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A 3-WEEK-OLD NEWBORN WITH BILATERAL PSEUDOTUMOR OF THE STERNOCLEIDOMASTOID MUSCLE: A CASE REPORT

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3 კვირის ახალშობილი სტერნოკლეიდომასტოიდური კუნთის ორმხრივი ფსევდოსიმსივნით: შემთხვევის მოხსენება

მაღალი ტექნოლოგიების სამედიცინო ცენტრი, საუნივერსიტეტო კლინიკა; თბილისის სახელმწიფო უნივერსიტეტი, თბილისის სახელმწიფო სამედიცინო უნივერსიტეტი

რეზიუმე

მკერდლავიწდვრილისებრი კუნთის ფსევდო ტუმორი, ლიტერატურაში სხვადასხვა ავტორების მიერ ასევე წოდებული ფიბრომატოზი colli-ს სახელწოდებით, არის იშვიათი დაავადება და პროცესი ლოკალიზდება მკერდლავიწდვრილისებრი კუნთის შუა ან ქვედა ნაწილში. ფსევდო სიმსივნე იწოდება იმის გამო, რომ იგი არ წარმოადგენს ჭეშმარიტ ავთვისებიანი მიმდინარეობის პროცესს. იგი ვლინდება როგორც კონგენიტალური პათოლოგია ფიბროზული ქსოვილის სიჭარბით და სიმსინისმაგვარი ზრდით მკერდლავიწდვრილისებრი კუნთის სიღრმეში. უფრო ხშირად წარმოდგენილია ცალმხრივად მიმდინარე პროცესის სახით. ბილატერალური მდებარეობით ძალზედ იშვიათია. ჩვენ სტატიაში განვიხილავთ მკერდლავიწდვრილისებრი კუნთის ბილატერალური სიმსივნის უიშვიათეს შემთხვევას 3 კვირის ახალშობილში. დიაგნოზზე ეჭვის მიტანა ხდება კლინიკური მონაცემების საფუძველზე და დასტურდება მაგნიტურ რეზონანსული კვლევით

Pseudotumor of the sternocleidomastoid muscle, also called fibromatosis colli, sternocleidomastoid tumor of infancy (STOI) by some authors [1,2], is a rare condition [3]. The process is localized in the middle or lower part of the pectoralis major muscle. It is called a pseudotumor because it does not represent a true neoplasm. It manifests as a congenital pathology with an excess of fibrotic tissue and a tumor-like growth in the depth of the pectoralis muscle. The incidence of the disease is 0.4% [4] in live-born babies. In 75% of cases, it presents in a unilateral form and, according to literature data, is mainly localized in the right pectoral muscle [7,8]. It is more common in boys than in girls [3,7,8]. Bilateral involvement is especially rare [2,5,6]. In the literature, abnormal childbirth is mentioned in 50% of cases [6,8]. Ultrasonography [4,12] and MRI are considered the primary diagnostic studies. We report a rare case of bilateral pectoralis major muscle tumors in a 3-week-old infant.

A 3-week-old newborn was brought to our clinic by the emergency services with a suspected diagnosis of lymphadenitis. The newborn was from the 8th physiological pregnancy and delivery, born at term, weighing 3200 g and measuring 51 cm in length. The infant was breastfed.

From the medical history, it is noteworthy that a small formation (initially considered to be a lymph node) was observed at 10 days of age by family members and later by the doctor. For the last 4 days, there had been a sharp increase in the size of this formation, which was followed by difficulty in breathing. Specifically, noisy, labored, and frequent breathing was observed, which worsened during exertion and was accompanied by feeding difficulties. Due to these symptoms, the patient was transferred to our clinic.

Upon admission to the clinic, the patient's condition was assessed as severe, with clinical signs of acute respiratory distress developing against the background of the underlying condition. Tachypnea, chest retractions, and the use of accessory muscles for breathing were evident. Prolonged difficult inspiration and inspiratory stridor were present. Bilateral formations on the anterolateral surface of the neck were notable. These masses were localized in the projection area of the pectoralis major muscle on both sides, measuring 3 cm in size, dense, and immobile. They were not lymph nodes. Palpation was not painful. Torticollis with the head turned to the left was observed.

The patient underwent an ultrasound examination of the soft tissues of the neck and an MRI examination with contrast: The neck is deviated to the left. In the right supraclavicular fossa, a somewhat hardened structure, not sharply demarcated, measuring up to 30 mm in size, with weak, uneven contrast enhancement is observed. On the contralateral side, at the same level, the soft tissues of the neck are easily deformed, revealing an unclearly defined area of similar structure up to 20 mm in size. The described process leads to lateral deviation and deformation of the thyroid gland, without apparent focal damage. There are no reliable MRI signs of the process spreading to the major blood vessels. Correlation with MRI data (STIR sequence) is highly consistent with the diagnosis - Sternocleidomastoid tumor of infancy (STOI). Morphological verification is recommended for unambiguous assessment.

