Sequelae of congenital oropharyngeal teratoma in the oral cavity of the child: case report

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Abstract

Congenital oropharyngeal teratoma is a rare tumor subtype with cells from the three germ layers, majorly being benign. Whereas teratoma develops early in intrauterine life, thus affecting the growth and development of adjacent structures, the purpose of this case report is to present the sequelae of this tumor in a child's oral cavity. Female child, 2 years and 3 months old referred by the otorhinolaryngologist for

dental evaluation. The mother´s main complaint was the the child could not close her mouth. According to the child's medical record, at 34th week of gestation, during the routine ultrasound examination, the presence of the anechoic tumor mass was observed without Doppler signal, presenting a 3.2cm x 2.4cm diameter outside through the oral cavity. At 17 and 32 days of life, respectively, the excision of the largest and residual tumor located on the floor of the mouth was performed. Upon anatomopathological examination, the tumor with was classified as a mature teratoma. In the intraoral examination, a deep and atresia palate incomplete cleft palate, bone growth in the region of the posterior alveolar ridge of the maxilla, and in the posterior region of the mandible on the left side, "V" shaped mandible and microglossia, crowding were observed dental agenesis of the lower lateral incisors, absence of caries lesions, gingivitis and enamel development defects. The child will remain under the regular dental pediatric follow-up. Several sequelae were observed in the child's oral cavity and the need for multi-professional follow-up after excision of congenital oropharyngeal teratoma.

Keywords: Teratoma, Oropharynx, Child, Preschool, Oral Cavity, Case Reports

1. Introduction

The word teratoma derives from the greek "teras", meaning "monster", while the suffix "oma" stands for "tumor" or "neoplasm". By definition, congenital oropharyngeal teratoma is a rare tumor subtype with cells from the three germ layers (ectoderm, mesoderm, and endoderm), majorly being benign[1]. Those originating from the hard palate, oral mucosa, thong, and chin, extending through the oral cavity, are called Epignathus[2]. Its etiology is unknown, however, the most accepted theory is that the teratoma would originate at the base of the skull, in cells in the region of the Rathke's pouch, growing in a disorganized manner[3]. Teratomas are associated with chromosomal abnormalities, early embryonic changes, or fetal syndrome and can be classified as mature and immature[4].

The occurrence in the oral cavity is 1:2000,000 live births, corresponding to approximately 10% of all teratomas, presenting a higher prevalence in females[4]. These tumors are characterized by rapid intrauterine growth and the indicated treatment is surgical[5].

Most studies[2,4,6-15] report some characteristics of teratoma and its removal, without describing the consequences in the oral cavity. Whereas congenital oropharyngeal teratoma develops early in intrauterine life, thus affecting the growth and development of adjacent structures, the purpose of this case report is to present the sequelae of this tumor in a child's oral cavity.

2. Case report

Female child, 2 years and 3 months old, white, attended the clinic of the Extension Project "Promotion of oral health for children with disabilities from zero to five years old", held at Hospital Odontológico da Faculdade de Odontologia da Universidade Federal de Uberlândia, accompanied by the mother and referred by the otorhinolaryngologist for dental evaluation. The mother´s main complaint was the the child could not close her mouth.

Initially, signatures of the consent form were requested for the anamnesis and clinical examination as well as the authorization to take photographs and footage for the production of didactic material. Then, the anamnesis was carried out with the collection of information about the child's pre-, trans- and postnatal and dental medical history through an interview. To complement the anamnesis, data were collected from the child's handbook and the discharge medical report provided by the mother containing summary data of the entire history since pregnancy. Later, in the intermission between the first and second dental consultations, data were collected from the child's medical record available at the Statistics Department of the Hospital de Clínicas da Universidade Federal de Uberlândia (HC-UFU), with prior authorization from the sector, for further clarification on the diagnosis, previous history, clinical findings treatment and evolution of the case.

The following information was obtained: the mother reported having become pregnant at 16 years of age, not needing medical care during pregnancy, although it was considered a risky pregnancy, and attended to ten prenatal consultations. According to the child's medical record, at the 34th week of gestation, during the routine ultrasound examination, the presence of the anechoic tumor mass was observed without Doppler signal, presenting a 3.2cm x 2.4cm diameter outside through the oral cavity.

