






## CASE SERIES

# Congenital atresia of the submandibular gland duct: report of two clinical cases with spontaneous regression and literature review

## Atresia congênita do ducto da glândula submandibular: relato de dois casos clínicos com regressão espontânea e revisão da literatura

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### OPEN ACCESS

#### Afiliación Institucional

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#### Citation:

Oliveira Freitas L., Seixas Mourão P., Aparecido Gonçalves M.W., Ramos-Jorge M.L., Marques Mesquita A.T. Congenital atresia of the submandibular gland duct: report of two clinical cases with spontaneous regression and literature review. *Rev Estomatol.* 2022; 30(1):e11829. DOI: 10.25100/re.v30i1.11829

**Received:** December 21<sup>th</sup> 2021

**Evaluated:** March 28<sup>th</sup> 2021

**Accepted:** April 20<sup>th</sup> 2022

**Published:** May 11<sup>th</sup> 2022

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#### ABSTRACT

**Background:** Congenital atresia of the submandibular gland duct is a rare condition, originated during the embryogenic process, characterized by swelling in the region of the oral floor. A few cases of congenital atresia of the submandibular gland duct have appeared in the literature and only one reports no surgical intervention to date.

**Case series:** In the present study reports two cases of babies with clinical findings demonstrating swelling well-defined in the region of the oral floor, slightly translucent bluish color, painless to palpation, without finding stones or salivary drainage and apparently with no perforation of the submandibular gland duct. Both patients were kept under observation, later presenting spontaneous regression.

**Conclusion:** This study highlights the importance of conducting a thorough clinical examination in addition to showing that the use of complementary exams may not be essential.

#### KEYWORDS

Congenital Abnormalities; Submandibular Gland; Salivary Ducts; Submandibular Gland Diseases.

#### RESUMO

**Introdução:** A atresia congênita do ducto da glândula submandibular é uma condição rara, originada durante o processo embriogênico, caracterizada por edema na região do assoalho bucal. Alguns casos de atresia congênita do ducto da glândula submandibular têm aparecido na literatura e apenas um relata nenhuma intervenção cirúrgica até o momento.

**Série de casos:** No presente estudo relata-se dois casos de bebês com quadro clínico demonstrando aumento de volume bem definido na região do assoalho bucal, cor azulada levemente translúcida, indolor à palpação, sem encontrar cálculos ou drenagem salivar e aparentemente sem perfuração do ducto da glândula submandibular. Ambos os pacientes foram mantidos em observação, apresentando posteriormente regressão espontânea.

**Conclusão:** Este estudo destaca a importância da realização de um exame clínico minucioso, além de mostrar que a utilização de exames complementares pode não ser imprescindível.

#### PALAVRAS CHAVE

Anormalidades congênicas; Glândula submandibular; Ductos salivares; Doenças da glândula submandibular.

## INTRODUCTION

Congenital atresia of the submandibular gland duct is a rare anomaly due to a failure in the canalization of the respective duct during the embryogenic process. During fetal development, ectodermal tissues of the oral mucosa begin to invaginate, originating salivary glands.<sup>1</sup> In the sixth embryonic week, the medial paralingual sulcus is formed as a solid cord of cells. In this phase, the epithelial tissue empties from distal to proximal, originating the patent duct.<sup>1,2,3</sup> The three anterior quarters of the paralingual groove form the submandibular duct and its posterior fourth, the submandibular gland itself.<sup>2</sup> Incomplete emptying of epithelial tissue, caused by a defect in the embryonic process, results in the absence of the submandibular duct, as well as its orifice.<sup>4</sup>

Congenital atresia of the submandibular gland's excretory ducts mainly affects males.<sup>5</sup> Such condition prevents the drainage of saliva, generating an accumulation of fluid in the floor of the mouth, which results in a swelling with characteristics similar to the ranula.<sup>6</sup> It can generate formation of salivary gland retention cysts, difficulty in feeding and speech development, being a lesion of relevant importance in the baby's development.<sup>7</sup> The lesion can be treated using different surgical methods, including simple incision, excision of the terminal portion of the submandibular duct with ductoplasty, duct stent and marsupialization.<sup>3, 4, 8-10</sup> Notably, there is a few cases report of congenital atresia of the submandibular duct in which there was spontaneous regression. This approach has advantages over the surgical technique, since it generates less discomfort for the child, there is less chance of intraoperative and postoperative complications.

The objective of this study is to present the report of two clinical cases, following the rules of the Case Report Guidelines (CARE)<sup>11</sup>, of a rare condition known as congenital atresia of the submandibular gland duct, in which both cases, the lesion regressed spontaneously. In addition, to compare the present cases with those already published in the literature and emphasize the importance of pediatric dentistry in recognizing this alteration to avoid errors in diagnosis and unnecessary treatments.

