



CASE REPORT

REVISED Case Report: Severe acute respiratory distress by tracheal obstruction due to a congenital thyroid teratoma.

[version 2; referees: 1 approved, 1 approved with reservations]

Previously titled: Case Report: Severe acute respiratory distress by tracheal obstruction due to a congenital thyroid teratoma: a case report and literature review.

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Abstract

Congenital teratoma is a rare condition and is a germ cell tumor composed of elements from one or more of the embryonic germ layers and contain tissues usually foreign to the anatomic site of origin. We report a case of a neck tumor diagnosed during pregnancy, initially thought to be a goiter. After birth the neck mass kept growing until it compressed the trachea and produced respiratory failure. The infant had a difficult tracheal intubation because of the compressing mass. The staff decided to surgically remove the neck mass. After that, the infant became eupneic. The histological analysis showed a mature teratoma with no atypias.

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REVISED Amendments from Version 1

This new version of the manuscript corrects one author name (Eliana Carla Armelin Benites) and removes from the title "case report and literature review", since it is redundant, as it is already at the beginning of the title.

See referee reports

Introduction

Congenital thyroid teratoma is a rare condition^{1,2}. We report a case of an infant with a neck mass diagnosed by ultrasound during pregnancy which was initially supposed to be a congenital goiter. Two doses of levothyroxine were administered into the amniotic fluid. The goiter kept growing after birth until it caused severe respiratory distress by compressing the trachea, necessitating immediate tracheal intubation. The tumor was surgically resected and the patient went eupneic for the first time in his life. The histological analysis demonstrated a mature teratoma with no atypias. Thyroid hormone substitute therapy was started and the infant is thriving well.

Case report

A 2-months-and-20-days-old Brazilian white male infant weighing 4.2 kg was admitted to the pediatric intensive care unit of our hospital (Santa Catarina Hospital, São Paulo, Brazil) in acute respiratory distress and was immediately intubated and placed in mechanical ventilation.

From a routine ultrasound during pregnancy, the fetus had been diagnosed with a cervical mass, considered initially to be a goiter (Figure 1) by doctors at another institution. Family history of the mother uncovered a cousin with hypothyroidism. The mother was previously healthy, but after diagnosis of the cervical mass of the fetus, she was tested for thyroid hormones and had hypothyroidism diagnosed during pregnancy (TSH: 5.0 mUI/mL – normal: 0.2 to 3.0 mUI/mL; free T4: 0.7 ng/dL – normal: 0. To 1.3 ng/dL; antithyroglobulin antibodies: 65 U/mL – normal: inferior to 60 U/mL and thyroid antiperoxidase antibodies: 166 UI/mL – normal: inferior

to 9 UI/mL). Two single doses of 200µg of levothyroxine were administered into the amniotic fluid, one during the 28th and one during the 31st week of pregnancy, in order to treat the supposed fetal thyroid hormone deficiency. Chorioamnionitis appeared after the second levothyroxine administration which triggered a premature cesarean birth which was undertaken in the other hospital. The premature newborn had sepsis due to maternal infection (chorioamnionitis) and remained in mechanical ventilation for 10 days. After tracheal extubation, he remained in nasal continuous positive airway pressure (CPAP) for 7 more days, and after that was kept on oxygen therapy for 10 days. He was discharged from the hospital 50 days after birth, still presenting with a laryngeal stridor that was attributed to tracheal malacia by the doctors that initially treated the patient.

After hospital discharge, he was observed by a pediatric endocrinologist who started research on thyroid disorders. Meanwhile, the infant maintained a euthyroid state, receiving no treatment, waiting for more investigation on the cause of the neck mass. However, the cervical mass kept visibly growing, was palpable and the infant presented a laryngeal stridor that was still attributed, by the pediatrician who followed the infant, to laryngomalacia. In the few days preceding hospitalization at our institution, the infant became increasingly dispneic each day, as related by his mother. One day, after choking and vomiting during breastfeeding he became hypotonic and went into acute respiratory distress.

He was admitted to our pediatric intensive care unit 25 days after he had been discharged from the other hospital, and was immediately intubated. An X-ray showed a small amount of interstitial infiltrate, compatible with aspiration pneumonia. However, the respiratory distress was attributed mainly to an upper airway obstruction. It was difficult to tracheally intubate the infant; only an uncuffed 2.5 mm endotracheal tube (ETT) was able to be inserted into the trachea and it was difficult to place this in the right position. The X-ray after intubation showed the ETT in a high position and the trachea displaced to the right (Figure 2). Magnetic resonance imaging (MRI) revealed the extent of the cervical mass and its compression on the trachea, and the latter's subsequent displacement (Figure 3a and 3b).



Figure 1. Prenatal ultrasound showing the neck mass.

