereas the vast majority of fibrous stenoses required an instrumental dilatation.[4] It therefore appears necessary to incorporate the condition of tissue quality in the diagnosis, classification, and planned treatment of a stenosis. In this present paper, 111 stenoses in 93 patients were classified according to tissue quality, intensity of luminal narrowing, extend, number, and location (Tables I–IV, Fig. 5). Distal and middle sections of the duct were the segments of the ductal system most often affected, and these results were not significantly different than those published elsewhere.[1] Papillary stenoses were differentiated as a distinct location because in all cases they were associated with previous surgical manipulations to the papilla (60%) or bruxism (40%). The lower rate of multiple stenoses in our group of patients was noteworthy at 12.9% compared with almost 30% in the sialogram-based study of Ngu et al.[1] One important reason might be the more exact and direct assessment of the ductal system provided by the sialendoscope. Precursors of stenosis as web-like encroachments of the duct, segmental inflammations, or fibrosis of the ductal system (Figs. 3A, 4A, B) can only be definitively diagnosed in this way, as is the case with mild to moderate web-associated fibrous stenoses (type II) (Fig. 3 B).

No data concerning the length of stenoses are given in the paper by Ngu et al.,[1] whereas in that of Marchal et al. punctate and diffuse stenoses are differentiated, but without further reference to their precise extent.[6] There is also hardly any data about the differing extent and/or remaining residual lumen. In the publication by Qi et al., a non-high-grade obstruction is distinguished from a high-grade stenosis.[7] The length of the stenosis and, in particular, the width of the residual lumen affect prognosis and are important information in planning effective treatment. Our classification includes, alongside additional changes of the ductal wall (web-like, circular changes), the degree of stenosis and also information about stenosis length. Knowledge about the average width of Stensen's duct from anatomical preparations[11] and the differently sized sialendoscopes of the new endoscopy set (Fig. 1)[2] were of great significance in this regard. The number of our patients who showed long or diffuse stenoses was relatively small (8.6% and 4.3%, respectively). The luminal narrowing seemed to be associated with the different appearance of the tissue in the stenotic area. No data have previously been published about the importance of the differing tissue quality in stenoses. Obstructive parotitis due to inflammatory changes of the ductal system or as a result of a fibrotic ductal stenosis has already been mentioned.[2,4,7] In our group of patients, three types of stenosis could be distinguished in terms of their different tissue quality. Type III stenoses were by far the most frequent at 65.5% (Table I). This differentiation has been included in the present proposed classification of parotid stenoses, which thus represents a modification and enlargement of current descriptions and classification (Fig. 5).[1,6] Type I stenoses were most often (26.7%) of the long variety, but were never diffuse or bilateral. Relative to the others, type II stenoses were most often multiple (17.6%) (Table II) and located in a high percentage near the hilum (35% of all type II stenosis) (Table III). The majority of type I and II stenoses showed a slight to moderate luminal narrowing, whereas type III stenoses were almost invariably high-grade stenoses. Of papillary stenosis, 80% were of type III (the highest proportion) (Table III). Of type III stenosis, 50% to 100% were associated with concurrent diseases, including their related treatment (Table IV). From data published before, it is assumed that tissue quality in the stenotic region requires a different therapeutic strategy, whereas the location could have a determining influence on the chosen surgical technique.[4] The data of the current study confirm these facts. That proposed classification may have influence on the further therapy; type I stenoses may be treated successfully in the majority of the cases by conservative means (e.g., application of cortisone into the duct system). Conservative treatment as single mode therapy may prevent the progression of a stenosis concerning tissue reaction (inflammatory to fibrotic) and/or luminal narrowing (grade of stenosis). A distinct number of patients with type I and II, and nearly all with type III stenoses, however, have to be treated by interventional sialendoscopy or other minimally invasive operative procedures of the duct system.

CONCLUSION

In conclusion, our results show that in addition to location, extent, and number, it is above all the quality of the tissue in the stenotic region that is of great

importance. A transition from one stenosis type to another, e.g., from type I to type III, is certainly conceivable, but does not appear crucial for the treatment strategy to be chosen. Because direct practical consequences can be drawn from the sialendoscopic findings, a classification of stenoses of Stensen's duct should accordingly include the criteria previously discussed.

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