Bilateral Fibromatosis Colli: A Case Report and Review of Literature

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MT, KA, and CN contributed to the literature search and writing. PA provided clinical input. All authors revised the article, approved the manuscript, and agreed with its submission.

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ABSTRACT

Fibromatosis colli is a rare benign fibrous tumour of the sternocleidomastoid muscle, affecting 0.4% of infants typically within the first 2-3 weeks of life. Fibromatosis colli is usually unilateral, appears more often on the right side, and affects male patients slightly more than females. Here we present a rare case of bilateral fibromatosis colli in a 5-week-old baby. To the best of our knowledge, there were only 9 cases described in the English literature before our report and review. Bilateral fibromatosis colli has a heterogeneous presentation, and its initial diagnosis can be clinically challenging. As such, we recommend supplementing the history and physical examination with ultrasound to improve diagnostic accuracy. With prompt diagnosis, bilateral fibromatosis colli can be initially managed with physiotherapy, minimising the need for surgical intervention.

CASE REPORT

BACKGROUND

Fibromatosis colli (FC) is a rare, benign fibrous tumour of the sternocleidomastoid muscle (SCM) with an incidence of 0.4%, typically within the first 2-3 weeks of life [1]. On physical examination, patients classically present with localised swelling of the SCM, restricted neck motion, and congenital torticollis with their heads facing the ipsilateral mass. It is usually unilateral, appears more often on the right side and closer to its inferior aspect, and affects boys slightly more than girls. While the exact aetiology is still unknown, it is believed to be secondary to a tilted and malpositioned foetal head during the intrauterine period. Diagnosis can be made clinically, supported with multimodalities such as ultrasound (US) or magnetic resonance imaging (MRI).

The initial diagnosis of bilateral FC can be a clinical challenge [1] and prompt diagnosis is important to prevent further complications [2]. However, as bilateral FC is extremely rare, the evidence from the literature is limited. Before our case report and review, there were only 9 cases described in the English literature [3]. Therefore, we review the literature and present a case report of a patient with bilateral FC. Diagnosis can be made clinically and confirmed with an US evaluation of the neck. If diagnosed early, FC can be managed conservatively with physiotherapy; otherwise, invasive or surgical intervention, such as distal tenotomy, muscle lengthening, and muscle excision, may be required for refractory cases or when the child is over age 1 [1].

CASE REPORT

A 39-day-old baby boy was admitted to the Montreal Children's Hospital with a preliminary diagnosis of sepsis of unknown aetiology. On physical examination, there were bilateral neck masses initially consistent with bilateral cervical lymphadenopathy.

Imaging findings

An US study of the neck was performed. The US examination showed slight heterogeneity, increased overall echogenicity, and fusiform dilation of both the right and left SCM, with a maximal thickening of 0.8 cm on the right and 0.7 cm on the left (Figure 1). Contrary to the initial impression, there were no associated calcifications, cystic changes, or size-significant lymphadenopathy. Colour Doppler evaluation showed no hypervascularity of the left or right SCMs or hyperaemia of the surrounding soft tissues. These US findings were suggestive of bilateral FC. Physiotherapy was recommended for the patient, and a follow-up US study of the neck was booked.

Follow-up

The follow-up US was performed 7 months after the patient's initial presentation and demonstrated significant improvement (Figure 2), with both right and left SCM showing normal echotexture and significant interval reduction of their previously noted thickening.

DISCUSSION

Aetiology and Demographics

FC, also known as "sternocleidomastoid tumour of infancy", is a benign, fibrous SCM tumour that typically presents in the second or third week of life [4], more commonly in boys than girls [5]. Infants typically present with a firm, mobile anterior neck mass [6]. The tumour may grow for several weeks, stabilise for a few months, and spontaneously decrease in size by 4 -8 months of age [7]. Early detection and treatment resolve the disease in about 80% of patients, though it may also cause complications such as torticollis, facial deformities, and lytic clavicular lesions [5]. Unilateral FC has a rare incidence of 0.4%, occurring on the right SCM more often than the left [8]. However, bilateral FC is exceptionally rare, and to the best of our knowledge, only 9 cases were reported in the English literature before this report [3].

