



## INTRAHEPATIC FOETUS IN FOETU IN ADULT MALE: A CASE REPORT AND REVIEW OF THE LITERATURE

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Article Info	ABSTRACT
<p>Received 19/06/2015 Revised 27/06/2015 Accepted 10/07/2015</p>	<p>Foetus in foetu is one of the rarest pathological conditions due to abnormal embryogenesis resulting unequal division of totipotent cells of blastocyst. Basically it is due to aberration of monozygotic diamniotic twin where a malformed foetus is formed as small cellular mass in the body of a more mature sister embryo. Usually such conditions come in notice during infancy and childhood but very rarely it may incidentally found in adulthood. Majority are found retroperitoneal in upper abdominal region but as reported rarely it may be located at sacrococcygeal region, intrathoracic or mediastinum, pelvis, intra cranial, intraventricular, intrarenal, intraoral, lymph nodes, adrenal gland, neck, undescended testis and scrotum. Its intrahepatic location is reported earlier only by few authors especially during infancy but not reported in adulthood. In this article we present a case of intrahepatic foetus in foetu in a 30 years male which was initially diagnosed as intrahepatic teratoma or partially healed hydatid cyst complicated by gas forming organism. Ultrasonography and CT scan showed a well defined cavitory mass of approximately 8×6 cm size with few calcifications in the lower part of the right lobe of liver. Complete surgical excision under general anaesthesia was done and after macroscopic and microscopic analysis accompanied by review of literature confirmed the mass had features consistent with a foetus in foetu.</p>
<p><b>Key words:</b> Foetus in foetu, Intrahepatic foetus, Intrahepatic teratoma.</p>	

### CASE REPORT

A 30 years old male was admitted in our institute with complaint of pain abdomen followed with mild fever, cough and vomiting since last three months. The pain was radiated to right shoulder followed by bilious vomiting resulting in relief from it. The patient claimed to be non alcoholic but had a history of twin pregnancy in his family. Except mild leucocytosis and slightly increase SGPT his other routine investigations were within normal limit. His report of  $\alpha$ -fetoprotein was also normal.

On ultrasonography the patient revealed an echogenic well defined mass of size 6.3 × 8.7 cm with hypo echoic brim and few calcification adjacent to gall bladder fossa posterior to inferior vena cava while contrast enhanced tomography [fig: 1] showed a large 8 × 6 cm

thick walled cavitory mass seen in the lower part of the right lobe of liver which was compressing the superior pole of right kidney. The mass contained solid calcified component and extensive air pocket. The wall seemed contiguous with liver parenchyma and was diagnosed initially as intrahepatic teratoma or partially healed hydatid cyst complicated by gas forming organism. No other focal lesion was seen in liver.

After proper evaluation and under general anaesthesia exploratory laparotomy was done where an ill defined mass in the lower part of the right hepatic lobe was found which was dissected carefully and excised completely with securing proper homeostasis. Finally the patient recovered uneventfully. The excised mass grossly showed strands of hair admixed with tissue like structure



and body parts [Fig: 2] which was confirmed histopathologically as foetus like structure [Fig: 3].

## DISCUSSION AND CONCLUSION

Foetus in foetu is a rare entity which was first described by Johann Fedrich Meckel [1] in the late 18<sup>th</sup> century and first time this term was quoted by R H Lewis [2] and first case was reported by George William Young [3] and later G W Nicholson and R A Willis formulated its diagnostic criteria [4, 5]. It is found in one out of 500000 of cases [6- 8] with male predominance [9, 10] but some authors also reported equal sex ratio [11]. Most of the cases are found as a single parasitic foetus but there have been reported multiple fetuses [12-15]. Till now approximately 200 cases have been reported worldwide and most of them have been detected before 18 months of age [16, 17]. Majority of the cases are discovered as retroperitoneal mass (80%) and commonly located in abdomen [5] (70%) but many authors reported that it may occur at sacrococcygeal region [9-11,17,33] orocervical region [18], intrathoracic [19], lung or mediastinum [19, 20] pelvis [21] and unlikely site of occurrence are intra cranial [15,22] intraventricular [23,24] intrarenal [25] intraoral [26], lymph nodes [25], adrenal gland [25, 27], neck [28], undescended testis [29] and scrotum [9-,11,30]. Only few authors reported in infants its intrahepatic location as Al-Baghdadi R in 1992 first reported a foetal sac which was found in the right lobe of liver [31] and later Magnus K. G. *et al* in 1999 found multiple fetuses in foetu which were removed surgically by hemihepatectomy [32] but present case of intrahepatic foetus in foetu is a unusual and to our knowledge such condition is not reported in adulthood.

Due to awareness of routine antenatal care and increasing use of ultrasound, nowadays most of the cases of foetus in fetu have been identified earlier during infancy or intrauterine period. The oldest reported case was a 47 years old male [33] and reasons for such late reporting is lack of appropriate imaging facilities in many places and delayed presentation of symptoms. Its clinical manifestations in early childhood are usually due to the compressive abdominal mass effect [34] to the adjacent area leading to vomiting; jaundice, gut obstruction, hydronephrosis, muconium peritonitis and breathing difficulty but such findings are usually not so overt in adulthood.

Foetus in foetu is a result of abnormal twinning process where suppressed monozygotic twin develop partial or complex external morphology of a foetus [35]. Initially during intrauterine period their growth occurs parallel but then abruptly impedes due to vascular dominance of the host twin or an inherent defect in the parasitic twin [33] which may lead to spectrum of anomalous embryogenesis like conjoined symmetric twins, parasitic foetus, embryonic foetal inclusion and organised teratoma [25, 36]. Although triggering factors for compromising blood supply to parasitic twin are still not

clear but some theories focus on host fact factors or nutritional supply while according to others authors parasitic twin continue to grow till enough vascularization [34, 41].

It is a matter of debate that whether foetus in foetu is a distinct entity or a highly organised fetiform teratoma [36]. Since fetiform teratoma is a rare form of mature teratoma that includes one or more components resembling a malformed foetus. Both forms may contain or appear to contain complex organ system, even major body parts such as torso and limbs. Foetus in foetu differs from fetiform teratoma in having spine and bilateral symmetry [5]. The teratoma usually occurs in the lower abdomen, ovaries or sacrococcygeal region and have tendency to malignant transformation [7, 11, 38] while foetus in foetu is composed of a complex fibrous membrane that is equivalent to chorioamniotic complex which contains some fluid and foetus suspended by a chord or pedicle [21, 37]. According to Willis the presence of a vertebral column is an important diagnostic criterion of foetus in fetu [5]. Even identification of notochord in term of an advanced primitive streak which is the precursor of the vertebral column can be sufficient to differentiate this from other [32, 36]. Although it's total absence does not exclude diagnosis of foetus in foetu probably due to invisibility at imaging of underdeveloped and non-calcified spinal column [8, 33].

Several genetic and molecular diagnostic modalities have been used to confirm foetus in foetu as genetic marker for uni-parental isodisomy of chromosome 14 and 15 shows no genetic difference between host and fetiform mass in foetus in foetu [42]. Analysis of blood group as well as karyotype between the host and foetus like mass showed identical finding [39]. PCR and DNA finger printing techniques confirm identical genetic material shared by the host and the mass [40].

Role of serological examination of  $\alpha$ -fetoprotein and  $\beta$ -HCG in foetus in fetu are not so clear [7,8,43] and the host may have reported normal or elevated level of  $\alpha$ -fetoprotein but in newborns reference value of  $\alpha$ -fetoprotein have yet to be clearly defined [44]. In one case the raise level maternal of  $\alpha$ -fetoprotein has been reported [45].

Complete surgical excision along with covering membrane is treatment of choice as presence the remnant membrane which may contain immature cells and likely to have malignant potential [2, 7]. So proper preoperative consideration and precise planning of surgical access are important and careful dissection may help to avoid injury to surrounding structures as a case of bile duct injury has been reported [25]. If mass is asymptomatic and incidentally found during investigations even though it should be removed as cases of malignant transformation has been reported [7, 11, 38]. Apart from this there are possibilities of associated risk as mass effect, infection, haemorrhage and pleura-peritoneal inflammation due to



leak from cavity mass [25].

