The Laryngoscope

TRIOLOGICAL SOCIETY CANDIDATE THESIS

Diagnosis and Treatment of Congenital Dilatation of Stensen's Duct

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Objectives/Hypothesis: This study aimed to describe the diagnosis and management of congenital dilation of Stensen's duct (CDSD) in seven cases.

Study Design: Retrospective study.

Methods: We collected data including medical records, radiology, and histopathology findings and follow-up for seven patients (four males) with CDSD. The mean age was 23.9 years (range, 2–72 years).

Results: The clinical features of CDSD were the primary presentation of painless swelling in the cheek without an obvious cause that was unilateral or bilateral and occurred at any age, and a swelling along Stensen's duct. In patients without a history of inflammation, aggressive massage of the swelling could produce abundant intraoral salivary flow. Parotid sialography demonstrated a dilated Stensen's duct with a smooth margin but no obvious obstruction. All seven patients underwent superficial parotidectomy including the intact Stensen's duct. No patient showed recurrent swelling after a follow-up of 6 to 65 months.

Conclusions: CDSD is an uncommon congenital disorder of the parotid gland. Management with parotidectomy is effective.

Key Words: Congenital dilatation, Stensen's duct, diagnosis, management.

Level of Evidence: 4.

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INTRODUCTION

Congenital dilatation of Stensen's duct (CDSD) is a rare heteroplasia of the parotid gland, which may have a hereditary background.[1,2] Clinically, CDSD presents as a painless, peculiar swelling along the Stensen's duct. The swelling becomes gradually more pronounced. Most patients do not realize the swelling until they experience secondary infection with pain, fever, and intraoral purulent flow from Stensen's duct.[1–4]

Sometimes the diagnosis of CDSD is difficult. As a rare disorder, treatment of CDSD is also controversial. We aimed to explore the diagnostic points and management of this uncommon disease by analysis of clinical data for seven patients with CDSD.

MATERIALS AND METHODS

From November 2003 to February 2010, we reviewed the records of 200 patients with non-neoplastic diseases of the parotid gland who had received surgical management. Among them were seven patients with a diagnosis of CDSD at the Center of Salivary Gland Disease, Peking University School and Hospital of Stomatology.

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We collected data from medical records, radiology, histopathology findings, and follow-up records. The mean duration of follow-up was 31 months (range, 6–65 months).

RESULTS

Clinical characteristics are shown in Table I. The mean age of patients was 23.9 years (range, 2–72 years); four were males. Bilateral swelling was found in only one patient. No patient had a family history of CDSD.

Medical History

The case history of patients ranged from 4 months to 10 years. Painless swelling was present at the beginning on one or both cheeks for all patients. The swelling increased in size slowly but remained the same size during mastication. One patient experienced increased salivary secretion and decreased swelling after massaging the swollen area, but total remission was never achieved. Four patients obtained medical consultation because of repetitive infections, the symptoms of which could be alleviated by antibiotic therapy. Four patients underwent local puncture and aspiration with needles, which yielded pale pink or milky-white liquid.

Clinical Symptoms and Signs

All patients showed a tube-like swelling in the cheek area following the route of Stensen's duct (Fig. 1). The texture of the mass varied according to the severity of the infection. In three cases without any infection, the swelling was soft, well defined, and movable. In the other

TABLE I.
Clinical Data of the Seven Patients With Congenital Dilation of Stensen's Duct.

No.	Gender	Age (yr)	Course (yr)	Position	Original Diagnosis	Course of Follow-Up (mo)
1	F	4	1/3	Left	CDSD	65
2	M	38	10	Right	Chronic obstructive parotitis	56
3	F	53	13	Bilateral	Chronic obstructive parotitis	48
4	F	2	1	Left	Dilation of salivary duct	12
5	F	72	3	Left	Dilation of salivary duct	20
6	M	63	11	Right	Dilation of salivary duct	10
7	M	7	1	Right	CDSD	6

F = female; CDSD = Congenital dilation of Stensen's duct; M = male.

four cases, the swelling became harder with repeated infection. In one case with severe infections, the entire parotid gland presented as a diffused solid mass.