## CASE REPORT

### Case one

Female patient, three months old, melanodermic, was referred by the pediatrician to the Stomatology Clinic for analysis of volume increase in the right region of the floor of the mouth. During examination, the mother reported that, at birth, she noticed a swelling in the child's sublin-

gual region and that this lesion did not impair the child's breastfeeding. The doctors reported that the anomaly would completely regress without any intervention; however, the lesion persisted until the date of the dental clinical evaluation, which occurred four months after the medical evaluation. In addition, the child had no history of trauma and had good general health.

Medical and family histories were not contributing. The extraoral examination, no alterations were observed (Figure 1A). The intraoral examination revealed an increase in volume, well defined, of bluish color slightly translucent, soft consistency and painless on palpation, located in the right region of the floor of the mouth. There was no salivary drainage and there was an absence of perforation of the submandibular gland duct (Figure 1B). The diagnostic hypotheses were ranula and congenital atresia of the submandibular gland duct. As the child did not show any sign of discomfort, we choose to keep him in follow-up, and the child's mother was instructed on the lesion and also on the child's oral hygiene methods to prevent infections.

### Follow-up

The child returned to the clinic 11 months later, with complete spontaneous regression of the lesion (Figure 1C). The mother reported that she was unable to attend the scheduled return, but said that, three months after the first consultation, the lesion had regressed. After two years and nine months of follow-up, the patient did not present recurrence of the lesion. The mother's perception of the proposed treatment was positive. According to the report, the mother was satisfied since there was no need for surgical intervention, avoiding possible risks for the child.

### Case two

Male patient, five months old, melanodermic, was referred by a dental surgeon to the Stomatology Clinic for evaluation of volume increase in the left region of the floor of the mouth. According to the mother's report, the lesion had been present in the oral cavity since birth, and the child showed no signs of pain or discomfort.

Medical and family histories were not contributing. On extraoral examination no changes were noted. The intraoral examination revealed a well-defined volume increase of bluish color, slightly translucent, soft consistency, in the left region of the floor of the mouth. No salivary drainage was observed and there was no perforation of the submandibular gland duct (Figure 2A). In view of these clinical aspects, the diagnosis was



**Figure 1.** Case 1 (A) Extraoral appearance showing normality. (B) Intraoral aspect demonstrating an increase in volume of translucent color, well delimited, on the floor of the mouth, right side. (C) Intraoral feature showing spontaneous lesion regression, after 11 months of follow-up.

congenital atresia of the submandibular gland duct. The mother of the child was instructed on the nature of the lesion and the possibility of spontaneous regression, and she agreed with the proposed conduct for monitoring the lesion. In addition, she was instructed on oral hygiene to prevent infections.

*Follow-up*

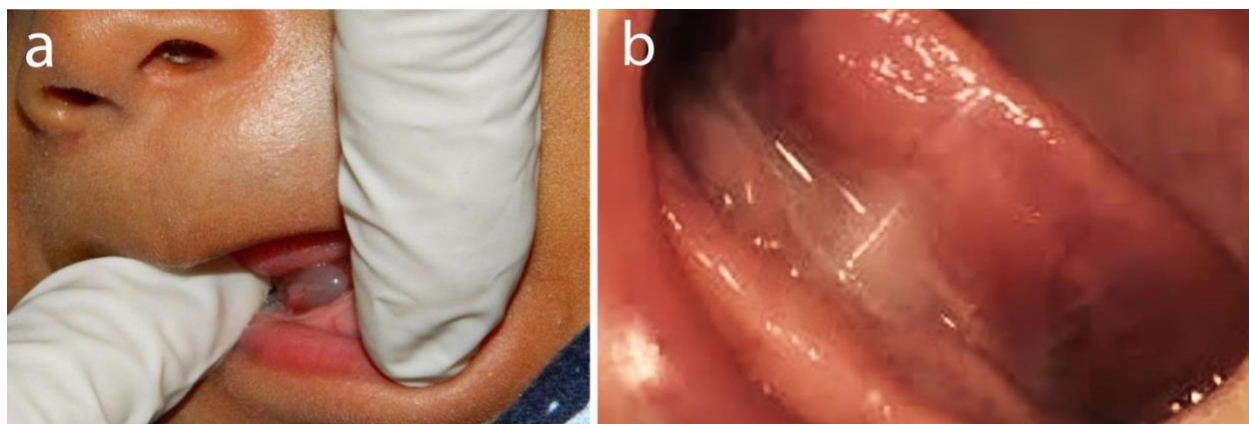
After three weeks of follow-up, complete spontaneous regression of the lesion was observed (Figure 2B). The child remained without signs of recurrence of the lesion after two years and two months of following up. The patient's mother's perception of the proposed treatment was positive, as in the first case.