The child is the first daughter, of a non-consanguineous couple, newborn at term, of elective cesarean delivery, performed at 39 weeks of gestational age, weighing 2,930 grams and measuring 47 centimeters in length, Apgar score in the first and fifth minutes of 8 and 8.

After birth, still, in the delivery room, nasotracheal intubation guided by bronchoscopy was performed, due to the size of the tumor and the possibility of risks to the child's life. Upon clinical examination, shortly after birth, the presence of two tumors was observed: a larger one, measuring approximately 3.2cm x 2.4cm in diameter obliterating the oral cavity with a similar aspect to the skin and mucosa previously seen on ultrasound, without the involvement of the nose, lips, and tongue; and a smaller one of approximately 1.0cm in diameter originating from the body of mandible, which had not been observed during the ultrasound examination.

Skull and sinuses tomography performed after birth, revealed maxillary disjunction with interposition of the pseudo maxilla, communication tumor with sphenoid bone, and bone defects.

Then, the child was referred to the Neonatal Intensive Care Unit and, after admission, had a sudden worsening due to hypertensive pneumothorax.

At 17 and 32 days of life, respectively, the excision of the largest and residual tumor located on the floor of the mouth was performed. The resection was performed respecting the safety margin and no components of malignancy were identified. Upon anatomopathological examination, the tumor with the largest dimension was classified as a mature teratoma, mostly containing adipose tissue, with areas of cartilaginous and bone consistency with structure in its interior compatible with a tooth. On the other hand, the mandibular specimen had a structure partially covered by smaller salivary glands surrounded by abundant adipose tissue, and the presence of spongy bone tissue containing hematopoietic marrow, without specifying whether it was mature or immature.

At postoperative follow-up, it was observed that the child had an open mouth posture, due to the process of upward growth of the mandible, preventing the suction movement and making oral feeding impossible, as well as lip and lingual protrusion. Hence, in the second postoperative week, indirect therapy was

started to stimulate the orofacial muscles and nasotracheal feeding. During the hospitalization period, the child had several complications, requiring supplemental O2 and performing a tracheostomy and gastrostomy. There are no reports in the medical record that the child breastfed and the mother was unable to report whether the child breastfeed or not.

After 60 days of hospitalization, with good postoperative evolution, the child was discharged and is undergoing multi-professional follow-up carried out by the Home Care Service in the city where she resides (São Gotardo, Minas Gerais), which includes: social worker, nursing technician, nurse, pharmacist, physiotherapist, physician, nutritionist and psychologist, and monitoring of regular consultations at HC-UFU.

Only at 5 months of age, an alpha-protein test was performed which obtained a value of 82.30 ng/mL. It was noted in the medical record that the child also had pituitary duplication and sickle cell trait.

Regarding the dental history, the mother reported that it was the first consultation and also that she was not instructed on brushing. Nonetheless, the mother reported that she performed her daughter's oral hygiene daily, with a brush and toothpaste that she does not know the brand of, at least three times a day, without flossing. Currently, the child's eating habits involve pasty meals and the probe is used only for liquids. Concerning non-nutritive sucking, the child never showed any habit and during the care, the presence of these was not observed. The mother was instructed on the relationship between sugary foods and dental caries. In the extra-oral examination, macrostomy and absence of lip sealing were observed (figure 1).

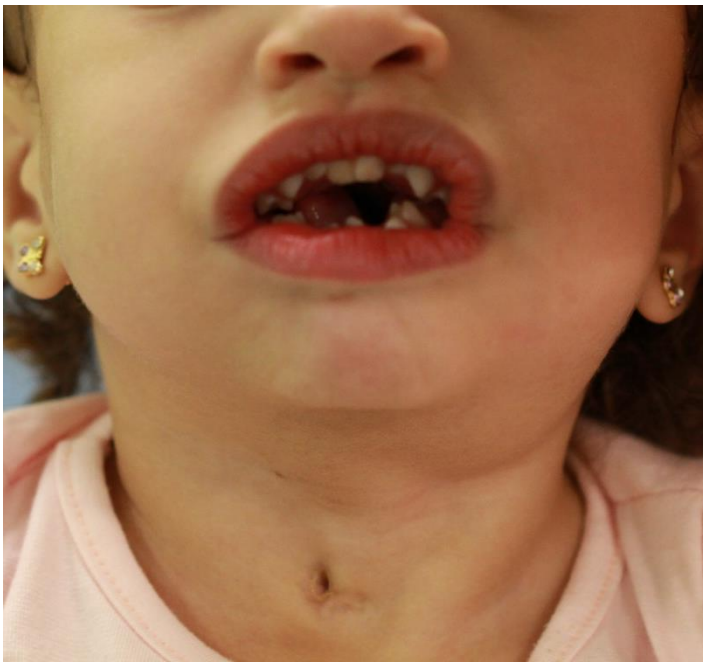

Figure 1 - Presence of macrostomy and absence of lip sealing.