Radiography

Parotid sialography, ultrasonography, and computed tomography (CT) scan were used for evaluation. All patients underwent parotid sialography (Fig. 2), which demonstrated dilated Stensen's duct with a distinct border in cases with no inflammation. Although cases with repeat infections showed irregular duct margins, we found no evidence of obstructive factors such as stones. Two of the sialograms revealed distended branch ducts but no sialectasis of distal ducts. Four patients underwent ultrasonography. An echoless area corresponded to the location of Stensen's duct, and the sonogram showed a tube-like structure with diameter from 4 to 19 mm (Fig. 3). CT scans of two cases showed dilated tubular soft tissues running along the course of the parotid duct (Fig. 4).

Management

All patients under general anesthesia underwent surgical management including superficial parotidectomy, resection of the entire Stensen's duct, and facial nerve dissection. The procedure is described briefly as follows.[5] A conventional S-shaped preauricular and submandibular incision was made. A skin flap was elevated in the plane deep to the parotid fascia up to the anterior border of the gland. Stensen's duct, usually seen to be dilated, was identified. The duct was dissected forward intraorally as far as possible. Then the surgery continued intraorally. A circular incision was shaped on the mucosa surrounding the ductal orifice. Via blunt dissection laterally and posteriorly, the dilated duct was visualized from the buccinator muscle. The blunt dissection was continued until the pathway of Stensen's duct was perforated completely from intraoral to extraoral region. Then, the Stensen's duct was drawn outside, the facial nerve underwent retrograde dissection, and superficial parotidectomy with the intact Stensen's duct was performed (Fig. 5). The skin and mucosa incisions were closed. A suction drain was placed in the wound.

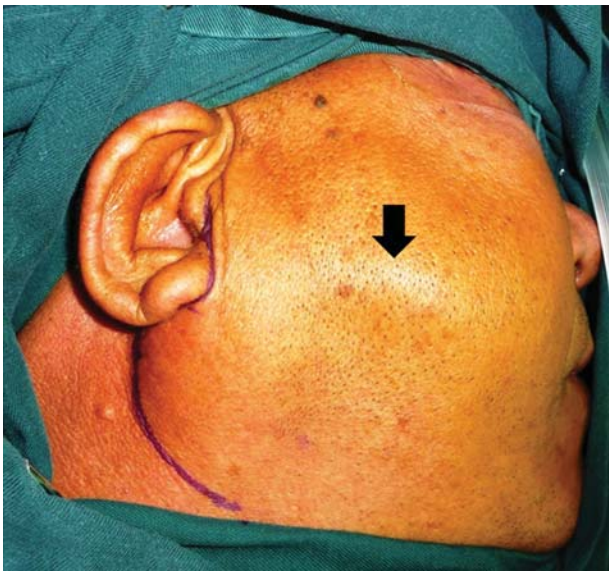

Fig. 1. Facial swelling along the course of Stensen's duct. [Color figure can be viewed in the online issue, which is available at wileyonlinelibrary.com.]

Histopathology

The surgical specimen showed a dilated cystic-like Stensen's duct. If the patient had a history of infection, the duct might not be well identified from its surrounding tissues. Microscopy examination showed the dilated duct lined by single or multiple layers of cuboidal epithelium. Infiltration of inflammatory cells was observed in the parotid gland tissues (Fig. 6). Acinar atrophy and tissue fibrosis were found in two cases.

Follow-Up

Three patients showed temporary facial weakness due to the involvement of the marginal mandibular and buccal branches. The function of the facial nerve recovered completely by 3 months after surgery without recurrent swelling.

