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Clinical Characteristics and Management of Congenital Submandibular Duct Atresia: A Systematic Review

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ABSTRACT

Congenital submandibular duct atresia (CSMDA) is a rare congenital oral lesion that appears as a cystic swelling along the course of the submandibular duct (SMD) without a lumen. Several cases of CSMDA have been reported with varying presentations and management. This review aims to identify the clinical characteristics and recovery outcomes among infants with CSMDA. The review was conducted by searching several databases from inception to August 2023 following the Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines. The search identified 234 studies, with 25 meeting the inclusion criteria. A total of 33 infants aged 0 days to 22 months were included, with a male predominance (75.8%). About 72.7% had unilateral cystic swelling. Infants with bilateral CSMDA (87.5%) were more likely to present with swallowing difficulties than unilateral CSMDA. Around 12% of the included children recovered spontaneously. Marsupialization of SMD is the most favored mode of treatment (83%) with no documented recurrence. Surgical treatment is recommended for infants presenting with swallowing difficulties. Yet, the quality of evidence is inadequate to recommend a widespread mode of treatment until a better quality study has been completed.

Keywords: Congenital oral lesion, congenital salivary gland anomaly, floor of mouth, infants, submandibular duct imperforate

Introduction

Congenital submandibular duct atresia (CSMDA), also known as imperforate submandibular duct (SMD), entails the absence or narrowing of the SMD duct lumen, resulting in cystic swelling on the floor of the mouth. The cystic swelling results from the accumulation of saliva following the disruption of the normal flow of saliva in the oral cavity.[1] Typically, infants may present with swallowing or airway issues, albeit most swellings go unnoticed.

Albeit rare, the clinical implications of CSMDA cannot be underestimated as it may significantly impact infants' breathing and swallowing processes, leading to a substantial impact on quality of life.[1] Parallel to that, delay or failure of timely decompression of the obstructed duct predisposes a high chance of submandibular gland infection or atrophy.[1] A satisfactory outcome has been widely documented following simple incision and marsupialization. Yet, reports on spontaneous regression of the cystic swelling challenge the need for early intervention among the affected infants.

The current literature highlights the clinical presentation, management, and outcomes of CSMDA. The clinical characteristics and treatment outcomes among infants presenting with this entity are aimed to be identified, and the available evidence is synthesized and analyzed to shed light on the current knowledge surrounding CSMDA.

Methods

The review was conducted based on PRISMA guidelines to identify studies that presented the clinical course and management of CSMDA.

Eligibility Criteria

The PICOS framework was adopted in this review, with infants and neonates under 24 months being included. The intervention or exposure group is children with CSMDA diagnosed clinically with an imperforate duct opening or with additional investigations such as imaging. Comparators are none.

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Outcomes Are Classifieds:

1. Primary outcome: Resolution of floor of mouth swelling, saliva presence, and improvement of symptoms: swallowing/breathing/weight gain.
2. Secondary outcomes: Revision surgery, complications, adverse effects, length of follow-up.

Conference abstracts, letters to editors, commentaries, review articles, publications without peer review, and gray literature were excluded. As the present publications on CSMDA are scarce, no restrictions on sample size were placed.

Data Sources and Searches

A literature search was completed to address the aim of this study. In September 2023, the electronic databases of PubMed/Medline, CINAHL, Scopus, and Google Scholar were searched using the terms, syntax, and Boolean combinations. Peer-reviewed articles published in the English language were included. The following key terms were searched: submandibular duct (SMD) atresia; imperforate SMD; SMD obstruction; salivary gland congenital anomaly; congenital oral lesion; and pediatric.

Complete details of the search strategy are in Figure 1. References to the included studies were also included to ensure a robust search procedure. Duplicate studies were excluded by using EndNote X10 software. The search was conducted over a period of 1 month (August 2023) in accordance with Preferred Reporting Items for Systematic Reviews and Meta-analyses guidelines.[2] After being scrutinized by the panel members, 25 peer-reviewed studies were included for review.

