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Submission template

TITLE OF CASE Do not include "a case report"

Pseudo appendicitis. Abdominal pain arising from thoracic spine dysfunction – a forgotten entity and reminder of an important clinical lesson

SUMMARY Up to 150 words summarising the case presentation and outcome (this will be freely available online)

Acute or chronic abdominal pain can be mimicked by thoracic spine dysfunction. However, it is comparatively rare and there is frequently a delay in its diagnosis that may lead to unnecessary surgery, or a patient's symptoms being undiagnosed or labelled psychosomatic.

The failure to associate thoracic spine dysfunction with abdominal pain persists, despite it first being recognised over 80 years ago. Two recent such cases are presented. The clinical presentation and diagnostic tests are described, with clear explanation of treatment and outcome.

The case for including the thoracic spine examination in the assessment of patients presenting with acute abdominal or post appendectomy pain that is of unexplained origin is made.

BACKGROUND Why you think this case is important - why did you write it up?

Abdominal pain is a common condition and around 40,000 people are admitted to hospital with appendicitis each year in England[1]. Approximately, one in every 13 people will develop abdominal pain at some point in their life, and whilst it can develop at any age, it is most common in those aged under 30[1].

Whilst appendicitis is the most common cause of abdominal pain, the American College of Surgeons recognize that other medical disorders may have similar symptoms, such as inflammatory bowel disease, pelvic inflammatory disease, gastroenteritis, urinary tract infection, right lower lobe pneumonia, Meckel's diverticulum, intussusception, and constipation[2]. Interestingly however, thoracic spine dysfunction is not described in their list of different potential symptom sources[2].

It is our aim therefore, to bring this entity to the attention of our general surgical colleagues who are not as familiar with musculoskeletal disorders such as thoracic spine dysfunction.

CASE PRESENTATION Presenting features, medical/social/family history

Two cases are presented with similar histories that were both referred to a pain management specialist from the general surgery team with ongoing pain post appendectomy. No visceral cause for symptoms could be found by the surgical team, and their source of symptoms had not been identified.

The first patient (Case 1) was a 19-year-old female who presented two-years after an appendectomy, with a history of pre-operative intermittent pricking pain over the right iliac fossa which had not resolved post surgery. She was review by two general surgeons, a colorectal surgeon, and a pain management consultant, and investigations via MRI, CT, Ultrasound, and an x-ray of the abdomen were all negative. The pathology report was normal. The "intractable pain" was labelled as psychosomatic.

Comment [TW1]: Grammar correction in accordance with comments from Reviewer 2

Comment [TW2]: Grammar correction in accordance with comments from Reviewer 2

Comment [TW3]: Added in response to comments from Reviewer 2

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The second patient (Case 2) was 25 years old and reported a 6-month history of right iliac fossa pain. He was diagnosed as having appendicitis and the appendix was removed laproscopically. Histology of the appendix was normal. Post surgery the pain continued and become more severe. He was reviewed by consultants in general surgery, colorectal surgery, gastroenterology, and radiology, with subsequent ultrasound, MRI, CT of the abdomen, a colonoscopy and a barium meal all found to be normal. He was left with 10cm^2 area of allodynia in the right iliac fossa.

INVESTIGATIONS If relevant

N/A

DIFFERENTIAL DIAGNOSIS If relevant

On examination, a thorough spinal assessment was completed for all patients by the pain management specialist, and in all cases a thoracic spine dysfunction was identified.

When both cases were examined in the prone position there was pain on posterior-anterior pressure to the right T11 and T12 transverse processes (right rotational force), but no pain found was found when this was repeated on the left side. This was accompanied by pain and tenderness in the right lower quadrant of their abdomen when they were both examined in the supine position.

For Case 2, in addition he was extremely tender on palpation over the right iliac fossa, and had exquisite para-spinal tenderness form the level of T1-L1

For both patients a diagnosis of right sided thoracic spine dysfunction at the levels of T11 and T12 was made in accordance with the diagnostic tests described by Maigne, Cope and Melnick[3,4,5].

In both cases, anterior cutaneous nerve entrapment syndrome (ACNES) was considered as a differential diagnosis due to the specific areas of abdominal tenderness on palpation. ACNES occurs when terminal branches of the lower thoracic intercostal nerves are entrapped in the abdominal muscles causing localized pain the abdomen. However, due to the accompanying spinal symptoms, a diagnosis of thoracic spine dysfunction was made for both patients.

TREATMENT If relevant

For Case 1 a therapeutic right paravertebral block (or dorsal root ganglion block) at the T11/12 level was performed under sterile conditions with x-ray control. Pain relief was obtained instantly.