DISCUSSION

Congenital sialectasis is one type of rare heteroplasia of the salivary glands, which can involve the ducts of the parotid and submandibular glands.[6,7] In the series reported by Seifert et al., only five cases were diagnosed as congenital sialectasis among the 360 cases of nonneoplastic cystic lesions of the salivary glands.[3]

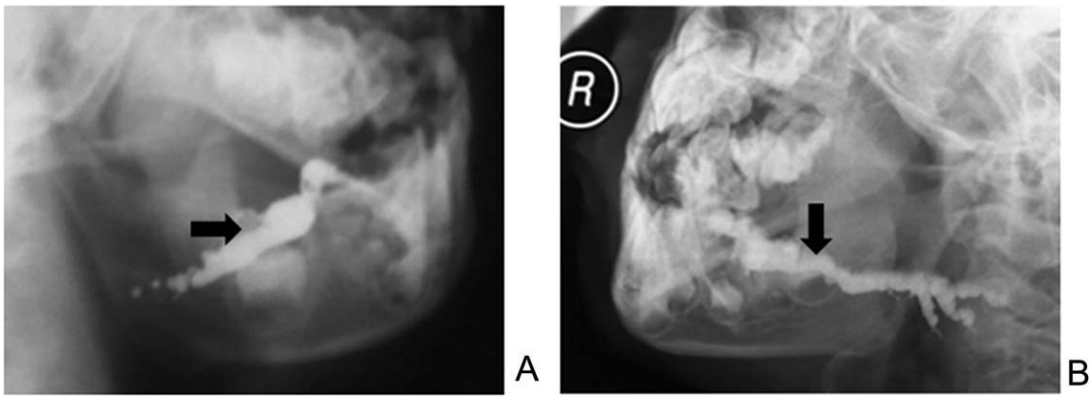

Fig. 2. (A) Dilated left Stensen's duct with a distinct border (arrow); (B) Dilation of right Stensen's duct and primary branching ducts (arrow).

According to the affected location, the condition can be dilation of the main duct or the terminal duct. Our series is among the few reports of CDSD in the English literature.

Etiology

Because of the difficulty in obtaining samples of human embryos, the pathogenesis of CDSD is unclear. Hereditary factors may play a primary role. However, most reports of congenital sialectasis describe the terminal duct.[2–4,6] Our cases did not show heredity. Previously, the presence of a constitutional defect in Stensen's duct was suggested to explain the dilation aggravated by external factors such as salivary retention and ascending infection from the oral cavity.[8,9]

Diagnosis

Because of asymptomatic characteristics in the early stages, CDSD is usually ignored unless patients have secondary infection. Louis[8] reported on five cases of grossly dilated Stensen's duct, all of which presented painless facial swelling. Four cases had no history of or demonstrated obstruction. In our series, similar features were evident in all cases. The swelling of CDSD is not exacerbated by eating in the initial period, although it might clinically act as a symptom of obstructive parotitis after repeated infections. So the history should be investigated for patients with unexplained swelling, and CDSD should be considered in the diagnosis.

Sialography of CDSD demonstrates a highly dilated Stensen's duct with a well-defined border, which is different from the sausage appearance seen on sialography of chronic obstructive parotitis (COP). In addition, even if the duct margin becomes more irregular with repeated infection, no obstructive factors can be observed inside the duct.[1,2,4] CDSD may involve the parotid glands bilaterally, and sialography can reveal the dilatation in both sides, whereas only unilateral swelling may be visible clinically.[2–4] Our study gave no support for this view. Two of our cases underwent bilateral parotid sialography. However, the sialogram showed a normal appearance on the healthy side. Ultrasonography and CT scans are helpful for diagnosis. Ultrasonography of four of our cases revealed a dilated tubular structure. The salivary retention inside the duct was seen as a homogeneous echoless area on the scans. The mean maximal diameter was about 12 mm (range, 4–19 mm), which is much larger than usual (range, 0.9–4 mm). CT