Study Selection

Through the initial screening of articles by the primary reviewer, duplicates, review articles, conference proceedings, editorials, poster presentations, and articles in languages other than English were removed. After this, 2 reviewers independently applied selection criteria to the title, abstract, and full-text articles. Most differences were resolved through consensus. A third party was consulted if differences were unresolved until a final decision was reached.

Two authors independently performed the data extraction, which was inserted into a predefined Excel spreadsheet. The following information was included: first author's last name; study design; country of the participants; the total number of patients; age; gender; comorbidities; presenting symptoms; associated symptoms including airway, swallowing and poor weight gain; diagnostic methods; adjunct investigations such as imaging, treatment, complications; and outcomes.

Main Points

- Congenital submandibular duct atresia (CSMDA) is one of the differential diagnoses of cystic lesions in newborns.
- Male gender is a risk factor for CSMDA.
- Neonates with CSMDA present with difficulty in swallowing and failure to thrive.
- Cystic lesion with the absence of duct opening at the floor of the mouth is adequate to diagnose CSMDA.
- Marsupialization of the submandibular duct is the ideal treatment of CSMDA, which prevents recurrence and requires no follow-up.

Data Synthesis

It was not possible to complete a meta-analysis as heterogeneity was found in (1) population diagnoses, (2) data collection methods and measures used in the included studies, and (3) study outcomes reported. A narrative approach was therefore used to synthesize the data on key study variables, study quality, and risk of bias.

Outcome Measures

The primary outcome was determined as the resolution of symptoms or changes in symptoms (resolved swelling). Secondary outcomes included revision surgery, the presence of complications, and follow-up.

Ethical Consideration

This review was conducted in accordance with ethical standards. All authors declare no conflicts of interest.

Results

Study Selection

A total of 234 English-language articles were identified. At the initial stage, the titles and abstracts were screened for eligibility. Forty-six studies described the presentation and management of CSMDA and were evaluated in-depth by reading their full texts. Studies were excluded for the following reasons: commentaries (5), the study was nontherapeutic in nature (24), reported only complications (1), involved removal of the gland (1), and was only published as an abstract (1). Twenty-five articles published from inception to August 30, 2023 fulfilled the selection criteria and were included in the review.[1,4-27] The included articles are retrospective cohort case series or case reports (Level IV).

Epidemiology and Clinical Features

Thirty-three patients were pooled (Female: 8 [24%]). The time of presentation ranged between birth and 22 months. Bilateral SMD involvement was observed in 27.3% of cases,[5,8,11,12,15,17,20,21] whereas unilateral SMD involvement was present in 24 (72.7%) children, with a left predominance (66.7%) being demonstrated. Table 1 demonstrates the summary of all articles included in this review. A meta-analysis was considered inappropriate in our study because of the heterogeneity found among the included studies.

No significant antenatal or postnatal history was noted among the included children, although 9% of the included infants were found to be premature.[4,7,17] Seventeen of the included children were asymptomatic. Feeding difficulties were reported among 48.5% of the included children,[1,5,8,11,13,15,17,19-21,24-26] whereby loss of weight was documented in 2 children.[5,20] Feeding difficulties (87.5%) were predominantly noted among infants with bilateral CSMDA.[5,8,11,15,17,20,21] None of the included patients with congenital SMD atresia had airway complaints.

Diagnosis and Investigations

The diagnosis of imperforate SMD was made clinically in 26 of the included patients. Additional investigations were performed in 10 infants, including imaging: 3 patients for magnetic

