



Iatrogenic post-surgical tension pneumosyrinx—a first reported case

Shyam S. Swarna¹ · Josephine Jung^{2,3} · Steve Connor⁴ · Maurizio Belci¹ · Gordon Grahovac²

Received: 14 March 2022 / Accepted: 13 June 2022 / Published online: 27 June 2022
© The Author(s) 2022

Abstract

We describe the first ever-reported occurrence of a post-operative tension pneumosyrinx occurring after a resection of an intradural intramedullary spinal tumour in a 40-year-old patient. Post-operatively, he developed sudden onset paraplegia and imaging revealed a tension pneumosyrinx which was subsequently surgically decompressed. He made a gradual neurological recovery. This is an extremely rare complication with potentially long-lasting deleterious effects on patients' neurological status if not recognized. We aim to bring this pathology to the attention of our neurosurgical colleagues and share our surgical approach and management to assist those who may encounter this pathology in the future.

Keywords Iatrogenic · Pneumosyrinx · Pneumorrhachis · Spinal cord tumour · Tension

Abbreviations

CSF	Cerebrospinal fluid
CT	Computer tomography
IDIM	Intradural intramedullary
IONM	Intra-operative neuromonitoring
ISNCSCI	International Standards for Neurological Classification of Spinal Cord Injury
MRI	Magnetic resonance imaging
MRC	Medical Research Council
SCIM	Spinal Cord Independent Measure

pneumosyrinx and review the existing literature on pneumorrhachis which is a condition closely associated with this. The authors describe the case of a 40-year-old who presented with an acute neurological deterioration after resection of a cervico-thoracic intradural intramedullary (IDIM) tumour due to severe tension pneumosyrinx caused by CSF entrapment in the arachnoid space. To the authors' knowledge, this is the first case of iatrogenic/post-surgical tension pneumosyrinx to be described in the literature. The patient required a return to theatre for exploration of the cervico-thoracic wound and release of the adhesive arachnoid tissue which was causing a one-way valve effect and compression of the spinal cord.

Introduction

Pneumosyrinx is a rare phenomenon defined as migration of air within the central canal of the spinal cord. In this case report, we discuss the presentation of a patient with

Case presentation

A 40-year-old male presented with a 3-year history of paraesthesia mainly affecting the fingers in his right hand. Magnetic resonance imaging (MRI) of the cervical spine revealed a cervico-thoracic IDIM tumour spanning from the C4 to T2 level (Fig. 1).

The patient underwent a laminectomy for complete tumour resection with intra-operative neuromonitoring (IONM). The myelotomy was performed on the dorsal midline of the spinal cord, immediately identifying the superficial tumour which was encased by the thin spinal cord. The tumour (from C3 down to T2) was carefully dissected with the cystic component drained (no excessive amounts of CSF were drained during this procedure). The myelon was not sutured during the initial surgery and dural closure was

This article is part of the Topical Collection on *CSF Circulation*

✉ Josephine Jung
Josephine.Jung@nhs.net

¹ National Spinal Injuries Centre, Stoke Mandeville Hospital, Aylesbury, UK

² Department of Neurosurgery, King's College Hospital NHS Foundation Trust, Denmark Hill, London SE5 9RS, UK

³ Institute of Psychiatry, Psychology and Neurosciences, King's College London, London, UK

⁴ Department of Neuroradiology, King's College Hospital, London, UK



Fig. 1 Pre-operative T2-weighted sagittal MRI of the cervical spine showing a hyperintense cystic intradural intramedullary lesion spanning from C4 to T2 and expanding the cord

performed with 5.0 prolene with copious irrigation. Post-operatively, the patient displayed a new-onset mild right grip and right leg weakness with 4 out of 5 power on the Medical Research Council (MRC) scale in line with IONM findings. Ten hours after surgery, the patient started complaining of increased neck pain followed by haemodynamical instability and development of complete loss of power in both lower limbs and the right upper limb (0 out of 5 on MRC) with only preserved shoulder shrug bilaterally (4 out of 5 on

MRC) and left biceps flexion (3 out of 5 on MRC). He was also insensate to the indwelling urinary catheter.