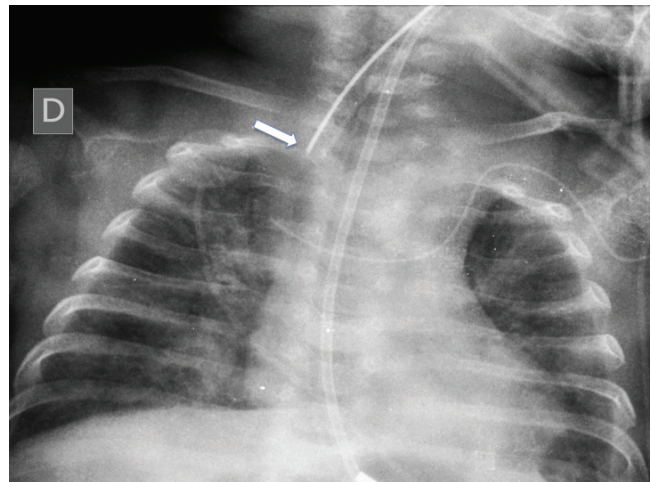


Figure 2. Endotracheal tube displaced to the right position.

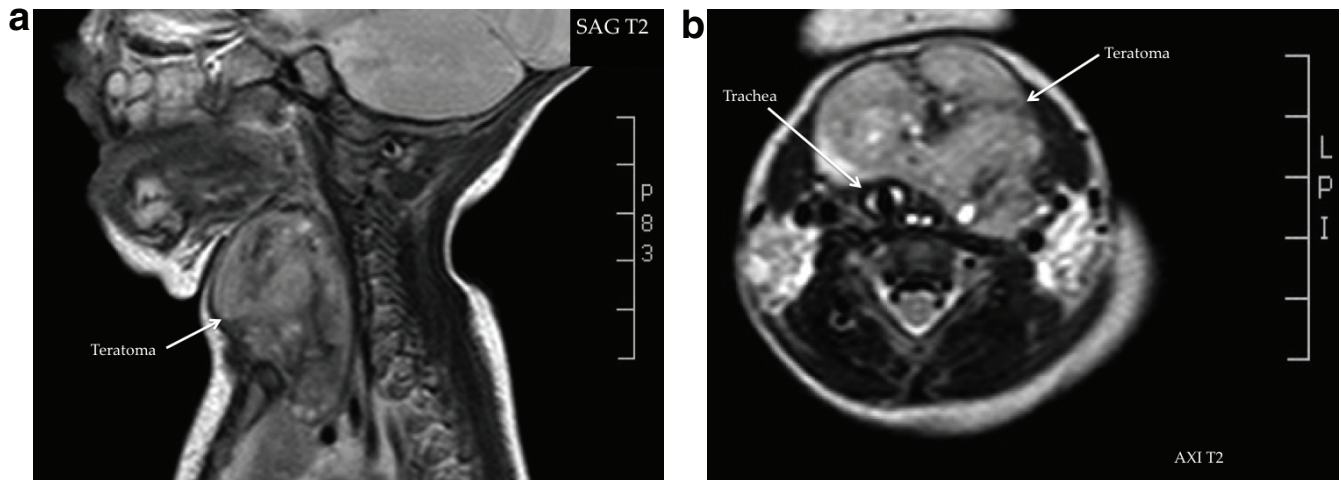


Figure 3. a. Sagittal MRI (T2) of the neck showing the teratoma. b. Axial MRI (T2) of the neck showing the teratoma and the tracheal displacement.

Meanwhile, we started investigation into the cause of the neck mass and performed blood tests on the infant: thyroid hormones were in the normal range (free thyroxine (T4): 1.3 ng/dL and thyroid-stimulating hormone (TSH): 4.7 ng/dL). Calcitonin levels, for investigations into potential malignancy, were normal (calcitonin: 21 pg/mL), as was the alpha-fetoprotein: 505 µg/L.

We decided to remove the cervical mass, since it was causing the tracheal obstruction. The surgery lasted 35 minutes and was uneventful. The mass was well circumscribed and could be easily dissected, weighed 20 grams and measured 33×61×45 mm (Figure 4). The infant returned from surgery in good condition. A bronchoscopy was performed the next day after surgery, during tracheal extubation, which revealed no malacia or any other disorders on the trachea or the upper respiratory tract. The patient has been eupneic since then. The histological analysis revealed a mature teratoma with no atypias or signs of malignancy.



Figure 4. The resected benign teratoma.

Levothyroxine was started (25µg, once a day) as thyroid hormone substitute therapy and the infant is thriving well according to the pediatric endocrinologist that continues following the patient.

Discussion

Teratomas originate from multipotent primitive germ cells and result in different tissues, diverging from the anatomical site of origin^{2,3}. They are most common during early childhood and the most common location is the sacrococcygeal region in children and the gonadal region in adults²⁻⁴. The frequency of these embryonic tumours is about 1:20,000–40,000 live births. However, only 1.5% to 5.5% of all pediatric teratomas are placed in the neck region. These tumours are usually solitary, with no other associated congenital malformations or chromosomal abnormalities⁴. Although 95% of all teratomas are benign, the cervical teratomas if not properly treated, lead to death in 80% of the cases due to obstructive respiratory distress³⁻⁵.

In this case, the acute clinical presentation of the neck mass with severe respiratory distress, needing ready intervention and immediate tracheal intubation should alert all pediatricians to the risk of these neck masses, and consider it as a potentially fatal case. In the presented case, surgical removal of the neck mass was both diagnostic and therapeutic.

Conclusion

Thyroid teratoma is rare in infants, it is usually benign, and can cause airway compression depending on the site and size of the mass. The likelihood of a malignant thyroid teratoma is low in infants, however it could be fatal by causing upper airway obstruction. Therefore, surgical resection is required both for diagnosis and treatment. If the surgical removal is a success, the long-term outcome and quality of life should be good⁵.

Consent

Written informed consent was obtained from parents of the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the editor of this journal.

Author contributions

JCJ drafted the first manuscript. UT is the pediatric surgeon and helped with the decision of resecting the tumor. FML did the endocrinological research and helped with the draft. ECB is from the

pediatric oncology team and helped with the diagnosis and the draft. WK, JHI and PRM contributed to treating the patient. WBC revised and edited the manuscript. All authors have read and approved the content of the final manuscript.

Competing interests

The authors declare that they have no competing interests.

Grant information

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Daniel Garros

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The authors describe an interesting case, with a positive outcome despite some delay in diagnosis. The title and the description of the case are all well written.

However, the **discussion and the conclusion** require more work. This case brings up an important and not so rare clinical scenario in Paediatric Critical Care, i.e. the difficulties inherent to extrinsic airway compression by a tumour. The authors, on their discussion, should take advantage of this case and discuss the approach to airway extrinsic compression, the use of extra-corporeal life support (ECLS) as a back-up plan, who should be present in such situations (ENT, anaesthesia), etc... The clinician could barely secure the child's airway, and the child survived and was able to have the teratoma resected. But it could have been a disastrous outcome. There are protocols for such scenarios, and they should be mentioned and described for the reader to be prepared when facing with similar situations

I have read this submission. I believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Competing Interests: No competing interests were disclosed.

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Coletti *et al* described in a case report of severe acute respiratory distress by tracheal obstruction due to congenital thyroid teratoma. This case report adds knowledge to the literature, not only by tumor description, but also the clinical management of the case.

Tumor description is interesting, however I believe that the authors should also discuss in this article the clinical management of malacia in premature, since this is very common in neonatal clinic. I believe that this discussion would make the most interesting article and is also an opportunity to develop the

discussion of the article, which I believe is short. For this reason there are some points which need to be considered and are necessary clarifications

Add data:

1. The patient was born with what gestational age?
2. After birth was not performed control with imaging for the mass?
3. What are the results of thyroid hormones tests after birth?
4. The staff investigated other causes for laryngeal stridor before discharge?

Suggestions:

The discussion could discuss the differential diagnosis of stridor. The staff not investigated a patient with stridor, attributing the malacia. Many patients in neonatology have stridor and I believe that this article could show that there is need for investigation in stridor, because sometimes the differential diagnosis can be serious pathologies.

Case reports are interesting, but I believe it should have a good discussion of the various aspects involved in the pathology, in which case I would like that you develop the discussion and mainly develop the discussion about the differential diagnosis.

We have read this submission. We believe that we have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.

Competing Interests: No competing interests were disclosed.

Author Response 10 Sep 2015

Jose Colleti Junior, Hospital Santa Catarina, Brazil

I would like to thank Dr Martinez and colleague for the remarks and suggestions above.

Answering the questions regarding this case report:

1. The patient was born with 31 weeks of gestational age, immediately after the second administration of levothyroxine in the amniotic fluid due to a chorioamnionitis.
2. After birth new imaging control was done in the previous hospital and in our medical center (figure 3).
3. After first hospital discharge, the patient was followed by a pediatric endocrinologist who asked for thyroid hormone tests which resulted normal for the age (TSH=1.8 mUI/mL – normal: 0.8 to 6.0 mUI/mL; free T4: 0.9 ng/dL – normal= 0.7 to 1.5 ng/dL).
4. Since the patient was born in other hospital we could not know if the staff did any other investigation for other causes of laryngeal stridor. In our medical center, the patient was admitted in a critical clinical status and the surgery was promptly performed.

I hope I have clarified your doubts and concerns about this case report. Thank you for the review.

Competing Interests: I declare no competing interests to disclose.