**DISCUSSION**

Congenital atresia of the submandibular gland duct was

first described by Scher in 1955<sup>8</sup>, being a condition that is clinically similar to other congenital pathologies in the floor of the mouth, including lymphatic malformation, teratoma, dermoid cyst, thyroglossal duct cyst and heterotopic gastric cyst.<sup>3</sup> The present case series is the second to report the spontaneous regression of this lesion, whose clinical conduct was observation in both cases.

In a review of the literature, only 31 published cases of congenital atresia of the submandibular gland duct were found<sup>3-10, 12-27</sup>, being the majority affecting males with 21 cases, females with only seven cases, and three cases were not informed. The age in the reports ranged from 12 hours to one year and ten months, in agreement with the age range of both cases in this study. In most cases, the lesion affected the left side of the floor of the mouth (13 cases) and the most common diagnostic method was based on a thorough clinical examination (14 cases), similar to the present study (Table 1).



**Figure 2.** Case 2 (A) Intraoral appearance showing a bubble on the floor of the mouth near the opening of the submandibular gland duct, on the left side. (B) Intraoral aspect, showing complete regression of the lesion, after three weeks of follow-up.

**Table 1:** List of studies in the literature on diagnosed cases of Congenital atresia of the submandibular gland duct.

AUTHOR/ YEAR	COUNTRY	AGE	SEX	LATERALITY	DIAGNOSTIC METHOD	TREATMENT
<b>Scher and Scher (1955)<sup>8</sup></b>	-	25 days	Male	Bilateral	-	Incision
<b>Redpath (1969)<sup>15</sup></b>	England	12 hours	Female	Right	Histopathological examination	Marsupialization
<b>Foretich (1973)<sup>9</sup></b>	-	2 months	Male	Bilateral	-	Excision with stent, ductoplasty
<b>Hoggins and Hutton (1974)<sup>16</sup></b>	England	Not reported	Female	Bilateral	Thorough clinical examination	Incision
		13 days	Male	Left	Thorough clinical examination	Incision
<b>Addante (1984)<sup>13</sup></b>	Germany	1 year	Male	Left	Histopathological examination	Incision and Marsupialization
<b>Pownell et al. (1992)<sup>10</sup></b>	EUA	Not reported	Female	Bilateral	Thorough clinical examination	Incision and Marsupialization
		2 months	Female	Right	Thorough clinical examination	Incision and Marsupialization
		Not reported	Not reported	Left	Thorough clinical examination	Marsupialization
		1 month	Not reported	Left	Thorough clinical examination	Marsupialization and ductoplasty
		1 week	Male	Left	Magnetic resonance imaging	Excision
<b>Steelman et al. (1998)<sup>14</sup></b>	EUA	1 day	Male	Right	Thorough clinical examination	Spontaneous regression after 5 weeks
<b>Kawahara et al. (2000)<sup>17</sup></b>	Japan	1 year, 10 months	Male	Right	Histopathological examination and Magnetic resonance imaging	Incision and Marsupialization
<b>Amin and Bailey (2001)<sup>3</sup></b>	England	3 months	Male	Left	Thorough clinical examination	Incision
		2 days	Not reported			
<b>Hoffrichter et al. (2001)<sup>6</sup></b>	EUA	3 months	Male	Bilateral	Computed tomography	Marsupialization of the left duct and Incision of the right duct
<b>Wada et al. (2004)<sup>18</sup></b>	Japan	1 day	Male	Right	Ultrasonography and Histopathological examination	Marsupialization
<b>Walker (2005)<sup>4</sup></b>	Australia	2 months	Male	Left	Thorough clinical examination	Incision and Marsupialization
<b>Capaccio et al. (2007)<sup>19</sup></b>	Italy	4 months	Male	Bilateral	Videosialoendoscopic and ultrasonography	Incision

<b>Pal and Abdulla (2007)<sup>20</sup></b>	Saudi Arabia	1 day	Male	Right	Thorough clinical examination	Incision
<b>Ulualp et al. (2007)<sup>7</sup></b>	EUA	2 months	Male	Bilateral	Magnetic resonance imaging	Incision and Marsupialization
<b>Basaran et al. (2009)<sup>21</sup></b>	Turkey	2 months	Male	Left	Computed tomography and Histopathological examination	Marsupialization
<b>Rosow et al. (2009)<sup>12</sup></b>	EUA	5 months	Male	Left	Thorough clinical examination	Excision of the duct papilla
		1 month	Male	Not reported	Thorough clinical examination	Excision of the duct papilla
<b>Ellegård and Kjellmer (2010)<sup>22</sup></b>	Sweden	2 months	Male	Bilateral	Thorough clinical examination	Marsupialization
<b>Mandel and Alfi (2012)<sup>5</sup></b>	EUA	4 months	Male	Left	Magnetic resonance imaging	Incision
<b>Aronovich and Edwards (2014)<sup>23</sup></b>	EUA	5 months	Female	Bilateral	Computed tomography	Incision and Marsupialization
<b>Hseu et al. (2016)<sup>24</sup></b>	EUA	3 months	Male	Left	Thorough clinical examination	Excision
<b>Tanaka et al. (2016)<sup>25</sup></b>	Japan	5 months	Female	Left	Magnetic resonance imaging and Histopathological examination	Marsupialization and sialoductoplasty
<b>Pontes et al. (2018)<sup>26</sup></b>	Brazil	4 months	Male	Left	Histopathological examination	Excision
<b>Prosdócimo et al. (2018)<sup>27</sup></b>	Brazil	1 month	Female	Not reported	Histopathological examination	Excision
<b>Present study (2021)</b>	Brazil	3 months	Female	Right	Thorough clinical examination	Spontaneous regression after 3 months
		5 months	Male	Left	Thorough clinical examination	Spontaneous regression after 3 weeks

For the conclusion of the final diagnosis, it is of utmost importance that the dentist makes a complete evaluation, along with a thorough physical examination and, if necessary, uses complementary exams in order to identify particular characteristics of each lesion. Clinically, congenital atresia of the submandibular gland duct presents itself as an increase in volume in the region of the floor of the mouth that can be mistaken mainly for a ranula.<sup>12</sup> Such diagnosis can be defined when considering the previous history of the lesion, clinical characteristics and the use or not of complementary exams. In the cases reported in the present study, complementary exams were not used, only a thorough clinical exam, showing the importance of the pediatric dentist's attention to congenital lesions that may appear in the baby's oral cavity.

It is notable that clinically the ranula presents an opening in the duct ostium; the milking movement presents salivary drainage; it has a more diffuse characteristic, extending up to the center of the floor of the mouth, due to mucin overflow; has a history of exacerbation and remission and may be associated with some trauma.<sup>12</sup> There are reports in the literature that state that bilateral clinical manifestation of the ranula is highly unlikely, unlike congenital atresia of the submandibular gland duct that can manifest bilaterally.<sup>6</sup> When the histopathology of the ranula is performed, it is possible to observe areas with accumulation of mucoid material with reaction of the granulation tissue, in addition to mononuclear inflammatory infiltrate.<sup>3</sup>

In contrast, congenital atresia of the submandibular gland duct does not clinically present an ostium opening, nor does it present any saliva drainage during milking. In addition, it presents well-defined, circumscribed limits, with an enlarged lobed conformation and is not associated with a history of trauma. As in the present cases, there was no saliva drainage during milking and history of trauma. Histopathological examination reveals a cystic cavity covered by a pseudo-stratified columnar epithelium, with ciliated edges and fine connective tissue, consisting of an enlarged salivary duct.<sup>13</sup> In addition to clinical and histopathological findings, the treatment approach adopted in both diagnoses is also divergent, since a simple incision in a ranula usually leads to recurrence of the lesion and the most appropriate treatment is excision of the lesion along with removal of the salivary gland involved.<sup>28</sup> In the reported cases, it was decided to carry out with the observation, consequently there was no need for histopathological exams to confirm the diagnosis, since the lesions spontaneously regressed. Inversely, the bullous lesion resulting from congenital atresia of the submandibular gland duct presents good results reported in the literature with simple incision or marsupialization.<sup>5</sup>

Investigations with the aid of complementary exams such as, magnetic resonance imaging, computed tomography, ultrasonography, video sialoendoscopy and histopathological analyses have been described in the literature for better understanding of the other differential diagnoses such as lymphatic malformation, teratoma, dermoid cyst, thyroglossal duct cyst and heterotopic gastric cyst.<sup>3</sup> In our quantitative search, we found 14 cases diagnosed with detailed clinical examination, eight with histopathological examination, five with magnetic resonance imaging, three with computed tomography, two with ultrasonography and one with video sialoendoscopy, and in some cases a combination of complementary exams was performed (Table 1). However, complementary examinations are not essential for the diagnostic conclusion, as the diagnosis is notably possible of being performed with clinical examination and previous history of the lesion, being a more conservative, less invasive and economical option for the patient<sup>3</sup>, as shown in the two cases presented here. Thus, the Pediatric Dentist may be the first professional to diagnose and manage the case when this lesion does not present physiological damage to the patient.

