CASE REPORT

Acquired torticollis due to primary pyomyositis of the paraspinal muscles in an 11-year-old boy

S Ray,^{1,2} A Iyer,³ S Avula,⁴ R Kneen⁵

¹Department of Neurosciences, Alder Hey Children's Hospital, Liverpool, UK

²University of Liverpool, Institute of Infection and Global Health, Liverpool, UK

³Alder Hey Children's NHS Foundation Trust, Liverpool, UK

⁴Department of Radiology, Alder Hey Children's Hospital, Liverpool, UK

⁵Littlewoods Neurosciences Unit, Alder Hey Children's Hospital, Liverpool, UK

Correspondence to

Dr R Kneen,
rachel.kneen@liverpool.ac.uk

Accepted 24 February 2016

SUMMARY

Torticollis is characterised by tilting and rotation of the cervical spine in opposite directions. Causes can be congenital or acquired. Primary pyomyositis is a rare subacute deep bacterial infection of skeletal muscles that typically affects individuals under 20 years of age from tropical countries. Infrequently, pyomyositis occurs in individuals from temperate regions, usually in immunocompromised adults, and this is defined as secondary pyomyositis. We report a case of acquired torticollis due to primary pyomyositis of the paraspinal muscles in a previously healthy boy from the UK. A prolonged course of antibiotics and physiotherapy led to a complete resolution of his illness. We review how to differentiate pyomyositis from focal myositis, a more common inflammatory muscular cause of acquired torticollis.

BACKGROUND

Torticollis is characterised by tilting and rotation of the cervical spine in opposite directions. Causes can be congenital or acquired.^{1 2} It is common and occurs in all age groups.³ Congenital torticollis occurs in 1–2% of children, usually resulting from craniocervical vertebral abnormalities or muscular causes. Acquired torticollis is often secondary to trauma,^{1 2} but there are many other causes (table 1).

Primary pyomyositis (or tropical pyomyositis) is a rare subacute bacterial infection of skeletal muscles and is not associated with any contiguous infection of bone, soft tissue or skin. The majority of cases occur in healthy individuals under 20 years of age from tropical countries.⁵

Reported cases of pyomyositis in temperate regions are rare, being generally described in immunocompromised older individuals with conditions such as HIV, malignancy or malnutrition; this

is categorised as secondary pyomyositis (or non-tropical pyomyositis).^{6–8}

In contrast, focal myositis is a localised self-limiting focal inflammation (non-infective) of skeletal muscles and has more frequently been reported to present with torticollis;⁹ steroids are used in the treatment of this condition.

We report the first detailed case of acquired torticollis due to primary pyomyositis in a boy, where a prolonged course of intravenous antibiotics and physiotherapy led to full recovery. We also highlight how to clinically differentiate between focal myositis and pyomyositis—key to ensuring appropriate treatment is offered for these distinct causes of acquired torticollis.

CASE PRESENTATION

A previously well 11-year-old boy presented to a district general hospital with a 3-week history of severe left-sided neck pain and stiffness. He had been having high fevers and rigours for a week. There was no history of trauma. There was no family history of neurological conditions and he was on no regular medications. On examination, he had a left-sided torticollis, with significantly reduced neck movements, particularly of lateral movement of his head to the right. He had a fever and was tachycardic. There were no focal neurological signs, no carotid bruit and the rest of the physical examination was normal. He was referred to the tertiary neurology department with an acquired torticollis of unknown aetiology, where he underwent further investigation.

INVESTIGATIONS

Blood results: C reactive protein (CRP) 203 mg/L, white cell count (WCC) $12.6 \times 10^9/L$, neutrophils $9.0 \times 10^9/L$, clotting within normal range, IgG, IgM and IgA levels all within normal range.

