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Case Report(s)

A thirty eight year old man presented with difficulty in breathing, cough with brick red expectoration and heaviness in chest for ten days. A physician from another hospital had performed thoracocentesis on him following chest radiograph (figure 1) for these complaints. After repeating the radiograph (figure 2) he was referred to our tertiary care center for further management. We performed computed tomography of thorax with contrast, which showed bilateral iatrogenic pneumothorax with presence of cyst on both the sides showing serpent sign, double arch sign and air-bubble sign. He was advised surgery but was unwilling for the surgical intervention. He was treated with oral albendazole 400 mg for 3 weeks with 1 week gap for 3 cycles. Repeat chest radiograph showed decreased left sided cyst and right lower lobe thick walled cavity. To assess the activity of the disease, positron emission radiography (PET) scan was performed which showed metabolically active cystic lesion in anterior segment of left upper lobe and lingular lobe, showing multiple crumpled membranes within and surrounding collapse consolidation. PET scan also shows metabolically inactive large cavity in right lower lobe, possibly communicating with segmental bronchus with surrounding collapse consolidation.

Discussion

Hydatid disease is a parasitic infestation of humans caused by Echinococcus granulosus and multilocularis, which produce cystic lesions in any part of body. Dogs and some wild carnivores are definitive hosts. It spreads in incidental hosts like human by direct contact with infected dogs. Hydatid cyst consists of three layers – the outermost pericyst made of fibrous tissue represents the response of the host to the parasite, middle layer of laminated membrane (ectocyst), and innermost layer of germinal epithelium (endocyst) which secretes the hydatid fluid internally and laminated membrane externally producing new generation of parasites. Detachment of the endocyst with its collapse results into serpent sign. The rupture may be related to decreasing intracystic pressure, degeneration, host response, trauma, or response to therapy. In our case it was due to iatrogenic rupture of the cyst which could have been life threatening. It is important to be aware about possibility of hydatid cyst when radiograph shows ‘elevated dome of diaphragm’ and ‘air crescent sign’ and be aware about possible complication related to aspiration. Ultrasound, CT and magnetic resonance imaging (MRI) are all appropriate for imaging of the membranes floating in partially fluid filled hydatid cyst. This presence of floating membrane or serpent sign is pathognomonic of ruptured hydatid cyst. Our patient in addition showed “sign of the double arch” or “cyst-within-a-cyst” which is also diagnostic of hydatid cyst. Once rupture of the cyst begins, air penetrating the interior of the cyst may outline the inner surface of the exocyst, producing parallel arches of air that are referred to as “double arch” or “Cumbo’s sign”. Air bubble sign also seen in mediastinal window as double, rounded radiolucent areas within the periphery of a solid mass lesion. It is seen when there is dissection of air between the pericyst and parasitic membrane, due to erosion of a bronchiole by an expanding cyst.

The “air bubble” sign which is a relatively newly recognized radiological sign is reported to be very sensitive and specific (85.7% sensitivity and 96.6% specificity) in establishing diagnosis of ruptured, infected hydatid cyst.

PET scan role in hydatid cyst is to support the diagnosis in an established case (ruptured or infected or both), to show the extent of disease and to help in monitoring medical treatment in patient with contra indication to surgery or refused surgery. Thus, based on PET scan patient was advised removal of left lower lobe hydatid cyst. He declined the surgery. However, PET scan enabled us to decide about surgery. Thus, FDG-PET-CT can support the diagnosis in an established case of hydatid, either ruptured or infected or both, to show the extent of disease and to help in monitoring medical treatment in patient with contraindication to surgery or those refused surgery.

Familiarity with the crescent sign, serpent sign and double arch sign of hydatid cyst may prevent diagnostic delay and decrease the risk of
life-threatening complications. It is important to do PET scan for hydatid cyst do decide the activity of disease and response to medical management.

Acknowledgements

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References


Illustrations

Illustration 1

Figure 1: Hydatid cysts with B/L Pneumothorax

Illustration 2

Figure 2: Hydatid cysts with B/L Pneumothorax
Illustration 3

Figure 3: Partial improvement on medical management

Illustration 4

Figure 4: Improvement with medical management
Illustration 5

Figure 5: CECT Showing Serpent Sign

Illustration 6

Figure 6: Double Arch Sign
Illustration 7

Figure 7: Air Bubble Sign

Illustration 8

Figure 8: FDG-PET Showing Metabolically Cyst