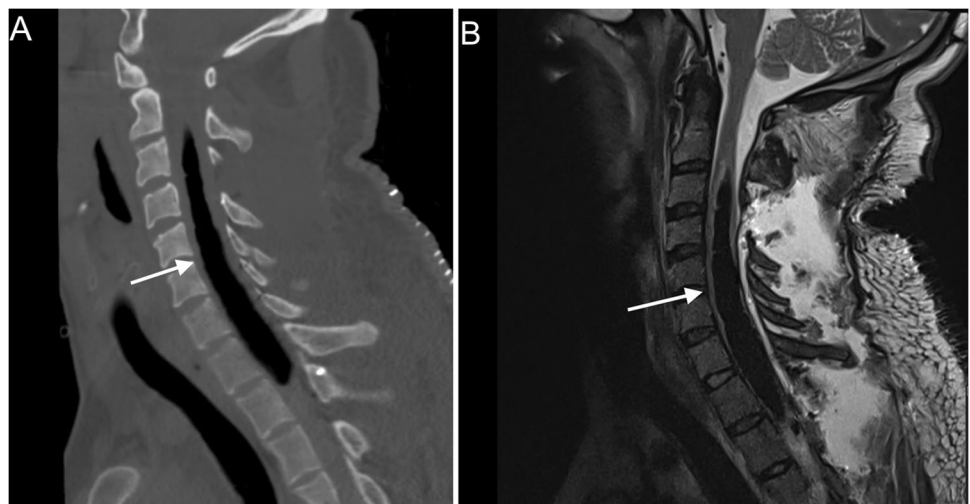
Post-operative computer tomography (CT) and MRI scan of the neck showed a long segment of tension pneumosyrinx (Fig. 2A and B) for which he subsequently underwent emergency re-exploration. CT brain imaging was normal and there was no pneumocephalus.

After opening of the dura, the cord herniated through the dural opening due to expansion of the arachnoid in front of the cord. After releasing the entrapped cerebrospinal fluid (CSF) from the ballooning arachnoid space in front of the cord (similar to an arachnoid cyst), it was decompressed and CSF flow re-established.

The patient made a gradual recovery and was transferred for further neuro-rehabilitation. He had residual weakness involving the right arm and right lower limb with ongoing bladder and bowel dysfunction. Follow up MRI scan showed resolution of the tension pneumosyrinx (Fig. 3). The tumour was diagnosed as an ependymoma WHO grade II for which the patient did not require any further adjuvant treatment.

On admission to the tertiary neuro-rehabilitation unit, he had residual weakness predominantly affecting the right upper and lower limb with motor sparing on the left as well as ongoing sphincter dysfunction. His neurological level according to the International Standards for Neurological Classification of Spinal Cord Injury (ISNCSCI) was C2 ASIA Impairment Score D. During neuro-rehabilitation, he made excellent gains in all aspects of the Spinal Cord Independent Measure (SCIM) which includes self-care (feeding, grooming, bathing, and dressing), respiration and sphincter management, and patient mobility (transfers to/from bed and mobility indoors/outdoors). The SCIM on admission was 41 and subsequently improved to 86 at discharge. The patient also improved to independency with bladder and bowel function and was mobilizing with walking aids at 12 months post surgery.

Fig. 2 **A** Post-operative cervical sagittal CT demonstrating a tension pneumosyrinx extending from C3 to the level of T2; **B** Post-operative sagittal T2-weighted MRI of the cervical spine demonstrating a hypointense lesion at the same level which is consistent with a tension pneumosyrinx



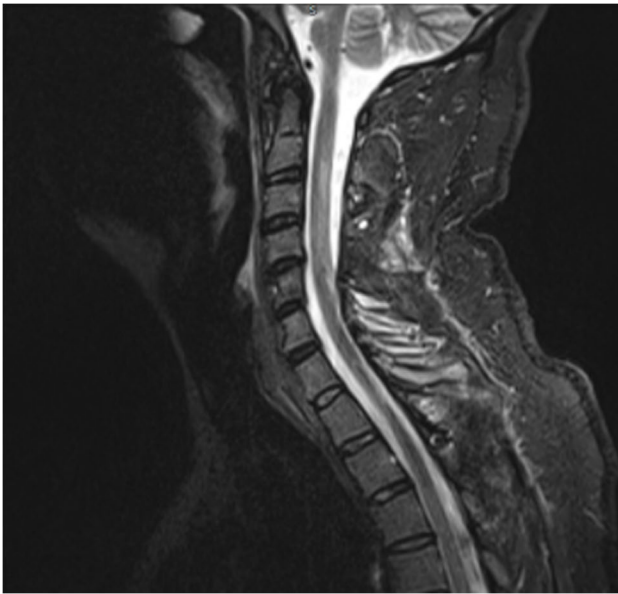


Fig. 3 T2-weighted MRI scan 14 weeks post re-exploration following tension pneumosyrinx

Discussion

Pneumosyrinx is an extremely rare complication. Upon search Pubmed, using the Boolean search term “pneumosyrinx,” there was only one article by Yanagawa et al. [19] which reported this condition. Pneumosyrinx was associated with pneumorrhachis. Pneumosyrinx is defined as entrapment of air or gas within the central canal of the spinal cord. Pneumorrhachis is the entrapment of air within the spinal canal. The previously published case report [5] describes a 26-year-old female who was involved in a road traffic accident. She was comatose on admission to the hospital and sadly died within 2 h of admission to hospital. Pneumosyrinx in this case was discovered on CT imaging of the thorax and abdomen and demonstrated air bubbles in the spinal canal between the second and fifth thoracic vertebrae. This was thought to be caused by migration of air into the central canal of the spinal cord. It was suspected that the existence of the pneumosyrinx could be an indirect sign of either spinal cord or medullary trauma with an open spinal canal injury, pneumocephalus, or pneumothorax. Pneumorrhachis was caused directly by the open injury. In other studies, direct open injury to the spinal canal, air migration into the spinal canal due to pneumomediastinum with barotrauma, or pneumothorax have been reported [3, 16, 19].

On Pubmed search using the Boolean search term “pneumorrhachis,” this was reported in 202 articles including both human and animal studies. Air in the spinal canal was first described by Gordon and Hardman in 1977 [9]. Pneumorrhachis has been subclassified depending on the location of

air which can be located either extradurally (epidural) or intradurally (in the subarachnoid space). The distinguishment of epidural pneumorrhachis is important as there are different pathophysiological mechanisms which may have different clinical implications and potential consequences. Pneumorrhachis can also occur in trauma without meningeal tears, with air simply accumulating in the epidural space [6]. The first report of epidural pneumorrhachis caused by blunt thoracic trauma was in a patient with bronchial rupture and pneumomediastinum following a road traffic accident [18]. The amount of air in epidural pneumorrhachis is generally small, as the driving pressure of the pneumothorax or pneumomediastinum is not high enough to push large volumes of air into the epidural space [6, 8]. The presence of pneumorrhachis signifies potential significant trauma, it however does not require specific treatment beyond the management of the precipitant such as a pneumothorax [5, 8, 10, 13].

In contrast, subarachnoid pneumorrhachis is usually associated with pneumocephalus [8]. The volume of air might raise intraspinal and by extension, intracranial pressure for which acute neurosurgical or interventional radiological treatment is required to facilitate drainage or to repair the dural tears [12, 20]. Less commonly, it may be atraumatic and cases have been reported where pneumorrhachis was due to violent coughing, forceful emesis, cardiopulmonary resuscitation, malignancy, infection, epidural anaesthesia [11], or marijuana or cocaine inhalation [1, 14]. Moreover, spontaneous pneumorrhachis has also been reported [7].

Some tumours are commonly associated with syringomyelia: two-thirds of haemangioblastomas and half of spinal ependymomas have an associated syrinx [2, 15, 17]. A possible explanation for syrinx development in association with IDIM tumours is that the cystic cavity is part of the tumour itself [17].

This is the second case of pneumosyrinx reported in the literature and the first one that is iatrogenic and associated with tension pneumosyrinx. In our patient, it was following the resection of an IDIM tumour. There are several possibilities regarding the mechanism for this to have happened. It could have occurred by the same mechanism leading to syringomyelia which is due to mechanisms affecting fluid dynamics referred to as “slosh” and “suck” [4]. The “slosh” effects are due to increased epidural venous flow triggered by activities such as coughing and sneezing. In this case, the potential culprit could have been positive pressure ventilation during the general anaesthetic as part of the operative procedure. The resection of the tumour formed a dead space within the cord structure thereby creating a vacuum effect. The second mechanism, “suck,” is the result of partial subarachnoid blockage. As the fluid is initially forced up due to increased epidural venous pressure, it returns slowly creating a pressure gradient across the partial subarachnoid block with negative pressure caudally to it [4]. Once the tumour

was removed, however, venous congestion and therefore epidural pressure should have decreased and no excessive amount of positive pressure ventilation was used during the surgery. A second possibility is that air was entrapped during wound closure; however, the post-operative MRI cervicothoracic spine showing the tension pneumosyrinx did not reveal any air tracking along the surgical wound and no surgical drain was used. The third and most plausible hypothesis is that air was already entrapped intradurally during surgery. The head and cervical spine are often positioned lower than the thoracic spine during prone cervicothoracic surgery. During the tumour resection, air may have become entrapped in a distal part of the thoracic dural sack which was not visible at the time of dural closure. With a post-operative change in position from probe to supine and sitting up in bed, this air could have then migrated upwards and entered the large pial defect causing the gradual increase in pressure within the cord and resultant neurological deterioration. A mechanism involving entrapment of intradural air and downwards migration seems unlikely since the CT brain did not demonstrate any pneumocephalus.

Conclusion

Pneumosyrinx is an extremely rare condition with potentially long-lasting deleterious effects on patients' neurological status. We present the first case of post-operative tension pneumosyrinx to the spine. We aim to bring this pathology to the attention of our worldwide neurosurgical colleagues and share our surgical approach and management to assist those who may encounter this pathology in the future.

Funding JJ is funded by a National Institute for Health Research (NIHR) Clinical Lectureship. The funder had no input into the study conception, design, analysis, or reporting. The views expressed are those of the author(s) and not necessarily those of the NIHR or the Department of Health and Social Care.

Declarations

Consent to participate The patient did not undergo any additional procedures and informed consent was taken prior to all procedures undertaken as per the standard of care pathway.

Conflict of interest The authors declare no competing interests.

Open Access This article is licensed under a Creative Commons Attribution 4.0 International License, which permits use, sharing, adaptation, distribution and reproduction in any medium or format, as long as you give appropriate credit to the original author(s) and the source,

provide a link to the Creative Commons licence, and indicate if changes were made. The images or other third party material in this article are included in the article's Creative Commons licence, unless indicated otherwise in a credit line to the material. If material is not included in the article's Creative Commons licence and your intended use is not permitted by statutory regulation or exceeds the permitted use, you will need to obtain permission directly from the copyright holder. To view a copy of this licence, visit <http://creativecommons.org/licenses/by/4.0/>.