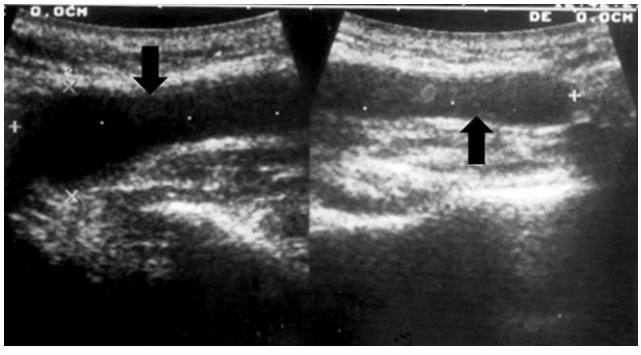

Fig. 3. Sonograms demonstrating an echoless area (arrow) corresponding to the location of Stensen's duct in a diameter of 14 mm.

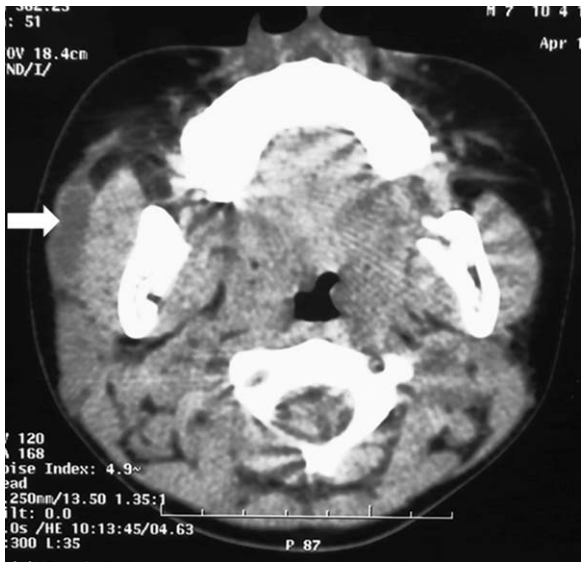

Fig. 4. Computed tomography scan showing dilated tubular soft tissues running along the route of the parotid duct (arrow).