In the literature, satisfactory results can be observed without recurrence rates associated with treatments such as simple incision<sup>3</sup>, marsupialization<sup>6</sup>, stenting of the duct<sup>9</sup> and excision of the terminal portion of the submandibular gland duct<sup>12</sup>; with reports of preference for marsupialization<sup>12</sup>. In our review, 15 cases were treated

with an incision, 15 marsupialization, four excisions, two ductoplasty, two excisions of the duct papilla, one case spontaneously regressed, one sialoductoplasty, one excision with stent, and one or more techniques could be used in the same case (Table 1). However, there is a discussion of the possible observation for spontaneous regression of the lesion, avoiding intervention in cases in which children are not being harmed by difficulty in feeding, breathing or infection in the region.<sup>14</sup>

The cases reported in the present study have the common characteristic of presenting spontaneous regression in patients at six months of age, coinciding with the time when food introduction occurs in the child's life. It is believed that such an event can generate friction or damage the abnormality, generating overflow and consequent spontaneous regression. There is a discussion in the literature associating the suction movement resulting from breastfeeding with the stimulation of salivary drainage, naturally perforating the duct and causing the lesion to decrease.<sup>10</sup>

When carrying out a quantitative bibliographic review of the documented cases, it is possible to observe only one documented report that presented the lesion observation conduct, which spontaneously regressed afterwards<sup>14</sup>, different from other findings in the literature which opted for an immediate intervention procedure (Table 1). Such report provides a basis for choosing the clinical conduct performed in the two cases presented. Here we present two cases of congenital atresia of the submandibular gland duct in which there was no need for immediate intervention since the children involved were not experiencing any damage associated with the lesion and the follow-up for more than two years did not show a recurrence. It is notable that follow-up was the best option for patients with congenital atresia of the submandibular gland duct, since there is the possibility of spontaneous opening of the ostium of the submandibular gland duct resulting in the disappearance of the lesion. This can also impact caregivers as there is no need for any invasive procedures such as excision/surgical intervention. However, future studies with representative samples and with follow-up are needed to confirm the non-invasive clinical management of this type of lesion.

## CONCLUSION

In the present study we show two cases of congenital atresia of the submandibular gland duct which did not require surgical intervention, due to the spontaneous regression of the lesions. This conduct emphasizes the importance of conducting a thorough clinical examination on the part of dentists and shows the importance of pediatric

dentists in cases like this, as they are in direct contact with children and newborns.

In addition, this study shows that the use of complementary exams may not be essential when there is a well-conducted clinical examination and reinforces the importance of follow-up when there is no harm to the patient, especially in the case of babies.

## ACKNOWLEDGMENTS

The authors thank the patients and their parents for all their collaboration during dental treatment and for their consent to publish this work. We would also like to thank: Dr. Raimundo Cirilo Marques Neto for his contribution in case 2; Dr Jorge Esquiche León for his opinion on the diagnosis; the Universidade Federal dos Vales do Jequitinhonha e Mucuri, the Coordenação Brasileira de Ensino Superior, Ministério da Educação (CAPES) and the Fundação de Amparo à Pesquisa de Minas Gerais (FAPEMIG) for providing all the support for this work to be carried out.

## ETHICS APPROVAL

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the Helsinki Declaration of 1964 and its subsequent amendments or comparable ethical standards. The study was approved by the Research Ethics Committee of the Universidade Federal dos Vales do Jequitinhonha e Mucuri (Number: 3.707.743).

## AUTHORS' CONTRIBUTIONS

The author LOF was responsible for collecting patient data and writing the manuscript, as well as collecting all the necessary information for the literature review. The authors PSM and MWAG were responsible for contributing together with LOF in writing the work and reviewing it critically. The author MLRJ was responsible for critically reviewing the manuscript and reviewing the literature. The ATMM author was responsible for evaluating patients, collecting data, critically reviewing the manuscript and reviewing the literature. All authors discussed and approved the final version of this article.

## DECLARATION OF CONFLICT OF INTEREST

The authors declare no conflict of interest.

## SOURCES OF FUNDING

The study did not receive funding.

## CONSENT TO PARTICIPATE

Informed consent was obtained from those responsible for the children included in this case report.

## CONSENT FOR PUBLICATION

The guardians of the children signed the informed consent form regarding the publication of their data and photographs.

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