Pulmonary findings: No free fluid is seen in the bilateral pleural cavity or pericardium. Pneumatization of bilateral lung parenchyma is mildly impaired against the background of respiratory artifacts. No significant focal or infiltrative changes are observed.

Based on the decision of the multidisciplinary team and the results of the instrumental studies (as reviewed by the pediatrician, surgeon, maxillofacial surgeon, oncologist, hematologist, and intensivist), it was planned to conduct a biopsy and morphological analysis of the obtained material.

The patient underwent a biopsy of the sternocleidomastoid formation under general balanced anesthesia. The material was sent for morphological research. The diagnosis was confirmed by morphological examination. Following the treatment administered at the hospital, the patient's condition stabilized and showed relative improvement. Stridor is now only slightly evident during feeding. The postoperative period proceeded without complications.

Tumor of the pectoralis major muscle is a rare condition in children. According to literature, it presents as a firm, solid, circumscribed mass within the thickness of the sternocleidomastoid muscle (SCM), typically occurring in infants 1-8 weeks of age and may be associated with torticollis. This pathology is more commonly observed as a unilateral neck formation; bilateral occurrence is very rare, which was the case with our patient. The diagnosis is initially suspected based on clinical data and subsequently confirmed by magnetic resonance imaging.

Discussion. Sternocleidomastoid tumor of infancy (STOI) is reported in the literature by authors M.B. Coventry, L.E. Harris et al. to occur in 0.4% of newborns [1]. In the literature, it is also frequently associated with torticollis and is often referred to as congenital muscular torticollis. Torticollis may occur independently or with an existing tumor, which requires differential diagnosis [2]. In the majority of cases, the tumor usually appears between 1-8 weeks of age [3,4]. This aligns with our patient's case, where the tumor was noticed by the parents at 10 days of age in the projection area of the sternocleidomastoid muscle on both sides of the neck, and was later misidentified as a lymph node by the doctor. According to the literature, most cases appear between 2 and 4 weeks of age. STOI typically presents as a firm, painless, isolated mass, 1 to 3 cm in diameter, within the thickness of the sternocleidomastoid muscle. It is usually located in the middle or lower third of the muscle. In our patient's case, bilateral masses on the anterolateral surface of the neck were noteworthy. The tumors were localized in the projection area of

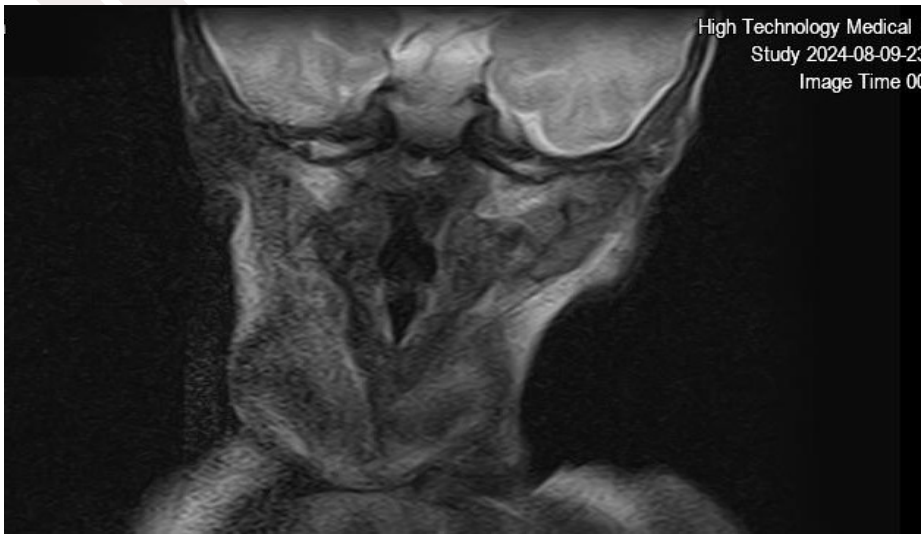
the sternocleidomastoid muscle on both sides, measuring 3 cm in size, dense, and immobile. They were not lymph nodes, and palpation was not painful. Torticollis with the head turned to the left was observed. The mass is typically located in the middle or inferior part of the SCM muscle [4]. There is no preference for side [5]. One previous case of bilateral involvement has been reported [6].