In the intraoral examination, a deep and atresia palate incomplete cleft palate, bone growth in the region of the posterior alveolar ridge of the maxilla, and in the posterior region of the mandible on the left side, "V" shaped mandible and microglossia (figure 2), crowding were observed dental agenesis of the lower lateral incisors, absence of caries lesions, gingivitis and enamel development defects (figure 3).

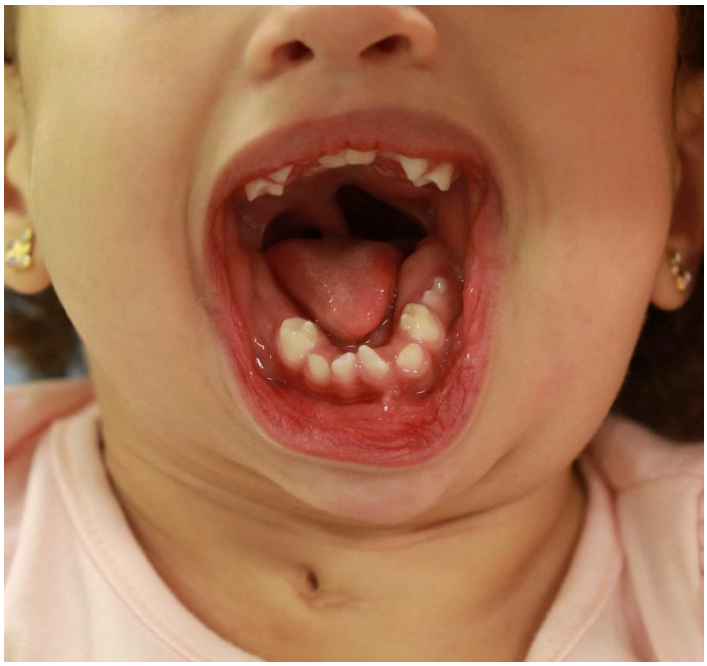

Figure 2 - Deep and atretic palate, incomplete cleft palate, bone growth in the region of the posterior alveolar ridge of the maxilla and in the posterior region of the mandible on the left side, "V" shaped mandible and microglossia.

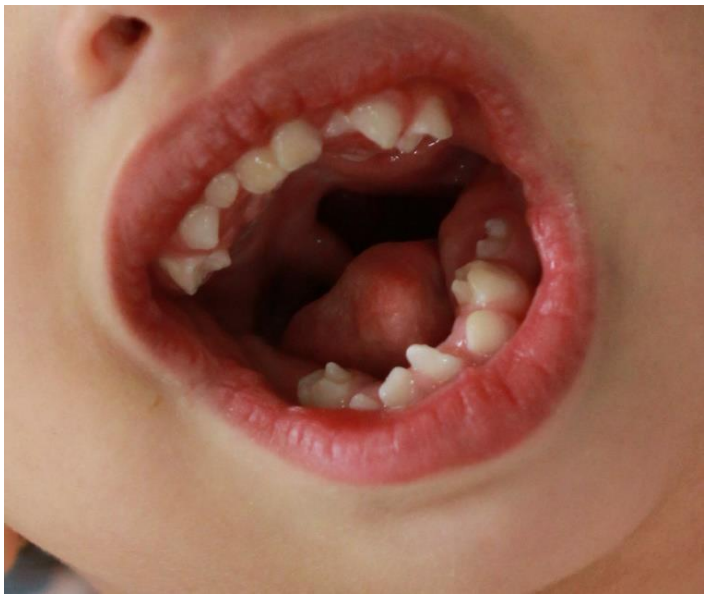

Figure 3 - Presence of dental crowding, agenesis of the lower lateral incisors, absence of caries, gingivitis, and enamel development defects.

Brushing was performed with a children's brush and a fluoride toothpaste with a concentration of 1,100ppmF- (Oral-B - Procter & Gamble, Cincinnati, Ohio, United States) and the use of assembled dental floss (Easy way bag - Powerdent, Barueri, São Paulo, Brazil). The mother was instructed on the amount and concentration of fluoride toothpaste, the brushing technique appropriate for her age, the number of

times a day, and the use of dental floss (figures 4A and B).

Figure 4A and B - Brushing with children's brush, children’s fluoride toothpaste, and flossing.

Throughout dental care, basic behavioral management techniques were used: talking-showing-doing, distraction, and positive reinforcement. The child was a collaborator in most of the care with short periods of crying.

At the end of the first consultation, a medical opinion was requested, which is a standard procedure in all consultations, carried out at the Extension Project Clinic "Oral Health Promotion for Children with disabilities from zero to five years old" to find out the general condition of the child.

At the second consultation, the mother presented the complete medical opinion, signed and stamped by the responsible physician, who described that the child has sequelae of oropharyngeal teratoma resected at birth, with incomplete cleft palate, and is currently being monitored by the otolaryngology, oncology team, pediatric surgery, and speech therapy. She can feed herself orally, and progressive weaning from gastrostomy is being carried out and, currently, general health conditions are good and there is no need for prophylactic antibiotic therapy for dental treatment.

In this session, professional prophylaxis was performed with the aid of a Robinson brush, rubber cup, and infant fluoride toothpaste with a concentration of 1,100ppmF- (Oral-B - Procter & Gamble, Cincinnati, Ohio, United States), obtaining clean surfaces to perform the procedure. The child had 15 deciduous teeth (51, 52, 53, 54, 61, 62, 63, 64, 71, 73, 74, 75, 81, 83 e 84). The data obtained were noted on the clinical record and radiographic examination was indicated for diagnostic completion. The child's behavior was collaborative and basic techniques of behavioral management, talk-show-do, distraction, and positive reinforcement were used.

The genetic evaluation was not found in the medical files, and according to the mother, the consultation with the geneticist doctor never took place, although in the discharge medical report the referral was made for evaluation of possible association with syndromes.

The child will remain under the regular pediatric and dental pediatric follow-up to prevent major oral diseases and referral for orthodontic correction at the appropriate time.

3. Discussion

Although congenital oropharyngeal teratomas are considered rare benign tumors, when present, they can cause several sequelae in the child's oral cavity, as observed in this case report, requiring long-term multidisciplinary follow-up to correct them.

This clinical case has many similarities with others found in the literature[2,6-8,10,11,15-19]. Regarding gender, most studies reported the presence of oropharyngeal teratomas in female children. Studies show a higher prevalence, in the proportion of 3:1, in girls than in boys, without justification for the difference between them. Other studies[3,12-14,21,22] described the presence of teratoma in male neonates.

In this case, it was not possible to define the etiology of congenital oropharyngeal teratoma. However, several hypotheses are considered[1,4,7]. The etiology is multifactorial ranging from abnormal migration of primordial cels in regions of the mediastinum or hypothalamus to abnormalities in embryonic development involving chromosomal changes that may include mosaicism with an inactive X chromosome ring, trisomy 13, X ring chromosome and X pentassomy, Aicardi syndrome, Pierre-Robin syndrome and even the abortive growth of a twin. Therefore, it is consiered extremely important that a genetic investigation be carried out to complement diagnosis and genetic investigation be carried out for the purpose of complementing diagnosis and genetic counseling according to the data consulted. According to the literature, the prenatal karyotype and the search for associated abnormalities are mandatory in all teratomas[1]. It appears in the discharge medical report that the child was referred to the geneticist. However, in the anamnesis, the mother reported that it has not yet been carried out during the consultation.

The presence of the larger tumor was found during the ultrasound exam in the 34th week of pregnancy, similar to other reports[9,10,12,16,19] in which the diagnosis also occurred in the third trimester. Other studies[7,8,20,23] reported the presence of teratoma in the second trimester of pregnancy, and in some[2,14,15], only after birth. According to Tonni et al.[1] and Kumar et al.[10], the presence of teratoma can be identified early in the 15th to 16th week of gestation, thus allowing delivery and postpartum maneuvers to be planned, increasing the child's survival. In the present report, a tumor was found before birth, which made it possible to plan the child's delivery and prepare for possible complications.

Regarding the treatment, Dohlman & Sjovall[24] recommend that it be performed in stages, with immediate resection of the tumor if possible and reassessment of the residual tumor if present, during the first weeks of life. In this case, the removal of the teratoma was performed early, o the 17th day of life, requiring removal of the residual teratoma two weeks later.

The tumor described in this case was classified anatomically and pathologically as mature, as well as the tumors reported by most authors[2,4,6,9,11,12,14,16,19,21,22]. However, even though it is an uncommon occurrence, Kumar et al.[7] and Chung et al.[8] reported cases of immature teratoma. According to Sauter et al.[25], there may be the occurrence of malignant epicanthus-type teratomas, however, this fact is considered rare.

Fetal manifestations are described in approximately 6% of cases, the most common being bifid tongues, left ventricular hypoplasia, pulmonary hypoplasia, hydrops, hypertelorism, imperforate anus, micrognathia, glossoptosis, cleft palate, and macrostomy[3,7,17,19].

In this clinical case, two alterations mentioned in the literature were observed, such as macrostomy[19] and cleft palate[11,16-19,21-23]. The shape of the oral cavity corroborates with the suggestive diagnosis for

macrostomy, a rare congenital anomaly understood as an enlargement of the mouth in the region of the oral commissures which may present itself unilaterally or bilaterally and the reported incident varies from 1:60,000 to 1:300,000 live births[26].

This anomaly is the result of a failure in the fusion of the maxillary and mandibular processes of the first branchial arch, which occurs around the seventh week of pregnancy. She needs surgical treatment and the correction is indicated between 3 and 12 months of life so that there is no impairment of the speech process[26,27]. In the consulted literature, only Teixeira et al.[19] described a case of macrostomy associated with congenital oropharyngeal teratoma that was corrected early, at [7] months of age. So far, the child has not undergone macrostomy correction. The mother was unable to report the reason why it had not yet been corrected. Nonetheless, the child is being accompanied.

The cleft palate, among craniofacial malformations, is considered the most common. This can be explained by the fact that this malformation occurs during intrauterine life, more precisely until the 12th gestational week, that is, at the end of the embryonic period, preventing the fusion of the nasal septum with the bilateral palatal processes[28]. In several reports[11,16-19,21-23], the presence of cleft palate was observed. Carreirão & Pitanguy[29] recommend that surgical intervention for the correction of cleft palate should occur at around 1 to 1.5 years of age, aiming to achieve better results due to the easier identification of anatomical structures and the age being the period acquisition of language function. In agreement with the literature, primary palatoplasty was performed to correct the cleft palate when the child was 1 year and 5 months old, according to the medical record, the surgery was successful, but a second one will be necessary to close the fissure.

The presence of microglossia, a developmental change, was also observed. So far, there is no conclusive etiology for the appearance of this alternation[30]. No report was found on the association between congenital oropharyngeal teratoma and microglossia. Regarding this alternation, there is no specific treatment and when it is indicated, the severity and nature of the condition must be considered. The recommended treatment is the follow-up with the speech therapist, orthodontist, and, if necessary, a surgical procedure[30]. The child is undergoing speech therapy and will need orthodontic correction later.

As described in the medical opinion, the child is being assisted by several health professionals. This opinion is always requested in the first consultation since its content explains in detail the procedure that will be performed and the drugs of choice so that the medical team can guide the dentistry about the recommendations regarding the safe care of the child. In this case, the child did not present any alteration that justified previous medication and/or prevent dental treatment.

One aspect worthy of attention is that the child attended to her first dental consultation when she was almost two and a half years old. The current recommendation is that the first examination should be performed at the time of the eruption of the first tooth and no later than 12 months of age. Fortunately, she had no caries lesions. The absence of the lower deciduous lateral incisors was also observed. The child will be accompanied by the pediatric dentist, on periodic return visits, for clinical examination assessment of growth and oral development, risk of caries, professional prophylaxis, topical fluoride applications, and sealing of pit and fissure when indicated. In addition, evaluation and/or treatment or referral for correction of malocclusion will be provided. It should also be noted that the strategies of the dental team seek to enable maneuvers that improve the child's quality of life by correcting the sequelae of congenital oropharyngeal teratoma, in a multidisciplinary way involving facial-maxillary surgery, orthodontics, and pediatric

dentistry.

4. Conclusion

Several sequelae were observed in the child's oral cavity and the need for multi-professional follow-up after excision of congenital oropharyngeal teratoma.

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