Table 1 Causes of childhood torticollis⁴

Congenital causes	Acquired causes
Muscular torticollis—thought to be due to restricted movement of the fetus in utero leading to fibrosis of muscle	Trauma, eg, fractured occipital condyle fracture
Osseous torticollis, eg, Klippel-Feil syndrome	Infection, eg, osteomyelitis
Central nervous system torticollis, eg, syringomyelia	Inflammation, eg, juvenile idiopathic arthritis
Ligamentous laxity, eg, Down syndrome	Malignant, eg, posterior fossa tumour
	Ocular torticollis, eg, superior oblique muscle palsy
	Drug induced torticollis, eg, olanzapine
	Gastro-oesophageal reflux (Sandifer's syndrome)
	Benign paroxysmal torticollis—considered to be a migraine variant
	Vascular abnormality, eg, arteriovenous fistula

CrossMark

To cite: Ray S, Iyer A, Avula S, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2015-213409

Urine microscopy and culture: No growth.

Throat culture: No growth.

Blood culture—First culture day one—*Streptococcus constellatus*—fully sensitive to penicillin, cefotaxime, erythromycin, clindamycin, vancomycin and teicoplanin. Second culture day three—No growth.

C-spine X-ray: No fracture, no atlantoaxial subluxation. Prevertebral soft tissue outline grossly normal.

Ears, nose and throat (ENT) review day 2: ENT examination unremarkable, low clinical suspicion that torticollis was due to Grisel's syndrome, alternative diagnosis more likely (CT felt not to be indicated); pyomyositis subsequently confirmed by MRI (figure 1A).

Ophthalmology review day 2: No evidence of squint as a contributing factor for the torticollis.

MRI of the head and neck day 2: Hyperintensity within the left paraspinal muscles at the level of C1–C3 demonstrating underlying infective/inflammatory change; interpreting this in correlation with the clinical picture, this suggests pyomyositis (figure 1A). Venous sinus thrombosis identified in the left transverse sinus, sigmoid sinus and the proximal segment of the left

internal jugular vein. Incidental finding of a mild Chiari malformation with a small syrinx at the 6th cervical level.

Neurosurgical review day 2: Incidental small Chiari malformation on MRI, no intervention required.

Infectious diseases review day 2: Prolonged course of antimicrobial therapy advised for pyomyositis (see below).

MRI of the head and neck at 6 month follow-up: Hyperintensity at paravertebral muscles C1–C3 fully resolved. Absence of flow in left transverse sinus, sigmoid sinus and internal jugular venous thrombosis remains.

MRI of the head and neck at 1 year follow-up: Previous signal abnormality in left transverse sinus, sigmoid sinus and internal jugular less prominent. Signal abnormality in paraspinal muscles C1–C3 resolved (figure 1B).

DIFFERENTIAL DIAGNOSIS

- ▶ Primary pyomyositis
- ▶ Focal myositis
- ▶ Grisel's syndrome: non-traumatic subluxation of the atlantoaxial joint due to infection or inflammation in the ENT tissues
- ▶ Mastoiditis
- ▶ Lemierre's syndrome: thrombophlebitis of the internal jugular vein as a complication of bacterial sore throat

TREATMENT

The infectious diseases team advised intravenous ceftriaxone 2 g once daily and 150 mg clindamycin four times daily for 2 weeks followed by 4 weeks of oral coamoxiclav 250/125 mg three times a day. A long line was inserted to facilitate this treatment. Additionally, physiotherapy sessions were performed three times a day for 3 weeks. Each session contained five to six stretches and lasted 20 min. Initially, stretches were mainly passive, as they were limited by pain and the severity of the contracture, but they became increasingly active over time as the infection improved. The patient was transferred back to the referring hospital to complete his antibiotic and physiotherapy treatment.

OUTCOME AND FOLLOW-UP

Within a week, the patient's inflammatory markers returned to normal. Three weeks into treatment he had complete resolution of the torticollis and regained full range of neck movements. His follow-up MRI scans showed complete resolution of the pyomyositis and resolving venous thrombosis. He was discharged from follow-up at 1 year.