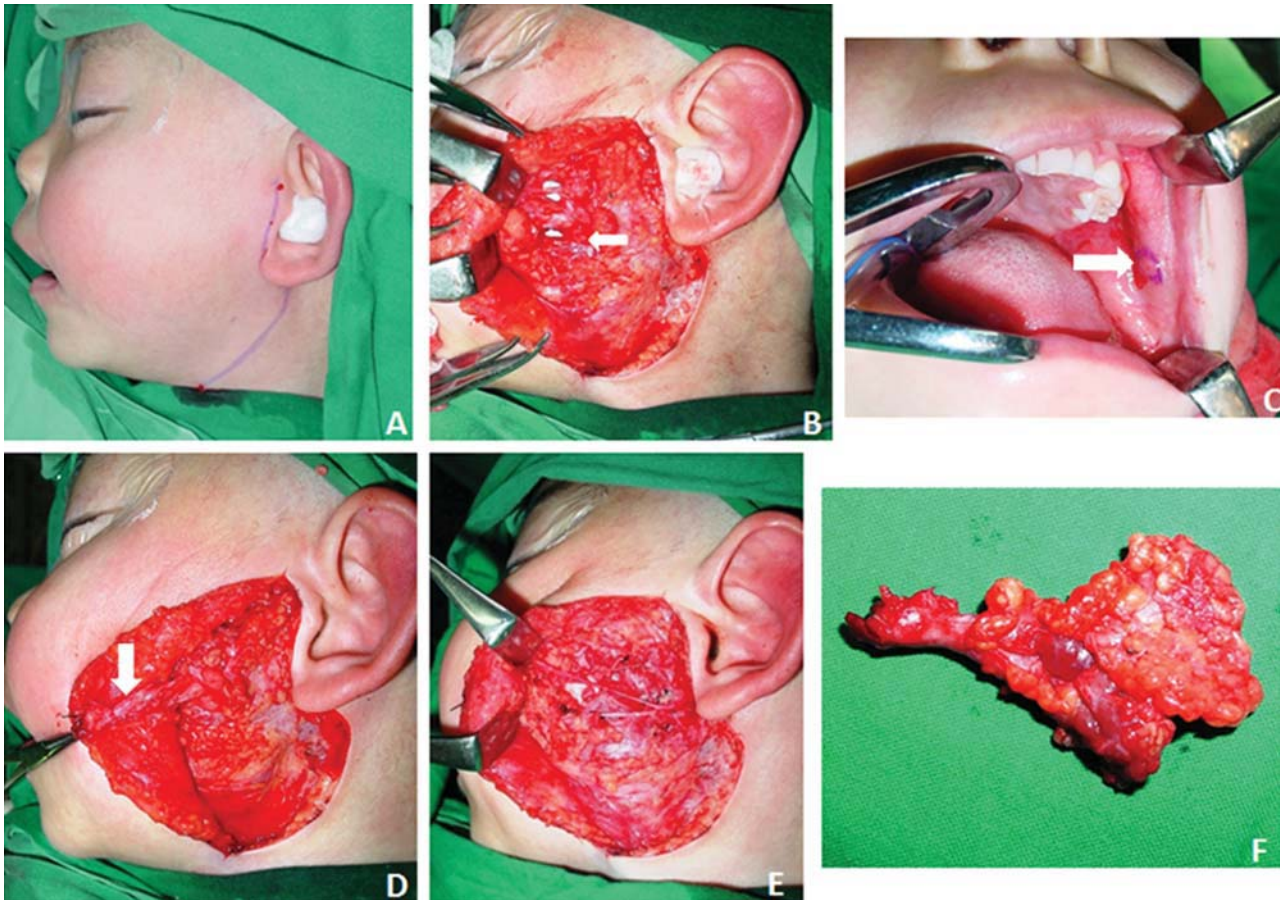

Fig. 5. Surgical procedures. (A) Conventional S-shaped preauricular and submandibular incision. (B) Dilated Stensen's duct (arrow). (C) Circular incision (arrow) was shaped on the mucosa surrounding the ductal orifice. (D) The intact Stensen's duct (arrow) was drawn outside. (E) Superficial parotidectomy accompanying Stensen's duct. (F) The surgical specimen. [Color figure can be viewed in the online issue, which is available at wileyonlinelibrary.com.]

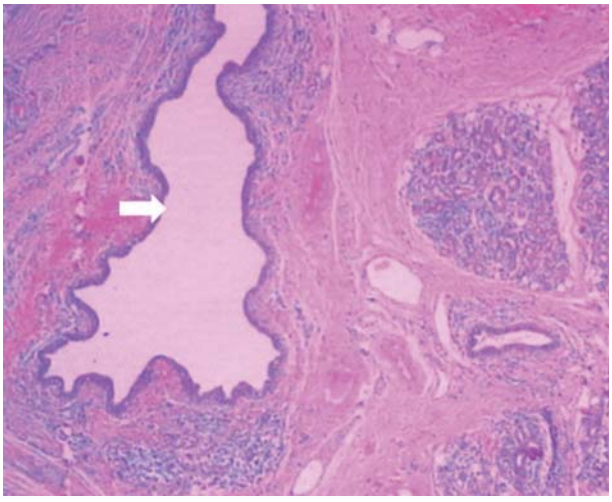

Fig. 6. Photomicrograph of the sample showing dilated Stensen's duct (arrow) lined by 2 to 5 layers of cubical epithelium and infiltration of inflammatory cells in the surrounding parotid gland tissues (hematoxylin and eosin stain, original magnification ×40). [Color figure can be viewed in the online issue, which is available at wileyonlinelibrary.com.]

scanning revealed a dilated duct overlying the masseter muscle sometimes accompanied by parotitis. Radiology examination, especially sialography of the parotid gland, is valuable in the diagnosis of CDSD.