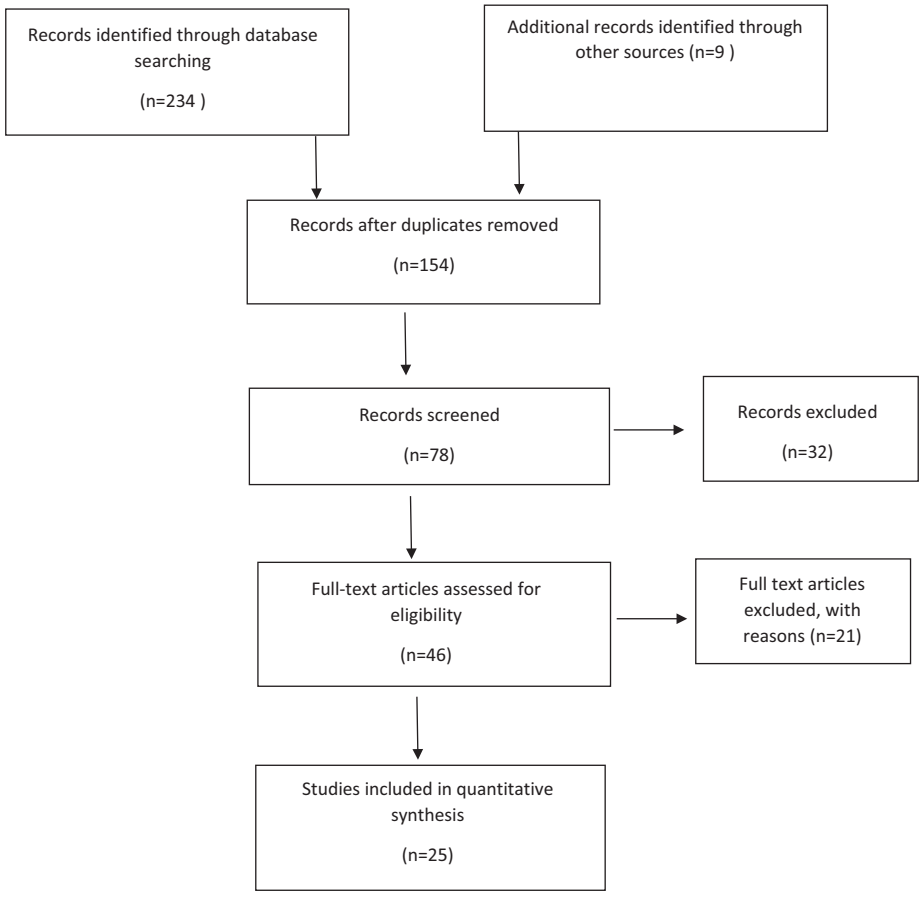


Figure 1. Summary of the search strategy.

resonance imaging (MRI),[10,17,25] 3 patients had computed tomography (CT),[11,21,26] 1 patient had multi-detector computer tomography (MDCT),[18] 1 patient had video-sialoendoscopy,[15] 1 patient had intraoperative ultrasound,[13] and 1 patient had intraoperative probing.[21] Histopathological findings were mentioned in 4 studies, whereby pseudostratified ciliated epithelium was found in 3 studies,[4,7,24] and single cuboidal epithelium in 1 study.[18]

Treatment and Outcome

Surgical treatment is the most common mode of treatment among 28 of the included patients, while 4 children were managed conservatively (12%).[9,26,27] Marsupialization of the SMD was the most favored surgical technique performed in 15 children (83%).[4,7,8,10,11,13,14,17-19,21,22,25]

Recurrence was reported in 2 studies involving 3 patients,[12,20] whereby one child underwent cystic enucleation and right sublingual gland excision,[12] the other child was not operated on as parents defaulted follow-up[12], and Ellegård et al[20] reported on bilateral SMD recurrence in a child which was managed with marsupialization and the other incision along the duct[20] with subsequent right sublingual gland excision[12] and marsupialization.[20]

Infection was documented in 1 study.[20] The child also developed recurrence and failure to thrive, which resolved with marsupialization of bilateral SMD. The follow-up period ranged between 3 months and 15 years.

Discussion

Since it was first described in 1955 by Scher, myriad reports on CSMDA have been published.[28] The development of the submandibular gland begins as a thickening of the epithelium, which protrudes into the underlying mesenchyme. The epithelium subsequently invaginates, forming the main salivary duct that connects the epithelial bud to the oral surface. This epithelial bud branches and creates a multilobed gland. The duct is formed from solid epithelium, which undergoes cavitation, enabling flow between the saliva-producing acini and the oral cavity.[29] Failure to recanalize the epithelium has been postulated to result in SMD atresia.[22]