DISCUSSION

Torticollis, commonly known as 'wryneck', is characterised by rotational deformity of the cervical spine with secondary tilting of the head and rotation of the chin in the opposite direction. Many diseases can present as torticollis (table 1).³ A previous case series briefly listed torticollis as a symptom in a child with pyomyositis of the levator scapulae,⁶ but we believe ours is the first detailed report of pyomyositis primarily presenting as torticollis. Pyomyositis often affects the largest muscle groups, most commonly around the pelvic girdle.¹⁰ The clinical picture often begins with a painful muscle that is tense on examination but initially presents minimal erythema. This progresses to a suppurative phase involving worsening pain and fever, evolving erythema and eventual formation of a muscle abscess. Left untreated, this leads to multifocal abscess formation followed by fulminant septicaemia and death.^{5 6 11}

The skeletal muscle is intrinsically resistant to bacterial infection in normal physiology and the pathogenesis of pyomyositis

Figure 1 An 11-year-old boy presenting with acquired torticollis. Coronal short tau inversion recovery image (A) from the MRI scan of the neck demonstrating hyperintensity within the left paraspinal musculature and surrounding soft tissue (arrows) suggestive of underlying infective/inflammatory change. Follow-up imaging 12 months later (B) showing complete resolution of the abnormality.

remains incompletely understood.^{11 12} It is thought to be a complication of bacteraemia because it develops without penetrating injury. Trauma is a risk factor because it alters the local muscle tissue structure that then acts as a sanctuary for bacteria.¹¹ *Staphylococcus aureus* is the causative pathogen in 70–95% of cases,¹³ while group A *Streptococcus* is the second commonest organism.¹⁴ Other pathogens have very rarely been reported.⁶ *S. constellatus*, the causative organism in our case, plus *S. anginosus* and *S. intermedius*, are three distinct species that comprise the *S. milleri* group. They are considered part of the normal oropharyngeal, urogenital and gastrointestinal flora. However, they are capable of pyogenic infection and abscess formation. There are only three previous reported cases of *S. milleri* causing pyomyositis,^{15–17} and all had risk factors (HIV or malnutrition). It is unclear why pyomyositis occurred in the reported child, who was immunocompetent and had no history of trauma. We also presume that the venous thrombosis he suffered was secondary to the infection as there was no underlying coagulopathy.

Owing to the rarity of pyomyositis it is unlikely to be considered as the diagnosis during the initial presentation of acquired torticollis. Focal myositis, a localised self-limiting focal inflammation of skeletal muscles, has more frequently been reported to present with torticollis.⁹ The mainstay of treatment for myositis is steroids, while for pyomyositis a prolonged course of intravenous antibiotics and sometimes surgical drainage can be required. Identifying the differentiating features between these two pathologies will help clinicians to make an earlier diagnosis of pyomyositis, prior to the formation of abscesses.^{10 11}

Features of infection such as fever, elevated inflammatory markers (WCC, CRP, erythrocyte sedimentation rate (ESR)) and bacteraemia, are often present in pyomyositis but absent in myositis.^{6 9 13} MRI is considered the gold standard imaging modality to diagnose pyomyositis and will identify diffuse muscle inflammation, reactive lymphadenopathy and abscesses, if they have formed.¹⁸ MRI findings in focal myositis highlight focal inflammation only.¹¹ In cases where pyomyositis is suspected, contrast-enhanced MRI scan can best identify underlying abscess formation helping support the diagnosis. In rare cases where the diagnosis remains unclear, fine-needle biopsy for histology will confirm the diagnosis.^{19 20}

Early diagnosis and appropriate antibiotic therapy (with full sensitivity to the causative organism) is key to successful treatment of pyomyositis. In refractory cases, drainage of purulent

material may be needed. This was not required in our case. With appropriate treatment, outcome is very good with complete resolution of symptoms in the vast majority of cases.¹¹

In summary, we report the first detailed case of acquired torticollis due to primary pyomyositis. Early MRI and identification of the organism facilitated the diagnosis. A prolonged course of antibiotics and physiotherapy led to complete resolution of the patient's torticollis.

Acknowledgements The authors acknowledge the following: Dr Kate Hunter for her clinical management of the case at the local district general hospital; the ENT, ophthalmology and neurosurgical teams for their support in the initial investigations, and the infectious diseases team for their specialist guidance on appropriate treatment.