According to literature, the tumor usually increases in size over several weeks, stabilizes at 2-3 months of age, and then slowly decreases at 4-8 months of age. In our patient's case, rapid bilateral growth of the tumor was noted from 2-3 weeks of age, causing a worsening of the patient's condition with clinical signs of acute respiratory distress. Specifically, tachypnea, use of accessory muscles for breathing, difficulty swallowing, prolonged difficult inspiration, and inspiratory stridor were observed [7,8]. Torticollis may be associated with the disease but is generally transient and regresses with disease progression [8]. In our patient's case, torticollis was present with the head turned to the left.

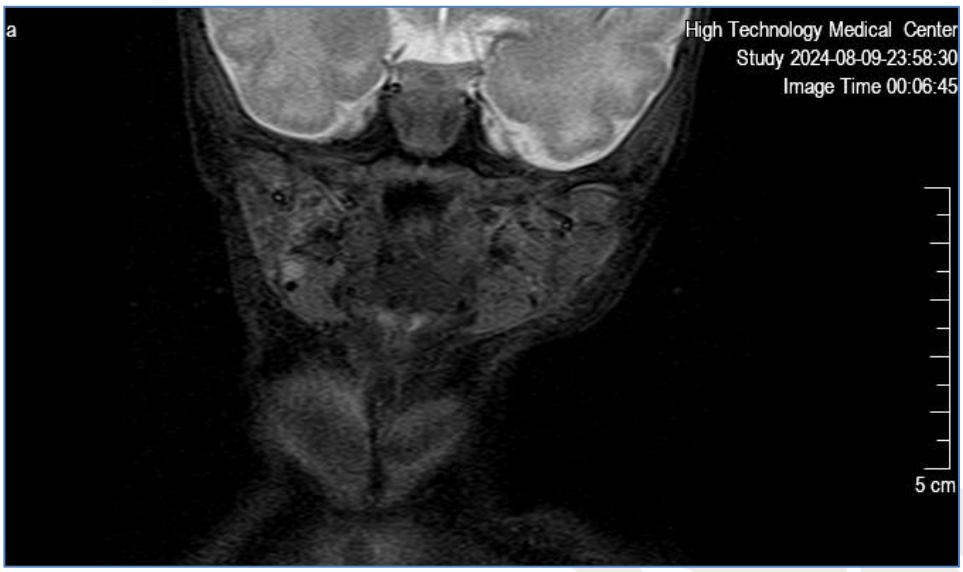
According to Coventry and Harris, based on an analysis of 35 cases, 86% of STOI cases regress by an average of 14 weeks of age with no residual effects [2]. In our clinical case, the mass was still present with the same dimensions at 14 weeks of age. Other authors [9] report that the tumor regresses (in 84 cases) by 7 months of age, with no residual changes observed in 77% of cases, while 23% of cases are associated with torticollis. Jones et al. suggest that early torticollis occurs in 9% of cases. Torticollis may also develop after the regression of STOI and is noted in 11% of cases [9, 10, 11,12]. In a large percentage of cases, the diagnosis is made based on a detailed medical history, clinical presentation, and physical examination. To confirm the diagnosis and differentiate the tumor from other conditions, ultrasonography and MRI are necessary. Tavill et al. [14] argue that MRI is essential to confirm the diagnosis. In our patient's case, it was confirmed by MRI:

The neck is turned to the left. In the right supraclavicular fossa, a somewhat hardened structure, not sharply demarcated, measuring up to 30 mm in size, with weak, uneven contrast enhancement is observed. On the contralateral side, at the same level, the soft tissues of the neck are easily deformed, revealing an unclearly defined area of similar structure up to 20 mm in size. The described process leads to lateral deviation and deformation of the thyroid gland, without apparent focal damage. There are no reliable MRI signs of the process spreading to the major blood vessels. Correlation with MRI data (STIR sequence) is highly consistent with bilateral sternocleidomastoid tumor of infancy (STOI). Morphological verification is recommended for unambiguous assessment.

Biopsy remains the last resort for cases with high suspicion of malignancy or those difficult to diagnose by cytology.



MRI showing bilateral STOIs with an isointense signal compared to muscle.



In most cases, the disease is treatable. Physiotherapy is indicated as the first line of treatment in most studies [6,7,8,9]. Treatment with physiotherapy is reported to be 76.8% successful. This is also confirmed by authors such as Canale and Morrison, among others. However, some researchers have recommended excision of the entire muscle [10]. According to other authors, the disease may regress even without treatment. However, after regression, there is a risk of developing torticollis and craniofacial asymmetry.

Conclusion. Bilateral STOI (Sternocleidomastoid Tumor of Infancy) is a very rare condition, with only a few cases reported in the literature. To our knowledge, this pathology has not been previously described in Georgia. The diagnosis may be suspected based on clinical presentation and medical history. The disease generally responds well to conservative treatment, with a high likelihood of full recovery. However, there remains a risk of developing torticollis and craniofacial asymmetry, which may require further treatment.