In this review, 78.5% of the infants diagnosed with CSMDA were male. Interestingly, other congenital salivary gland anomalies noted to have a male predominance include the aplastic or hypoplastic salivary gland.[30,31] Similarly, plunging ranulas, which result from the extravasation of mucus from a ruptured salivary gland duct, also demonstrate a male predilection.[32] It was found that no specific antenatal or postnatal characteristics or syndrome associated with CSMDA, although 3 (9%) of the included infants were premature.[4,7,17] Even though prematurity is known to be a risk factor for congenital anomalies, it was found that no significant association among the included patients.[33] Almost all the observed infants revealed a unilateral clinical presentation (73%) with left-sided predominance (66.7%), although the exact cause of the laterality pattern remains elusive. Any associations between modes of

Table 1. Summary of Main Findings

Author, Year, Country	Total Patients	Mean Age	Gender	Ante-natal/ Post-natal History	Uni/Bilateral	Presentation Airway/ Swallowing/ Voice	How Was the Diagnosis Made?	Additional Investigation	Intervention	Outcome/ Complication	Follow-up/ Recurrence
Redpath et al[4], 1969, UK	1	12 hours of life	Female	Premature	Uni Right	None	Clinical	None	Marsupialization Hpe: pseudostratified columnar epithelium	None	NR
Hoggins et al[5], 1974 UK	2	At birth	Female Male	Uneventful	1-Bilateral 1-Left	2-Feeding difficulty 1-Loss of weight	Clinical	None	Incision and polyglycolic acid seton insertion	None	No recurrence Duration not reported
Rees et al[6], 1979 UK	1	At birth	Male	Uneventful	Right	None	Clinical	None	Decompression	Recurrence after 2 weeks Swelling subsequently burst 2 days	1 year
Addante et al[7], 1984 USA	1	13 months At birth	Male	Premature at 34 weeks	Uni Left	None	Clinical	None	Surgical Marsupialization HPE: cystic cavity lined with pseudostratified columnar epithelium with brush borders and thin connective tissue consistent with a dilated salivary duct	None	24 months
Pownell et al[8], 1992, USA	4	At birth 0 × 2 2 1	3 Female 1 NR	Uneventful second branchial cyst	3 Uni (1 right 2 left) 1 both	None 3 Feeding issue	Clinical Imperforate SMD	None	Marsupialization with ductoplasty 2 Marsupialization 2	No recurrence No complications	1 year 4 months NR 2
Steelman et al[9], 1998 USA	1	Day 1	Male	Uneventful	Right	None	Clinical 1 × 0.5 cm mass	None	Conservative-resolved in 5 weeks	No recurrence	20 months
Kawahara et al[10], 2000 Japan	1	1 year 10 months	Male	Uneventful	Uni Right	None	Clinical and imaging congenital dilation of the submandibular duct	MRI Fluid-containing lesion extending from the submandibular gland to the anterior aspect of the floor of the mouth	Marsupialization	No recurrence	6 months
Amin et al[1], 2001, UK	2	3 months 2 day	Male	Term, no significant ante-post-natal	Uni: Both left side	Distressed at feeding	Clinical Cystic lesion 2 cm on left side	None	Surgical incision to decompress under LA	NIL	15 years 4 years
Hoffrichter et al[11], 2001 USA	1	3 months	Male	Uneventful	Bilateral	Distressed at feeding	Clinical + Imaging No saliva expressed on palpation	CT Dilated, SMD bilaterally	Incision bilateral Marsupialised Left Right had reduced at the time of surgery	NIL	5 months
Ugboko et al[12], 2002 Nigeria	2	At birth	Male	Uneventful	Bilateral Bilateral	None	Clinical	None	1-Puncture with needle 1-Incision	-1 child had cystic enucleation and sublingual gland excision -Other child defaulted follow-up	No reccurence after 6 months after excision

(*Continued*)

Table 1. Summary of Main Findings (Continued)

Author, Year, Country	Total Patients	Mean Age	Gender	Ante-natal/ Post-natal History	Uni/Bilateral	Presentation Airway/ Swallowing/ Voice	How Was the Diagnosis Made?	Additional Investigation	Intervention	Outcome/ Complication	Follow-up/ Recurrence
Wada et al[13], 2004 Japan	1	At birth	Male	Uneventful	Right	Distress at feeding	Clinical + Imaging + Laboratory analysis (S-type amylase isoenzyme)	USG: unicystic hypoechoic lesion in which irregular hyperechoic granular materials were found	Aspiration of fluid Marsupialization OK-432 injection 3 times (0.8 KE, 1.0 KE, and 2.5 KE solutions)		
Walker et al[14], 2005 Australia	1	Day 3	Male	Uneventful	Left	None	Clinical	None	Incision and Marsupialization	None	7 months
Capaccio et al[15], 2007 Italy	1	4 months	Male	Uneventful	Bilateral	Swelling evident during feeding and crying	Clinical and video-sialoendoscopy trifurcation of the main duct	Video-sialoendoscopy	Surgical incision Video-sialoendoscopy	NIL	19 months
Pal et al et al[16], 2007 Saudi Arabia	1	Day 1	Male	Uneventful	Uni (right)	None	Clinical	None	Incision	Nil	6 months
Ulualp et al[17], 2007, USA	1	2 month	Male	35 weeks, ASD, PS, gastrochiasis	Bilateral	Distress at feeding	Clinical + Imaging Cystic swelling FOM	MRI Symmetric dilation of both submandibular ducts along the entire length of the ducts	Stab incision + marsupialization of SMD	NIL	3 months
Basaran et al[18], 2009 Turkey	1	2 months	Male	Uneventful	FOM	9	Clinical and Imaging	MDCT Dilatation of both the intra- and extraglandular segments of duct of Wharton was presumed to be due to congenital stenosis of the orifice of duct of Wharton	Surgery Marsupialization HPE: single-stranded cuboidal epithelium with apical secretions	NIL	NR
Rosow et al[19], 2009, USA	2	5 months 1 month	Male	NR	Uni Left NR	Feeding difficulty since 3 weeks of age (treated as GERD)	Cystic swelling bluish translucent, no SD orifice	None	Surgery Sialodochotomy The terminal duct papilla was excised		19 months 30 months
Ellegård et al[20], 2010, Sweden	1	2 months	Male	C-section 37 + 2 (pre-eclampsia, asphyxia) SGA (BW: 1.84 kg)	Bi-lateral	Feeding difficulty, failure to thrive	Clinical 2 elongated translucent structures elevate tongue, submandibular ducts were enlarged 5 mm no saliva	None	Surgical Incision made along the duct LA	Infection: Resolve with antibiotics Recurrence both ducts, child not feeding and failure to thrive: Marsupialization of both ducts	10 months: no recurrence, weight, and feeding improve

(*Continued*)

Table 1. Summary of Main Findings (Continued)

Author, Year, Country	Total Patients	Mean Age	Gender	Ante-natal/ Post-natal History	Uni/Bilateral	Presentation Airway/ Swallowing/ Voice	How Was the Diagnosis Made?	Additional Investigation	Intervention	Outcome/ Complication	Follow-up/ Recurrence
Aronovich et al[21], 2014 USA	1	5 months	Female	Term, Preeclampsia	Bilateral at birth	Feeding difficulty	Clinical + Imaging (CT)	Probing CT Intraop USG small hockey-stick transducer probe at a frequency of 9 Hz was used to visualize the dilated Wharton ducts longitudinally and confirm the diagnosis	Incision, marsupialization bilateral	None	3 months
Hseu et al[22], 2016 USA	1	3 months	Male	NR	Left	Asymptomatic	Clinical Left FOM fluid-filled+blind ending duct with a fibrotic plug at the oral mucosa	None	Surgery Sialodochoplasty	NIL	3 months
Pontes et al[23], 2018 Brazil	1	4 months	Male	None	Left	Asymptomatic	Clinical	None	Incision, excision	No recurrence	51 months
Prosdócimo et al[24], 2018 Brazil	1	1 month	Female	NR	FOM	Difficulty in swallowing (Breast feeding)	Clinical Translucent, smooth cystic swelling FOM 3 × 2 cm	None	Excision Cystic cavity lined with pseudostratified columnar epithelium with brush borders and thin connective tissue consistent with a dilated salivary duct	NIL	6 months
Al-Badr et al[25], 2020 France	1	3 months	Male	Uneventful	Left	Difficulty in feeding, irritability	Clinical + Imaging	MRI Dilated left SMD	Excision and marsupialization		6 months
Gupta et al[26], 2021 India	1	2 months	Male	Anorectal malformation sigmoid colostomy	Left	-Difficulty in feeding -Slowly reducing in size	Clinical + Imaging	CT Tubular fluid attenuation in FOM USG Anechoic, tubular structure (the dilated left SMD)	Conservative	NR	NR
Freitas et al[27], Brazil, 2022	2	3 months 5 months	Female Male	Nothing significant	Uni Right Left (Male)	None	Clinical Cyst right FOM since birth Cyst left since birth Both with imperforate SD	None	Conservative	Spontaneous regression: 7 months Spontaneous regression at 6 months	2.9 years 2.2 years

HPE, histopathological examination; MRI, magnetic resonance imaging; NR, not reported; SMD, submandibular duct.

delivery such as Caesarean section, vaginal delivery, or breech presentation and laterality could not be elicited among the included patients.

In this review, while most of the infants with CSMDA were asymptomatic or were discovered (51.5%) during medical check-ups, 48.5% of the infants had swallowing issues, with failure to thrive demonstrated in 12.5% of the infants. 87.5% of the children with bilateral involvement were found to present with swallowing difficulties in comparison to the unilateral ones (45.8%). It is postulated that the enlarging cystic swelling located at the floor of the mouth hinders the sucking and swallowing process in infants, resulting in feeding difficulty and, subsequently, failure to thrive.

It is worth noting that several entities may appear with cystic swelling in the floor of the mouth in neonates, including duplication anomalies of the duct, teratomas, heterotopic gastric cysts,[1] dermal/epidermal inclusion cysts, thyroglossal duct cysts, bronchogenic cysts, lymphatic and vascular lesions, salivary gland tumors, and sialolithiasis.[8,19] Traditionally, the appearance of a bluish, cyst-like swelling filled with translucent fluid along the course of the SMD at the floor of the mouth with no lumen suggests CSMDA. Congenital submandibular duct atresia is often mistaken for a ranula, although ranulas are pseudocysts with no epithelial lining, whereas CSMDA has true epithelium-lined cysts.[19]

About 78.8% of the included infants were diagnosed clinically. Adjunct investigations were performed in 10 studies[10,11,13,15,17,18,21,25,26] which included intraoperative ultrasound, CT, video-sialoendoscopy, and MRI. These additional investigations are mainly performed to exclude the presence of other anomalies. The presence of an elongated and dilated tubular structure, with a segmental lobe in the floor of the mouth following the course of the SMD seen in the MRI, is suggestive of CSMDA.[34] In the same vein, histological examination may also aid in accurate diagnosis and differentiate it from ranula. Classically, a cystic cavity lined by respiratory epithelium with brush borders and thin connective tissue suggests a dilated SMD.[1,24]

Spontaneous regression of the cystic swelling was reported in 12% of the included patients.[9,26,27] Oliveira Freitas et al[27] postulated that introducing food as a contributory factor to the regression of the dilated SMD as friction or damage caused by the food may result in rupture and subsequent regression. Parallel to that, suction movement from breastfeeding has also been reported to stimulate salivary drainage, thus resulting in natural perforation of the dilated SMD, although most included articles did not mention the feeding mode.[35] Interestingly, none of the infants treated conservatively developed recurrence or documented complications.

Surgical treatment was the most favored mode of treatment amongst the included patients (84.8%). Numerous surgical options, from simple stab incision to excision of the end portion of the SMD with ductoplasty, stenting, and marsupialization, have shown promising results.[17] Marsupialization of the SMD was the most favored surgical technique performed in 15 children (15/28). This review found that a simple incision resulted in infection[20] and recurrence.[12,20] Needle drainage or stab incision results in rapid re-epithelialization, leading to reaccumulation of the fluid as a permanent drainage pathway failed to be established.[19] It is recommended that infants presenting with CSMDA swallowing difficulties undergo marsupialization of the SMD. At the same time, it is highlighted that CSMDA should be considered a differential diagnosis in infants presenting with swallowing difficulties.

The number and type of included studies limited the review. Hence, the provided conclusion was difficult to make. Therefore, the results should be interpreted cautiously, and future comparative studies are warranted. In the current review, there is insufficient data to demonstrate the epidemiology, clinical characteristics, and optimal management of the CSMDA. Additionally, significant heterogeneity was noted across the studies included.
Congenital submandibular duct atresia has a high male predilection (78.5%) with unilateral predominance (73%). Bilateral involvement results in swallowing issues (87.5%). Surgical treatment, notably marsupialization, is found to be the most favored mode of treatment. Yet, the quality of evidence is inadequate to recommend a widespread mode of treatment until a better-quality study has been completed.

Data Availability Statement: The data that support the findings of this study are available on request from the corresponding author.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept – J.S.; Design – J.S.; Supervision – E.T., G.V.D.M.; Resources – J.S.; Material – J.S., E.T.; Data Collection – J.S., E.T.; Analysis – J.S.; Literature serach – J.S., E.T.; Writing – J.S.; Critical Review – E.T., C.M., G.V.D.M.

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References

1. Amin MA, Bailey BM. Congenital atresia of the orifice of the submandibular duct: a report of 2 cases and review. *Br J Oral Maxillofac Surg*. 2001;39(6):480-482. [CrossRef]
2. Moher D, Liberati A, Tetzlaff J, Altman DG, PRISMA Group. Preferred reporting items for systematic reviews and metaanalyses: the PRISMA statement. *BMJ*. 2009;339:b2535. [CrossRef]
3. Moola S, Munn Z, Tufanaru C, et al. Chapter 7. Systematic reviews of etiology and risk. In: Aromataris E, Munn Z, eds. *JBI Manual for Evidence Synthesis*. 2020. Available at: https://synthesismanual.jbi.global.
4. Redpath TH. Congenital ranula. *Oral Surg*. 1973;31:446-557.
5. Hoggins GS, Hutton JB. Congenital sublingual cystic swellings due to imperforate salivary ducts: two case reports. *Oral Surg Oral Med Oral Pathol*. 1974;37(3):370-373. [CrossRef]
6. Rees RT. Congenital ranula. *Br Dent J*. 1979;146(11):345-346. [CrossRef]
7. Addante RR. Congenital cystic dilation of the submandibular duct. *Oral Surg*. 1984;59:656-658.
8. Pownell PH, Brown OE, Pransky SM, Manning SC. Congenital abnormalities of the submandibular duct. *Int J Pediatr Otorhinolaryngol*. 1992;24(2):161-169. [CrossRef]

9. Steelman R, Weisse M, Ramadan H. Congenital ranula. *Clin Pediatr*. 1998;37(3):205-206. [CrossRef]
10. Kawahara K, Hotta F, Miyachi H, Shirozu T. Congenital dilation of the submandibular duct: report of a case. *J Oral Maxillofac Surg*. 2000;58(10):1170-1172. [CrossRef]
11. Hoffrichter MS, Obeid G, Soliday JT. Bilateral submandibular duct atresia: case report. *J Oral Maxillofac Surg*. 2001;59(4):445-447. [CrossRef]
12. Ugboko VI, Hassan O, Prasad S, Amole AO. Congenital ranula: a report of two cases. *ORL J Otorhinolaryngol Relat Spec*. 2002;64(4):294-296. [CrossRef]
13. Wada T, Yamamoto H, Ootsuki R, Nakatani K, Fujita S. Congenital ranula. *Asian J Oral Maxillofac Surg*. 2004;16(1):63-66. [CrossRef]
14. Walker P. Imperforate submandibular duct. *Otolaryngol Head Neck Surg*. 2005;132(4):653-654. [CrossRef]
15. Capaccio P, Gaini LM, Pagani D, Sambataro G, Pignataro L. Videosialoendoscopic assessment of bilateral atresia of the Wharton's duct orifice in an infant. *J Pediatr Surg*. 2007;42(9):E5-E7. [CrossRef]
16. Pal K, Abdulla AM. Congenital imperforate submandibular duct in a newborn. *Indian J Pediatr*. 2007;74(7):687-688. [CrossRef]
17. Ulualp SO, Rodriguez SC, Hernandez J, Hay M. Bilateral atresia of the submandibular duct orifices. *Am J Otolaryngol*. 2007;28(3):184-186. [CrossRef]
18. Basaran C, Ulu EMK, Donmez FY, Tore HG, Aydin E, Coskun M. Unilateral congenital cystic dilatation of the submandibular gland duct: MDCT findings. *Int J Pediatr Otorhinolaryngol*. 2009;4(4):147-149. [CrossRef]
19. Rosow DE, Ward RF, April MM. Sialodochostomy as treatment for imperforate submandibular duct: a systematic literature review and report of two cases. *Int J Pediatr Otorhinolaryngol*. 2009;73(12):1613-1615. [CrossRef]
20. Ellegård E, Kjellmer I. Atresia of the submandibular duct orifices: an unusual cause of feeding problems and failure to thrive in an infant. *Acta Paediatr*. 2010;99(7):1100-1101. [CrossRef]
21. Aronovich S, Edwards SP. A case of imperforate Wharton duct. *J Oral Maxillofac Surg*. 2014;72(4):744-747. [CrossRef]
22. Hseu A, Anne P, Anne S. Congenital atresia of Wharton's duct. *J Clin Diagn Res*. 2016;10(2):MD01-MD02. [CrossRef]
23. Pontes FSC, de Souza LL, Pedrinha VF, Pontes HAR. Congenital ranula: a case report and literature review. *J Clin Pediatr Dent*. 2018;42(6):454-457. [CrossRef]
24. Prosdócimo ML, Barreto Nogueira AP, Aparecida de Albuquerque Cavalcante M, Agostini M, Benevenuto de Andrade BA, Romañach MJ. Congenital dilatation of the submandibular duct. *Int J Pediatr Otorhinolaryngol*. 2018;113:16-18. [CrossRef]
25. Al-Badri N, Nicot R, Touzet-Roumazeille S, Schlund M. Imperforate submandibular duct presenting as feeding difficulties. *Arch Dis Child*. 2021;106(4):383. [CrossRef]
26. Gupta A, Bansal A, Verma A, Naranje P, Sangwan A. Imperforate submandibular duct in an infant. *J Paediatr Child Health*. 2022;58(5):921-923. [CrossRef]
27. Oliveira Freitas L, Seixas Mourão P, Gonçalves MWA, Ramos-Jorge ML, Marques Mesquita AT. Congenital atresia of the submandibular gland duct: report of two clinical cases with spontaneous regression and literature review. *Rev Estomatol*. 2022;30(1):1-8. [CrossRef]
28. Scher LB, Scher I. Case of imperforate submandibular ducts in an infant. *Br Dent J*. 1955;98:324-325.
29. Calzada GG, Hanna EY. Cummings otolaryngology head & neck surgery. In: *Benign neoplasms of salivary glands*. Flint PW, Haughey BH, et al. eds. Philadelphia: Mosby Elsevier; vol 87. 1162-1177.
30. Matsuda C, Matsui Y, Ohno K, Michi K. Salivary gland aplasia with cleft lip and palate: a case report and review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 1999;87(5):594-599. [CrossRef]
31. Goldenberg D, Flax-Goldenberg R, Joachims HZ, Peled N. Misplaced parotid glands: bilateral agenesis of parotid glands associated with bilateral accessory parotid tissue. *J Laryngol Otol*. 2000;114(11):883-885. [CrossRef]
32. Chin SJ, Zeng ISL, Morton RP. The epidemiology of plunging ranula in South Auckland. *Laryngoscope*. 2016;126(12):2739-2743. [CrossRef]
33. Gunn-Charlton JK. Impact of comorbid prematurity and congenital anomalies: a review. *Front Physiol*. 2022;13:880891. [CrossRef]
34. Mandel L, Alfi D. Diagnostic imaging for submandibular duct atresia: literature review and case report. *J Oral Maxillofac Surg*. 2012;70(12):2819-2822. [CrossRef]
35. Gibson MHL. The prenatal human submandibular gland: a histological, histochemical and ultrastructural study. *Anat Anz*. 1983;153(1):91-105.