References

1. Atmaca Temrel T, Şener A, İçme F et al (2015) Subcutaneous emphysema, pneumomediastinum, and pneumorrhachis after cocaine inhalation. *Case Rep Emerg Med* 2015:134816
2. Barnett HJM, Jousse AT, Ball MJ (1973) Historical introduction, in Barnett HJM, Foster JB, Hudgson (eds): *Syringomyelia*. London, W.B.Saunders : 261 – 301
3. Bellil M, Hadhri K, Tebourbi A (2015) Cervical pneumorrhachis secondary to a basal skull fracture. *Neurol India* 63(5):800–801
4. Biyani A, el Masry WS (1994) Post-traumatic syringomyelia: a review of the literature. *Paraplegia* 32(11):723–731
5. Copetti R, Mattei G (2010) Traumatic epidural pneumorrhachis. *BMJ Case Rep* 2010:bcr0120102692
6. Derner M, Drugová B, Hořejší L, Skvára D, Druga R (2011) Massive pneumorrhachis, pneumocephalus and pneumoopticus following thoracic trauma and avulsion of the brachial plexus: case report and review of the literature. *Prague Med Rep* 112(1):56–66
7. Fonseca AZ, Santin S, Ribeiro M (2016) Spontaneous pneumorrhachis. *Am J Emerg Med* 34(12):2465.e3–2465.e4
8. Goh BK, Yeo AW (2005) Traumatic pneumorrhachis. *J Trauma* 58(4):875–879
9. Gordon IJ, Hardman DR (1977) The traumatic pneumomyelogram. A previously undescribed entity. *Neuroradiology* 13(2):107–108
10. Hadjigeorgiou GF, Singh R, Stefanopoulos P, Petsanas A, Hadjigeorgiou FG, Fountas K (2016) Traumatic pneumorrhachis after isolated closed head injuries: an up-to-date review. *J Clin Neurosci* 34:44–46
11. Hsieh XX, Hsieh SW, Lu CH et al (2015) A rare case of pneumocephalus and pneumorrhachis after epidural anesthesia. *Acta Anaesthesiol Taiwan* 53(1):47–49
12. Kara H, Akinci M, Degirmenci S, Bayir A, Ak A (2015) Traumatic pneumorrhachis: 2 cases and review of the literature. *Am J Emerg Med* 33(6):861.e1–3
13. Mangiardi JR, Brisson P, Weitzner I, Bednar M (1987) Traumatic intraspinal pneumocele. *Surg Neurol* 27(6):587–588
14. Oertel MF, Korinth MC, Reinges MH, Krings T, Terbeck S, Gilsbach JM (2006) Pathogenesis, diagnosis and management of pneumorrhachis. *Eur Spine J* 15(Suppl 5):636–643
15. Poser CM (1956) The relationship between syringomyelia and neoplasms. Charles C Thomas, Springfield
16. Rathore FA, Gill ZA, Amjad Yasin MM (2012) Pneumorrhachis of thoracic spine after gunshot wound. *J Coll Physicians Surg Pak* 22(8):529–530
17. Samii M, Klekamp J (1994) Surgical results of 100 intramedullary tumors in relation to accompanying syringomyelia. *Neurosurgery* 35(5):865–73; discussion 873
18. Scialdone CJ, Wagle W (1990) Intraspinal air: an unusual manifestation of blunt chest trauma. *Clin Imaging* 14(1):59–60

19. Yanagawa Y, Jitsuiki K, Yoshizawa T, Ishikawa K, Ohsaka H (2016) Traumatic pneumosyrinx and pneumorrhachis. *J Neuro-radiol* 43(5):358–359
20. Yuce I, Akbulut A, Ergun F, Sade R, Kantarci M (2016) Massive lumbar pneumorrhachis after severe cranial trauma. *Spine J* 16(1):e3

Comments

This is an unusual case of a postoperative pneumosyrinx after removal of intradural intramedullary tumor. The authors report on one other case that had pneumosyrinx, as well. This condition is extremely rare, but nevertheless should be considered in the differential after increased postoperative neurological deficit.

Publisher's note Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.

Volker Sonntag
Phoenix, AZ, USA