Case 3 was treated with a right T11, T12, and L1 transforaminal dorsal root ganglion with local anaesthetic and steroid block. Pain relief was immediate and has remained.

OUTCOME AND FOLLOW-UP

Both cases remain pain free to date.

DISCUSSION Include a very brief review of similar published cases

Diagnostic parsimony and the counterbalance it finds in Hickam's dictum have very

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important implications in medical practice. Any set of symptoms could be indicative of a range of possible diseases and disease combinations; though at no point is a diagnosis rejected or accepted just on the basis of one disease appearing more likely than another.

The two cases described highlight the importance of considering thoracic spine dysfunction in patients with acute and/or chronic abdominal pain of non-visceral origin. The patients suffered pain for between 6-24 months following appendectomy, and were seen by several senior doctors and underwent various procedures before correct diagnosis and treatment was achieved. Had the diagnosis been considered initially by examination of the thoracolumbar junction and prompt referral made, failed appendectomy and considerable distress to the patient may have been avoided. However, there is no criticism of the surgical teams involved because common conditions occur commonly, and in the case of appendicitis no chance could be taken due to the life threatening need for prompt treatment if appendicitis was believed to be the source of symptoms.

However, this report is an important clinical reminder due to the ongoing post-operative pain experienced by the patients, and because the link between thoracic spine dysfunction and abdominal pain has not been reported in the literature since Ashby demonstrated effective treatment of abdominal pain of spinal origin with spinal injection in 1977[6]. Prior to this the association between thoracic spinal dysfunction with the symptoms of abdominal pain was first made by Ussher in 1933 when he described "The Viscerospinal Syndrome" [7]. Subsequently, Wills and Atsatt described five cases of pseudo appendicitis of spinal origin which they cured by injecting the thoracic spine when other treatments failed[8], and Harman and Young successfully treated cases of chronic appendicitis, chronic cholecystitis and renal colic by injecting one per cent procaine solution into tender areas of the lower thoracic spine and in the muscles of the abdominal wall, when all other treatments failed[9]. More recently, there has been acknowledgement in the literature of chronic non-visceral sources abdominal pain but with no explicit explanation of thoracic spine dysfunction[10], and in other articles hip and/or low back pain symptoms have been found to contribute to abdominal pain[11], and the abdominal wall has also been highlighted as the cause of abdominal pain[12].

LEARNING POINTS/TAKE HOME MESSAGES 3 to 5 bullet points – this is a required field

- Thoracic spine dysfunction should be considered as a source of symptoms for all patients presenting with abdominal pain
- Differential diagnosis and thorough examination of the thoracic spine should especially be considered for the patient presenting with acute or chronic abdominal pain whose symptoms are not originating from the appendix or other common causes of abdominal pain, or is presenting post-operatively
- Prompt referral to a pain management specialist for patients with thoracic spine dysfunction and abdominal pain can lead to effective diagnosis and treatment
- The delay in diagnosis and treatment of these cases may be indicative of a wider lack of awareness of the condition

Comment [TW5]: Grammar correction in accordance with comments from Reviewer 2

Comment [TW6]: Grammar correction in accordance with comments from Reviewer 2

REFERENCES Vancouver style (Was the patient involved in a clinical trial? Please reference related articles)

- 1. NHS Choices. Appendicitis [Internet]. NHS Choices. 2015 [cited 20 Feb 2016]. Available from: http://www.nhs.uk/conditions/Appendicitis/Pages/Introduction.aspx
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- 6. Ashby E.C. Abdominal pain of spinal origin. Annals of the Royal College of Surgeons of England. 1977; 59:242-246
- 7. Ussher N.T. Spinal Curvature-Visceral Disturbances. California and Western Medicine. 1933; 38(6): 423-428. Available from:

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- 11. Rodeghero J.R., Denninger T.R., Ross M.D. Abdominal Pain in Physical Therapy Practice:
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- 12. Gallegos N.C., Hobsley M. Recognition and treatment of abdominal wall pain. Journal of the Royal Society of Medicine. 1989;82:343-344

FIGURE/VIDEO CAPTIONS figures should NOT be embedded in this document

PATIENT'S PERSPECTIVE Optional but encouraged

Case 1 said "Thanks a million for all your help and support, without your expertise and identification of my problem, I probably wouldn't now have my little family". Case 3 said "I was discharged from hospital in so much pain, thank you for identifying and resolving my suffering which had disrupted my life for 10 months".

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