Contributors SR was responsible for conception and design, acquisition of the data, drafting the article, final approval of final version to be published. AI was responsible for conception and design, revision of the draft for critically important intellectual content, final approval of version to be published. SA was responsible for contribution and analysis of radiological images, revision of the draft for critically important intellectual content, final approval of version to be published. RK was responsible for conception and design, acquisition of the data, revision of draft for critically important intellectual content, final approval of version to be published.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- Herman MJ, Wolf M. Torticollis in children. *Curr Orthop Pract* 2013;24:598–603.
- Haque S, Bilal Shafi BB, Kaleem M. Imaging of torticollis in children. *Radiographics* 2012;32:557–71.
- Tomczak KK, Rosman NP. Torticollis. *J Child Neurol* 2013;28:365–78.
- Herman MJ. Torticollis in infants and children: common and unusual causes. *Instr Course Lect* 2006;55:647–53.
- Hadjipavlou M, Butt DA, McAllister J. Primary pyomyositis: an unusual presentation in an older patient with no recognised risk factors. *BMJ Case Rep* 2012;2012:pii: bcr1220115342.
- Ovadia D, Ezra E, Ben-Sira L, et al. Primary pyomyositis in children: a retrospective analysis of 11 cases. *J Pediatr Orthop B* 2007;16:153–9.
- Chiedozi LC. Pyomyositis. Review of 205 cases in 112 patients. *Am J Surg* 1979;137:255–9.
- Geelhoed GW, Gray H, Alavi IA, et al. Pyomyositis—tropical and nontropical. *N Engl J Med* 1971;284:853–4.
- Manickavasagam J, Majumdar S, Bhattacharya AK. Focal inflammatory myositis of the paraspinal neck muscles. *Ear Nose Throat J* 2008;87:430–2.
- Giebaly DE, Horriat S, Sinha A, et al. Pyomyositis of the piriformis muscle presenting with sciatica in a teenage rugby player. *BMJ Case Rep* 2012;2012:pii: bcr1220115392.
- Bickels J, Ben-Sira L, Kessler A, et al. Primary pyomyositis. *J Bone Joint Surg Am* 2002;84-A:2277–86.
- Chauhan S, Jain S, Varma S, et al. Tropical pyomyositis (myositis tropicans): current perspective. *Postgrad Med J* 2004;80:267–70.
- Hassan FO, Shannak A. Primary pyomyositis of the paraspinal muscles: a case report and literature review. *Eur Spine J* 2008;17(Suppl 2):S239–42.
- Pogodina VV. Elizaveta Nilolaevna Levkovich-75th birthday. *Acta Virol* 1975;19:509.
- Walkty A, Embil JM, Nichol K, et al. An unusual case of *Streptococcus anginosus* group pyomyositis diagnosed using direct 16S ribosomal DNA sequencing. *Can J Infect Dis Med Microbiol* 2014;25:32–4.
- Yassin M, Yadavalli GK, Alvarado N, et al. *Streptococcus anginosus* (*Streptococcus milleri* group) pyomyositis in a 50-year-old man with acquired immunodeficiency syndrome: case report and review of literature. *Infection* 2010;38:65–8.
- Palomino-Nicás J, González E, Arroyo A, et al. Pyomyositis due to *Eubacterium lentum* and *Streptococcus constellatus* from a periodontal source. *Clin Infect Dis* 1996;22:176–8.
- Soler R, Rodríguez E, Aguilera C, et al. Magnetic resonance imaging of pyomyositis in 43 cases. *Eur J Radiol* 2000;35:59–64.
- Heffner RR Jr, Barron SA. Polymyositis beginning as a focal process. *Arch Neurol* 1981;38:439–42.
- Heffner RR Jr, Barron SA, Jenis EH, et al. Skeletal muscle in polymyositis. Immunohistochemical study. *Arch Pathol Lab Med* 1979;103:310–13.

Learning points

- Primary pyomyositis can present as acquired torticollis.
- *Streptococcus constellatus* is a very rare cause of pyomyositis.
- MRI is the gold standard imaging modality to identify pyomyositis and should differentiate it from myositis, particularly if contrast enhanced.
- Early identification and treatment of pyomyositis with appropriate antibiotics may prevent the need for surgical drainage.

Copyright 2016 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow