World Journal of Advanced Research and Reviews
eISSN: 2581-9615 CODEN (USA): WJARAI
Cross Ref DOI: 10.30574/wjarr
Journal homepage: https://wjarr.com/

(CASE REPORT)

Check for updates

Apical pulmonary hydatid cyst: About a rare cause of Pancoast syndrome

Bennani H [1,*], Benchekroun Z [1], Bennani M M [3], Akkamar A [2], Bouardi N [1], Haloua M [2], Alaoui Lamrani M Y [1], Boubbou M [2], Maâroufi M [1] and Alami B [1]

[1] *Department of radiology, CHU Hassan II, Fès, Morocco.*
[2] *Department of Mother and child radiology, CHU Hassan II, Fès, Morocco.*
[3] *Université libre de bruxelles Brussels, Belgium.*

World Journal of Advanced Research and Reviews, 2023, 19(03), 072–075

Publication history: Received on 01 July 2023; revised on 28 August 2023; accepted on 31 August 2023

Article DOI: https://doi.org/10.30574/wjarr.2023.19.3.1604

Abstract

Objectives: Our purpose is to illustrate the radiological aspects of an apical hydatid cyst as a rare cause of Pancoast Tobias syndrome.

Material: We report the case of a 35 years-old man, who was admitted to our department with a Pancoast tobias syndrome.

Results: The patient presented to our department with a history of shoulder and right chest pain radiating downward toward the ulnar side of his homolateral arm. He also reported hemoptysis and hydatidemesis (hydatid cysts and membranes in the vomitus).

Radiological investigations suggested the diagnosis of hydatid cyst of the left lung apex which was confirmed by surgical excision and pathological examination of the lesion. This case highlights an uncommon etiology of Pancoast syndrome which might mislead physicians in their practice.

Conclusion: In conclusion, a primary hydatid cyst should be considered in the differential diagnosis of Pancoast's syndrome especially in endemic regions.

Unlike Pancoast tumors, this benign lesion has a very favourable prognosis and good surgical results.

Keywords: Pancoast syndrome; Hydatid cyst; Imaging

1. Introduction

Pancoast syndrome is a rare syndrome commonly caused by bronchogenic tumors in the pulmonary apex. It is characterized by shoulder pain radiating toward the axilla or ulnar aspect of the arm, atrophy of the arm and hand muscles and homolateral Horner syndrome. Benign causes of Pancoast syndrome are very rare. We report this uncommon case of Pancoast syndrome related to lung apex hydatid cyst occurring in a 35 year- old man.

2. Observation

We report the case of a 35-year-old man with history of pulmonary hydatid cyst surgery in 2019; who presented to the emergency for dyspnea.

* Corresponding author: Bennani H

He complained of a one month history of shoulder and right chest pain radiating downward toward the ulnar side of his homolateral arm. He also reported hemoptysis and hydatidemesis (hydatid cysts and membranes in the vomitus).

On presentation, the patient's vital signs were stable, with a blood pressure of 120/70mm Hg, heart rate of 85 bpm, and no fever.

Physical examination revealed Horner's syndrome of the right eye, namely a ptosis and enophtalmos. The diagnosis of horner syndrome was made.

A chest X-ray showed a right apical opacity. A cervicothoracic computed tomography scan revealed a large right apical cystic mass containing membranes and gaz with cervical extension and inominate venous trunk compression. (figure 1). Given the presence of neurological signs a magnetic resonance imaging (MRI) of the cervico thoracic region was performed revealing a large cystic mass of the right pulmonary apex which was hypointense on T1 weighted images. The lesion was hyperintense on T2 weighted images and delineated by a hypointense rim and containing hypointense membranes that did not enhance. There was also a compression of the axillary nerve.

Surgical exploration by an anterior cervicothoracic approach revealed a hydatid cyst in the apical portion of the lung apex. The cyst was completely resected. Postoperative finding was satisfactory with total recovery. Adjuvant Albendazole based chemotherapy was administrated.

3. Discussion

The Pancoast syndrome was first described by Pancoast in 1924 , as shoulder and arm pain which is radiated to ulnar aspect of arm and fore- arm, atrophy of the intrinsic muscles of the hand, associated with ipsilateral Horner syndrome (which is characterized by ipsilateral anhidrosis of the face, miosis, and ptosis with narrowing of the palpebral fissure secondary to paralysis of the Muller muscle). [1].

It is commonly associated with primary lung carcinoma in the superior pulmonary sulcus [1-4]. Other malignancies have been described such as thyroid cancer, cervical cancer metastasis, and multiple myeloma [3]. Infectious etiologies like aspergillomas, fungal abscess, staphylococcal or lymphoid granulomatosis have been reported but still are very rare [3,5,6].

However, this syndrome is very rarely associated with a pulmonary apex hydatid cyst even in endemic areas like morroco .

The manifestations are corelated with the anatomy of the superior sulcus which contains from front to back; the subclavian vein, then the subclavian artery as well as the trunks of the brachial plexus in the interscalene triangle and then the costovertebral groove as well as the roots of the brachial plexus and the stellate ganglion. [1,4].

Imaging is the main tool for diagnosis and can relate the radiologic features of the anatomy to surgical landmarks when planning the surgical approach [1-4].

Classic chest radiography shows an apical opacity which suggests a large diagnosis panel. Cross sectional imaging allows an etiological approach.

Computed tomography (CT) is the main tool for radiologic diagnosis [4]. It shows a cystic lesion with no enhancement, that may contain membranes and daughter cysts [4,7]. Complicated cysts are described as ruptured or infected cysts, many signs may suggest a contained rupture like 'air crescent sign', 'inverse crescent sign', and 'air bubble sign' like it was the case in our patient. Infected cysts show similar radiological manifestations as pyogenic abscesses [7].

On MRI, hydatid cysts are hypointense on T1 weighted images and hyperintense on T2 weighted images with sometimes a rim sign which consists of a low signal intensity rim surrounding the cyst and corresponding to the pericyst [4,6]. This modality is also better to delineate soft tissue involvement, invasion of brachial plexus, vasculature, and spinal involvement as part of the resecability assessment [2].

The differential diagnosis mainly includes tumors, but also hematologic conditions, infectious diseases, neurogenic thoracic outlet syndromes [4].

This pathology requires surgical management to decompress neurologic structures by total removal of the cysts [4] and Paulson thoracotomy seems to be suitable for some authors [4].

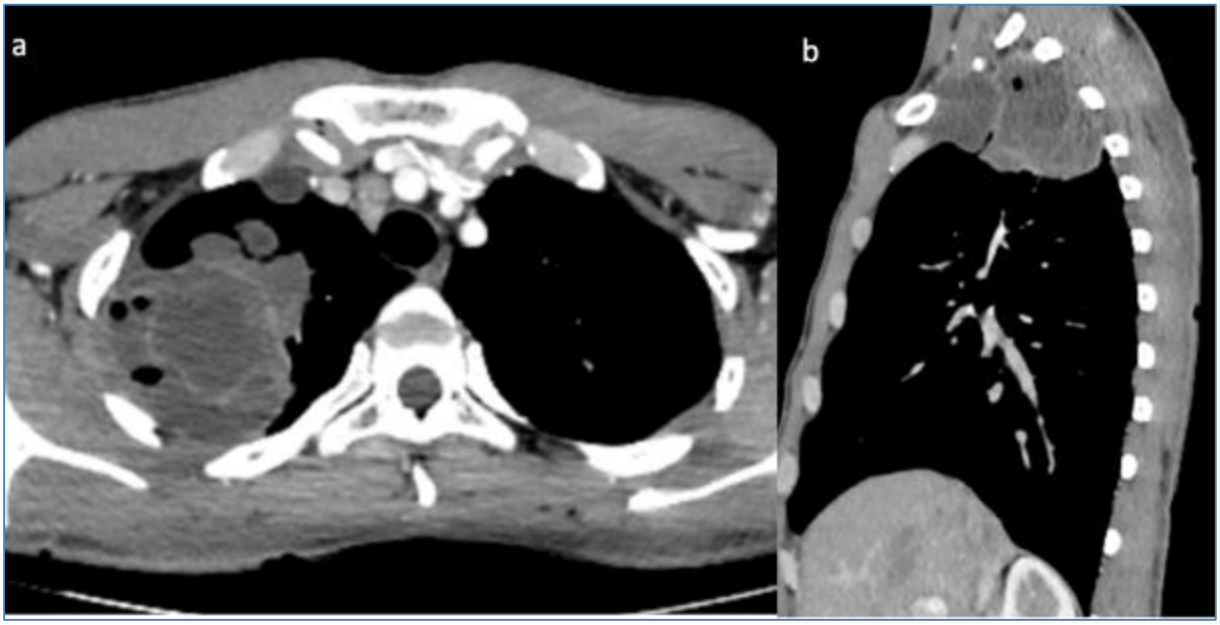

Figure 1 Axial (a) and sagittal (b) thin-section enhanced cervicothoracic computed tomography scan showing a large right apical cystic mass containing membranes and gaz with cervical extension and inominate venous trunk compression

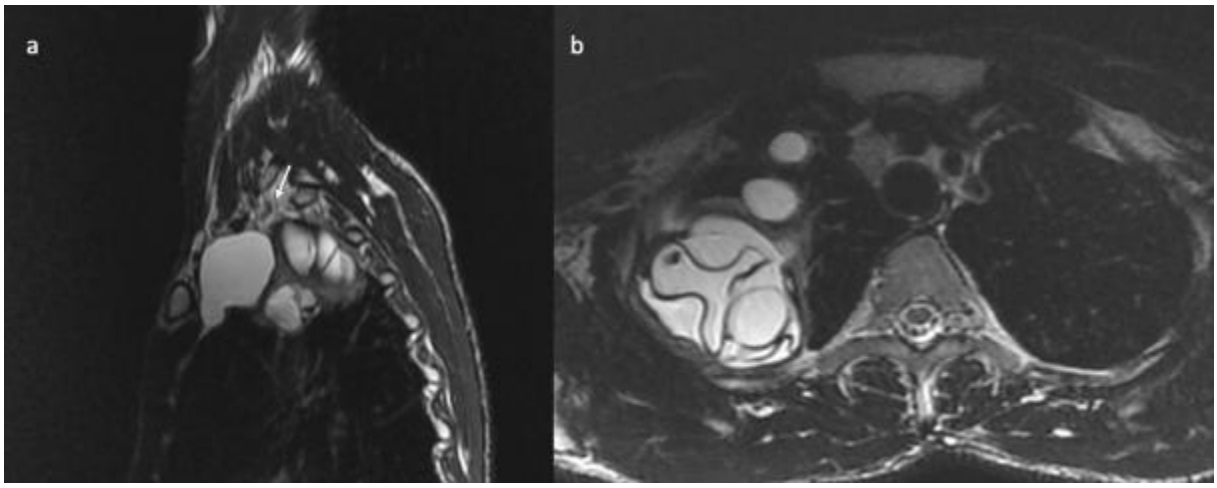

Figure 2 Sagittal (a) and axial (b) MRI T2 weighted images revealing a large cystic mass of the right pulmonary apex which is hyperintense on T2 weighted images and delineated by a hypointense rim and containing hypointense membranes. There was also a compression of the axillary nerve (white arrow)

4. Conclusion

In conclusion, a primary hydatid cyst should be considered in the differential diagnosis of Pancoast's syndrome especially in endemic regions.

Unlike Pancoast tumors, this benign lesion has a very favourable prognosis and good surgical results.

Compliance with ethical standards

Disclosure of conflict of interest

No conflict of interest to be disclosed.

Statement of ethical approval

If studies involve use of animal/human subject, authors must give appropriate statement of ethical approval. If not applicable then mention 'The present research work does not contain any studies performed on animals/humans subjects by any of the authors'.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

References

[1] Bruzzi JF, Komaki R, Walsh GL, Truong MT, Gladish GW, Munden RF, Erasmus JJ. Imaging of non-small cell lung cancer of the superior sulcus: part 1: anatomy, clinical manifestations, and management. Radiographics. 2008 Mar-Apr; 28(2):551-60; quiz 620. doi: 10.1148/rg.282075709. PMID: 18349457.

[2] Villgran VD, Chakraborty RK, Cherian SV. Pancoast Syndrome. 2023 Jan 30. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2023 Jan–. PMID: 29489146.

[3] Rabiou S, Issoufou I, Belliraj L, Ammor FZ, Lakranbi M, Ouadnouni Y, Smahi M. Pancoast syndrome revealing a hydatid cyst of the lung. QJM. 2016 May; 109(5):337-8. doi: 10.1093/qjmed/hcv192. Epub 2015 Oct 15. PMID: 26475600; PMCID: PMC4888328.

[4] Dao I, El Mostarchid B, Onen J, Mandour C, El Asri CA, Boucetta M. Pancaost syndrome related to hydatid cyst. Pan Afr Med J. 2013 Mar 27; 14:118. doi: 10.11604/pamj.2013.14.118.1754. PMID: 23717731; PMCID: PMC3664867.

[5] Das A, Choudhury S, Basuthakur S, Das SK, Mukhopadhyay A. Pancoast's Syndrome due to Fungal Abscess in the Apex of Lung in an Immunocompetent Individual: A Case Report and Review of the Literature. Case Rep Pulmonol. 2014; 2014:581876. doi: 10.1155/2014/581876. Epub 2014 Sep 11. PMID: 25302130; PMCID: PMC4180649.

[6] Zarca-Diaz de la Espina MAZ, Lopez-Menendez C, Ruiz-Martinez R, Molino-Trinidad C. Pulmonary actinomycosis with thoracic soft tissue mass: a rare onset form. Eur J Radiol 2001; 37:195-199.

[7] Garg MK, Sharma M, Gulati A, Gorsi U, Aggarwal AN, Agarwal R, Khandelwal N. Imaging in pulmonary hydatid cysts. World J Radiol. 2016 Jun 28; 8(6):581-7. doi: 10.4329/wjr.v8.i6.581. PMID: 27358685; PMCID: PMC4919757.