References:

1. Fibromatosis colli. Satish Kumar Gupta on 3 Aug 2023. Radiopaedia.org. doi: 10.53347/rLD-172957.
2. Sargar KM, Sheybani EF, Shenoy A, Aranake-Chrisinger J, Khanna G. Pediatric Fibroblastic and Myofibroblastic Tumors: A Pictorial Review. Radiographics. 2016 Jul-Aug;36(4):1195-214. doi: 10.1148/rg.2016150191. PMID: 27399243.
3. Diagnostic Imanging: Head and Neck 2017, Diagnostic Imaging: Head and Neck, Bernadette L. Koch, Bronwyn E. Hamilton,..H. Ric Harnsberger.
4. Sargar KM, Sheybani EF, Shenoy A, Aranake-Chrisinger J, Khanna G. Pediatric Fibroblastic and Myofibroblastic Tumors: A Pictorial Review. Radiographics. 2016 Jul-Aug;36(4):1195-214. doi: 10.1148/rg.2016150191. PMID: 27399243.
5. Sara Haseli, Bahar Mansoori et al. Fibroblastic and Myofibroblastic Soft-Tissue Tumors: Imaging Spectrum and Radiologic-Pathologic Correlation. Radiographics. 2023 Aug; 43(8):e230005. Doi: 10.1148/rg.230005.
6. Haseli S, Mansoori B, Christensen D, Abadi A, Pooyan A, Shomal Zadeh F, Mau B, Khalili N, Murphey M, Chalian M.Radiographics. 2023 Aug;43(8):e230005. doi: 10.1148/rg.230005.PMID: 37440448
7. Roula Hourani, Bedros Taslakian et al. Fibroblastic and myofibroblastic tumors of the head and neck: comprehensive imaging-based review with pathologic correlation. Eur J Radiol. 2015 Feb; 84(2):250-60.

8. Hourani R, Taslakian B, Shabb NS, Nassar L, Hourani MH, Moukarbel R, Sabri A, Rizk T.Eur J Radiol. 2015 Feb;84(2):250-60. doi: 10.1016/j.ejrad.2014.10.017. Epub 2014 Nov 1.PMID: 25467227 Review.
9. Harshita Agarwal, Lavleen Singh et al. Non-malignant fibroblastic/myofibroblastic tumors in pediatric age group: Clues and pitfalls to the cytological diagnosis. Cytopathology, 2020 Mar; 31(2):115-121. Doi: 10.1111/cyt.12786.
10. Agarwal H, Singh L, Gupta N, Sengar M.Cytopathology. 2020 Mar;31(2):115-121. doi: 10.1111/cyt.12786. Epub 2020 Feb 7.PMID: 32034815
11. Oscar M Navarro. Pearls and Pitfalls in the Imaging of Soft-Tissue Masses in Children. Semin Ultrasound CT MR. 2020 Oct; 41(5):498-512.
12. Navarro OM.Semin Ultrasound CT MR. 2020 Oct;41(5):498-512. doi: 10.1053/j.sult.2020.05.014. Epub 2020 May 28.PMID: 32980096 Review.
13. A clinicopathologic study of 45 pediatric soft tissue tumors with an admixture of adipose tissue and fibroblastic elements, and a proposal for classification as lipofibromatosis. Fetsch JF, Miettinen M, Laskin WB, Michal M, Enzinger FM.Am J Surg Pathol. 2000 Nov;24(11):1491-500. doi: 10.1097/00000478-200011000-00004.PMID: 11075850
14. M.A. Tavill, R.F. Wetmore, A case of familial sternocleidomastoid tumor of infancy. Int J Pediatr Otorhinolaryngol. 1996 Dec 20; 38(2):163-8.

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SUMMARY

Tumor of the pectoralis major muscle is a rare condition in children. According to literature, it presents as a firm, solid, circumscribed mass within the thickness of the sternocleidomastoid muscle (SCM), typically occurring in infants 1-8 weeks of age and may be associated with torticollis. This pathology is more commonly observed as a unilateral neck formation; bilateral occurrence is very rare, which was the case with our patient. The diagnosis is initially suspected based on clinical data and subsequently confirmed by magnetic resonance imaging.

We report a rare case of bilateral pectoralis major muscle tumors in a 3-week-old infant. To our knowledge, this pathology has not been previously described in Georgia. The diagnosis may be suspected based on clinical presentation and medical history. The disease generally responds well to conservative treatment, with a high likelihood of full recovery.

Keywords: Pulmonary, Sternocleidomastoid muscle, Newborn, MRI, breathing