Both the literature and our experience indicate that CDSD shows few characteristics on pathology examination. Microscopy examination can reveal the dilated ducts with the lining structure of normal ones. The duct can be lined by single or pseudostratified columnar epithelium. Inflammatory infiltration, acinar atrophy, and tissue fibrosis can be observed in the parotid gland, which reflects chronic parotitis.

Combining the literature with our experience, the diagnostic features of CDSD can be summarized as follows: 1) the primary symptom of painless swelling in the cheek that is not related to eating, without any evident etiology; 2) may be unilateral or bilateral, and may occur in any age group; 3) clinically, the presence of swelling along the Stensen's duct (in patients without a history of inflammation, aggressive massage of the swelling can produce abundant intraoral salivary flow); and 4) parotid sialography demonstrates dilated Stensen's duct with a smooth margin but no evidence of obstruction.

Differential diagnosis of CDSD is clinically uncommon. The medical history and parotid sialography play essential roles in diagnosis, and three disorders involving swelling of the parotid gland should be considered in the differential diagnosis: juvenile recurrent parotitis (JRP), COP, and salivary duct cyst (SDC).

JRP is the most frequently encountered inflammatory disease of salivary glands in children next to mumps, which usually starts before adolescence, especially between age 5 and 7 years. The condition is characterized by repeating swellings associated with fever, pain, and malaise. The disease is mostly unilateral, and when bilateral, the symptoms are usually prominent on one side. A family history may be present in some cases. The condition is self-limiting after adolescence in more than 90% of cases. Unlike in CDSD, sialography of JRP demonstrates sialectases in terminal ducts.[10,11]

CDSD accompanied by infection can induce the repetitive swelling of the parotid gland and may result in misdiagnosis of COP. However, the two conditions can be differentiated on both history and radiography. History reveals that the glandular swelling of COP is usually induced by eating. Referring to radiography, types of obstructive factors can be revealed by sialography. Furthermore, the sausage-shaped appearance of Stensen's duct and sialectases in terminal ducts can be visible in severe cases of COP.[12,13]

Another disease distinguished from CDSD is SDC, which is usually located at the border of the parotid gland. On palpation, the swelling is soft and fluctuant. SDC is considered one type of retention cyst. It occurs most frequently between age 60 and 70 years, which indicates that congenital hypoplasia is not responsible.[1–3]

Management

Because there are no symptoms in the early stage of CDSD, conservative management is considered the first choice. Milking the swelling in the intraoral direction is helpful in emptying the retained saliva. Even if no serious consequences have developed during longstanding history, the possibility of secondary infection should not be ignored. Maintaining good oral hygiene, drinking more water, and chewing gum to stimulate the secretion of saliva are also helpful in preventing secondary inflammation.

Acute exacerbations can be treated with symptomatic treatment. Recently, Baurmasb reported an intraoral approach suturing the dilated duct segment to the buccal mucosa, provided the dilated portion of the duct is anterior to the ramus where it is accessible.[8]

However, conservative management is not suitable for patients with repeat infections. In the current study, all patients underwent superficial parotidectomy together with excision of the intact Stensen's duct.[7] The results of follow-up were satisfactory, which suggested that the definitive therapy for the dilated duct with chronic parotitis should be surgical treatment.

The surgical procedures are generally the same as for superficial parotidectomy, and an elliptical or circular incision should be made on the mucosa surrounding the ductal orifice to ensure the complete removal of the duct. Protection of the facial nerve is critical during parotidectomy[5] and is more difficult in CDSD because of inflammatory fibrosis of the gland and close adhesion of the facial nerve with glandular tissues. Vitamin B1 and B12 should be prescribed for postoperative facial weakness. In addition, physical therapy and training of facial muscles can facilitate the recovery of the facial nerve and avoid atrophy of facial muscles.[14]

CONCLUSION

CDSD is an uncommon congenital disorder of the parotid gland and presents particular features in clinical diagnosis and management.

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