Clinical and Imaging Findings

The clinical presentation of bilateral FC varied greatly when reviewing previous case reports (Table 1). Only one case reported the classical finding of torticollis towards one side of the neck [9], while other clinical presentations included non-tender masses within the neck [10], restricted neck movements [11], or a short-appearing neck with upward tilting of the face [1]. As these cases are limited in quantity and variable in presentation, our report summarises the literature, adds to the evidence, and makes management recommendations.

Differential Diagnosis

On history and physical exam alone, the differential diagnosis of a neck mass in a young infant includes cervical lymphadenopathy/lymphadenitis, cervical teratoma, branchial cleft remnants, congenital thyroid goitre, lymphatic or vascular malformations, or neoplastic tumours such as sarcoma [7]. Bilateral FC presents as a firm, non-tender nodule, usually in the distal two-thirds of the SCM [20]. It results in apparent shortening of the neck and upturning of the chin [20]. Radiological imaging improves the accuracy of the differential diagnosis. US shows unilateral, fusiform expansion and increased echogenicity of the sternocleidomastoid muscle [4,6]. The intramuscular mass and its margins are well-defined, with no invasion of adjacent soft tissues (Figure 1). There is no lymphadenopathy or surrounding inflammatory changes. Therefore, based on the size and echogenicity of the SCM on US, lymphadenopathy, branchial cleft remnants, thyroid pathology, lymphatic malformations, vascular malformations, and neoplastic lesions such as teratoma or sarcoma are ruled out.

US is the imaging modality of choice due to its wide availability, low cost, lack of ionising radiation, and a 100% sensitivity rate [21]. US is usually diagnostic of FC and is often the only imaging study performed. In the case of inconclusive results, MRI may be used. MRI findings suggestive of fibromatosis colli include a fusiform or eccentric expansion of the sternocleidomastoid muscle[6]. The mass is isointense to muscle on T1-weighted imaging, and hyperintense on T2-weighted imaging, with subtle patchy and linear areas of decreased signal intensity [6]. If suspicious of malignancy (*e.g.,* multiple, firm, non-tender, non-mobile nodes), biopsies may be used. Pathological evaluation typically demonstrates the presence of myoblasts, fibroblasts, and myofibroblasts [6]. Fine needle aspiration shows benign spindle cells, which helps differentiate from other entities [8].

Treatment & Prognosis

The clinical course of FC is favourable and many cases will resolve spontaneously within 4 to 8 months [20]. If discovered within the first month of life, 90% of patients will have resolution after a year of physiotherapy [8]. Physiotherapy consists of massages, stretching exercises, strengthening exercises, and heat [8].

It is important to recognise and treat FC correctly. If left untreated, persistent torticollis may lead to the development of cranial or facial asymmetry. If symptoms persist beyond a year or craniofacial abnormalities develop, then surgical intervention may be needed.

CONCLUSION

FC is a rare, benign condition caused by a benign enlargement of the SCM, representing a cause of congenital torticollis. Bilateral FC is even rarer, with only 9 cases reported in the English literature before our case report and review, with a significant variability in the clinical presentation and diagnostic approach of these cases. Bilateral FC may be diagnosed based on history and physical exam, but US improves diagnostic accuracy. US is sufficiently sensitive and specific to make the diagnosis [18]. Fine-needle aspiration cytology can also be used to diagnose FC when there is suspicion of malignancy; however, it should be reserved for when both the US and MRI

do not show typical findings. These additional approaches should be considered individually based on clinical features on presentation.

Once the diagnosis of unilateral or bilateral FC has been made, physiotherapy should be the treatment of choice and surgical interventions are rarely required. In the few cases reported in the literature of bilateral FC resolution followed physiotherapy, as is expected with unilateral cases. After treatment, patients are followed to document resolution and ensure no complications develop.

In summary, it is important to recognise this rare presentation of bilateral FC to diagnose promptly, improve prognosis, and limit complications.

TEACHING POINT

Bilateral FC is a rare, benign fibroblastic tumour of the SCM that occurs in neonates. While bilateral FC cases lack a unifying clinical presentation, they can be readily diagnosed and followed using US, which demonstrates fusiform dilation of the distal two-thirds of the SCM and no surrounding tissue invasion.

QUESTIONS

Question 1: Which of the following statements regarding the clinical presentation of bilateral fibromatosis colli (FC) is correct?

1. All patients present with torticollis.
2. It always presents with bilateral cervical lymphadenopathy.
3. The masses are typically painful and inflamed.
4. The clinical presentation can vary and may include neck masses or restricted movement. (applies)
5. It is only diagnosed through biopsy.

Explanation:

1. Only one reported case had classical torticollis; others had variable presentations.
2. While lymphadenopathy was suspected clinically, ultrasound (US) ruled it out.
3. FC masses are described as firm and non-tender.
4. Presentations are heterogeneous, including masses, restricted motion, or postural abnormalities.
5. Diagnosis is typically made via US; biopsy is reserved for unclear or suspicious cases.

Question 2: Which imaging features are typically seen on US in FC?

1. Lymphadenopathy surrounding the sternocleidomastoid muscle (SCM).
2. Fusiform dilation and increased echogenicity of the SCM. (applies)
3. Cystic degeneration within the SCM.
4. Calcifications and heterogeneous enhancement.
5. Hypervascularity on colour Doppler.

Explanation:

1. Lymphadenopathy was specifically ruled out on US.
2. These are characteristic findings in FC.
3. FC is a solid fibrous tumour; cystic change is not typical
4. No calcifications or abnormal enhancement were seen.
5. Doppler shows no increased vascularity in typical FC cases.

Question 3: Which of the following statements regarding the treatment and prognosis of FC is accurate?

1. Surgery is the first-line treatment.
2. Antibiotics are required to resolve the condition.
3. Most cases resolve spontaneously or with physiotherapy. (applies)
4. It usually results in permanent craniofacial deformity.
5. Radiation therapy is an effective option.

Explanation:

1. Surgery is only used if physiotherapy fails or complications develop.
2. FC is not infectious; one misdiagnosed case was treated unsuccessfully with antibiotics.
3. Physiotherapy or spontaneous resolution is the mainstay of treatment.
4. If treated early, permanent deformities are uncommon.
5. Radiation is not part of the management of FC.

Question 4: Which differential diagnoses should be considered for a neonatal neck mass?

1. Congenital thyroid goitre. (applies)
2. Hepatic haemangioma.
3. Cervical lymphadenopathy. (applies)
4. Osteosarcoma.
5. Branchial cleft remnants. (applies)

Explanation:

1. Differentials include cervical lymphadenopathy/ lymphadenitis, cervical teratoma, branchial cleft remnants, and congenital thyroid goitre.
2. This is not a neck lesion and is not listed as a differential.
3. This was the initial clinical impression in the presented case.
4. This is not relevant in neonatal neck masses.
5. This is part of the listed differentials as mentioned above.

Question 5: Which of the following statements about imaging in FC is correct?

1. Magnetic resonance imaging (MRI) is the primary imaging modality due to its high sensitivity.
2. Biopsy is routinely needed to confirm the diagnosis.
3. US has a 100% sensitivity rate for detecting FC. (applies)
4. Computerised tomography (CT) is the imaging modality of choice.
5. 5. US typically shows aggressive features invading soft tissue.

Explanation:

1. MRI is used only when US is inconclusive.
2. Biopsy is reserved for atypical cases or suspicion of malignancy.
3. This is stated in the article.

4. CT is not routinely used, as US is preferred.
5. US shows well-defined intramuscular masses with no adjacent invasion.

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FIGURES

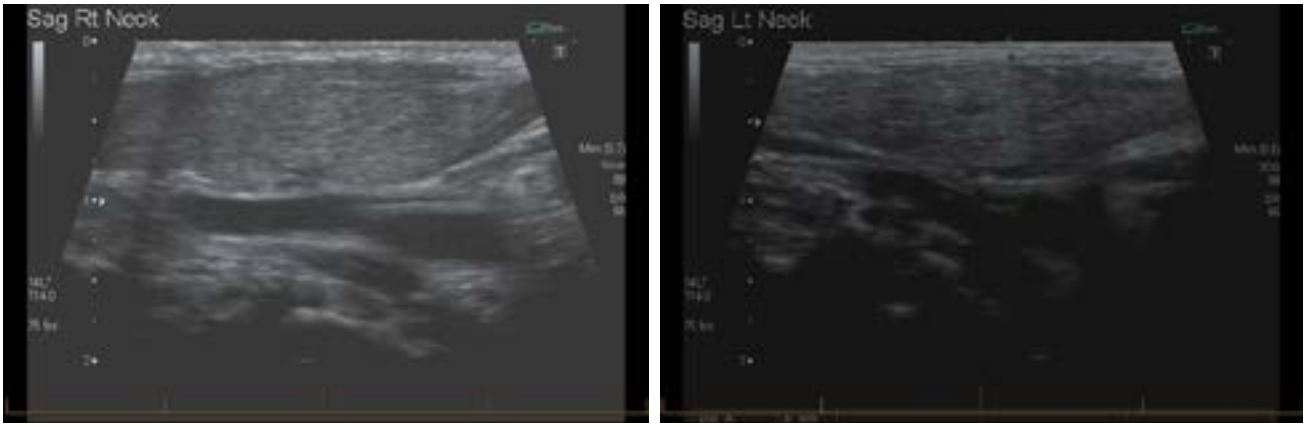

Figure 1: Initial ultrasound of a 39-day-old baby boy with bilateral fibromatosis colli. FINDINGS: Sagittal views show mildly increased overall echogenicity and fusiform dilatation of both the right and left sternocleidomastoid muscles, measuring 0.8 cm and 0.7 cm, respectively.

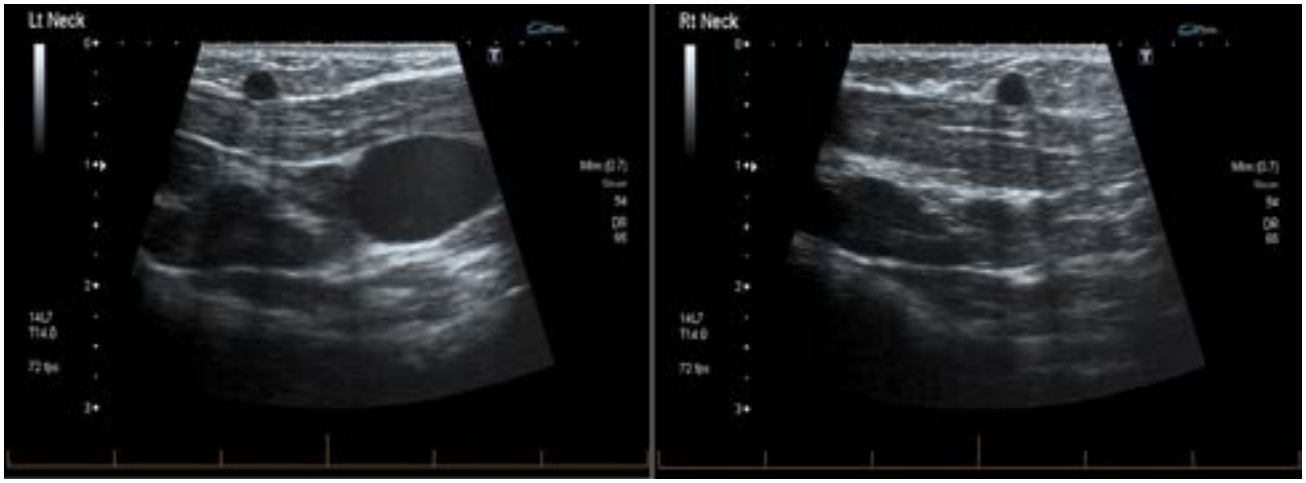

Figure 2: Follow-up ultrasound of a 39-day-old baby boy with bilateral fibromatosis colli. FINDINGS: Sagittal views of the left and right sternocleidomastoid muscles demonstrate normal overall echogenicity and interval normalisation their thickness bilaterally.

Table 1. Previously reported cases of bilateral fibromatosis colli in the English literature.

	Patient						
Author	#	Sex	Age	Clinical history	Diagnostic modality	Treatment	Outcome
Thomsen[10]	1	M	5 wks	Uneventful birth. Bilateral 3-4 cm non-tender, hard, lobulated masses at distal end of both sternocleidomastoid muscles noted shortly after birth.	Unremarkable cervical radiograph. CT showing discrete solid masses. Ultrasound showing bilateral solid masses. Open biopsy showing diffuse fibrosis compatible with fibromatosis colli.	Physiotherapy	Resolved
Wright[12]	2	NR*	NR*	One reported case of bilateral fibromatosis colli within the series of 100 neonates and children Clinical presentation unclear for specific case	NR*	Physiotherapy†	Resolved†
Chan[13]	3	NR*	8 mths	One reported case of bilateral fibromatosis colli within the series of 36 children Clinical presentation was unclear for the specific case	Ultrasound showing mass involving mid-distal third of sternocleidomastoid muscle, of patchy or homogenous echotexture‡	NR*	NR*
Manchanda[11]	4	M	8 wks	Uneventful birth. Bilateral neck masses for 2 weeks. Physical exam showing firm, non-tender mass bilaterally. Restricted horizontal mobility.	Clinical diagnosis	Physiotherapy	NR*
Kumar[1]	5	M	7 wks	Uneventful delivery. Diagnosed as lymphadenitis and treated unsuccessfully with antibiotics. Physical examination with firm bilateral masses, located in the upper third of the neck. Restricted horizontal movement.	FNAC showing spindle-shaped fibroblasts separated by collagen and degenerating skeletal muscle.	Physiotherapy	Resolved
	6	M	4 wks	1 week history of bilateral neck masses, 1.5x1.5 cm hard, non-tender within the middle third of the sternocleidomastoids, horizontal mobility. Neck appeared short with chin elevated and face tilted upwards.	Ultrasound demonstrating bilateral ovoid homogenous masses within the body of the sternocleidomastoid.	Physiotherapy	Improved
Tufano[9]	7	F	2 wks	Bilateral neck masses, firm, well-circumscribed within the SCM. Torticollis and face turned to the right shoulder.	MRI demonstrating prominent SCMs, increased enhancement post-contrast, consistent with STOI.	Physiotherapy	Resolved

Durnford[3]	8	M	5 wks	Delivery complicated by intraoperative trauma. Neonate presents with bilateral, palpable anterior neck masses of 4x3 cm (right) and 3x2 cm (left). Neck was slightly flexed.	Ultrasound illustrating well-defined soft tissue lesions within both SCMs	Physiotherapy	Resolved
Dangi[14]	9	M	3 wks	11-day history of firm to hard, well-circumscribed neck masses within lower 1/3 of SCM	Ultrasound illustrating hypoechoic lesions of 1.7*0.8 cm (right) and 1.9*0.9 cm (left)	Physiotherapy	Improved
Sabounji[15]	10	NR*	NR*	One reported case of bilateral fibromatosis colli within the series of 26 neonates and children. Mobile mass with SCM, not inflammatory, no change in size or tension with manoeuvres.	Cervical ultrasound showing a subcutaneous formation	NR*	NR*
Humar[16]	11	F	5 wks	1-week history of bilateral neck masses rapidly increasing in size.	Ultrasound and CT scan inconclusive, excisional biopsy performed on the right side	NR*	Resolved
Sönmez[17]	12	NR*	NR*	One reported case of bilateral fibromatosis colli within the series of 52 children Clinical presentation unclear for specific case	NR*	Surgery	Resolved
Matuszewski[18]	13	M	12-year-old	Asymmetry of head and face, smaller right cheek, rotated chin. Limited range of motion. Deformation of cervical and thoracic spine, increased thoracic kyphosis, elevation of right shoulder	Radiographs	Surgery	Resolved
Öhman[19]	14	F	2.5 mths	Prenatally in breech position, turned by healthcare provider some weeks before delivery in cephalic presentation. Referred for moderate brachycephaly and found to have bilateral thickened SCM with active and passive rotations affected.	Clinical diagnosis	Physiotherapy	Resolved (at about 5 months old)

[21]* Unreported data
† All sternomastoid tumors resolved, however six patients returned with contracture. Patients who returned with contracture were treated with surgical division and physiotherapy. It is not specified which patients had complications.
‡ Imaging findings of individual cases not reported.
Mths = months. Wks = weeks.

KEYWORDS

Fibromatosis colli; sternocleidomastoid tumour of infancy; pseudotumor of infancy; sternomastoid tumour of infancy; infancy sternocleidomastoid pseudotumor; congenital pseudotumor

ABBREVIATIONS

US = Ultrasound
FC = Fibromatosis Colli
SCM = Sternocleidomastoid muscle
Mnths = Months
Wks = Weeks