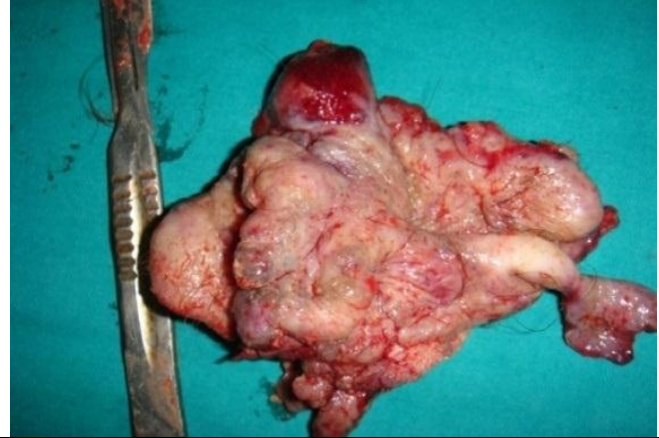
In conclusion, intra hepatic foetus in foetu is a rare entity and has not been reported especially in adult previously. Use of current imaging modality like ultrasonography, CT or MRI are valuable adjuncts for its

preoperative diagnosis and surgical planning [8,24] and finally surgical exploration and macroscopic and microscopic finding of excised specimen can establish the diagnosis.

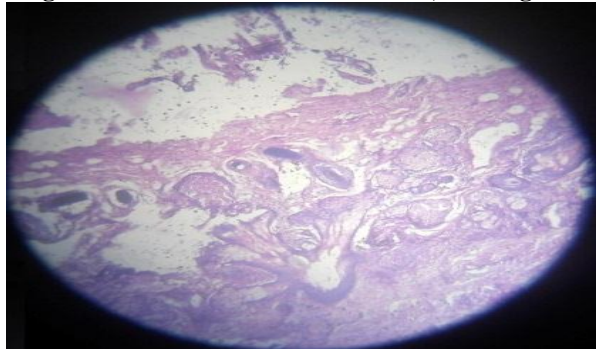
**Fig 1. CT scan showing a partially healed calcified thick walled cavitory mass of 8 × 6 cm size in the lower part of the right lobe of liver compressing the superior pole of right kidney**



**Fig 2. Excised mass showing strands of hair admixed with tissue like structure and body parts.**



**Fig 3. Histopathology showing hair admixed with fibrotic tissue, cartilage and bones of vertebral column**



## REFERENCES

1. Nastanski F, Downey EC. (2001). Fetus in fetu: a rare case of a neonatal mass. *Ultrasound Obstet Gynaecol*, 72-75.
2. Levis RH. (1960). Foetus in foetu and retroperitoneal teratoma. *Arch Dis Child*, 36, 220-6
3. George W. (2012). Case of a foetus found in the abdomen of a boy.
4. Nicholson GW. (1950). Studies on tumor formation. London, Butterworths.
5. Willis RA. (1935). The structure of teratoma. *J Pathol Bacteriol*, 40, 1-36
6. Grant R, Pearn JH. (1969). Foetus-in-fetu. *Medical J Aust*, 1, 1016-20.
7. Hopkin KL, Dickson PK, Ball TI, Ricketts RR, O'Shea PA. (1997). Abramovosky CR: Fetus in fetu with malignant recurrence. *J Pediatr Surg*, 32, 1997, 1476-1479.
8. Hoeffel CC, Nguyen KQ, Phan HT, et al. (2000). Fetus in fetu: A case Report and literature Review. *Pediatric*, 105, 1335-44.
9. Federici S, Ceccarelli PL, Ferrari M, Galli G, Zanetti G, Domino R. (1991). Fetus-in-fetu: report of three cases and review of the literature. *Pediatr Surg Int*, 6, 60-5.
10. Patankar T, Fatterpekar GM, Prasad S, Mukherji SK. (2000). Fetus in fetu: CT appearance-report of two cases. *Radiology*, 214, 735-737.
11. Thakral CL, Maji DC, Sajwani MJ. (1998). Fetus-in-fetu: a case report and review of the literature. *J Pediatr Surg*, 33, 1432-4.
12. Luzzatto C, Talenti E, Trednaghi A, Fabris S, Scapinello A, Gugliema M. (1994). Double fetus in fetu: diagnostic imaging. *Pediatr Radiol*, 24, 602-603.

13. Gangopadhyay AN, Srivastva A, Srivastva P, Gupta DK, Sharma SP, Kumar V. (2010). Twin fetus in fetu in a child: a case report and review of the literature. *J Med Care Rep*, 25, 4, 96.
14. Pourang H, Sarmadi S, Mireskandari SM, Soleimani M, Mollaeian M, Alizadeh H, Alehosein SM. (2009). Twine fetus in fetu with immature teratoma: a case report and review of the literature. *Arch Iran Med*, 12(5), 507-10.
15. Hung CF, Lam MS. (1993). Intracranial fetus in fetu: report of a case. *J Formos Med Assoc*, 92(10), 920-2.
16. Kakizoe T, Tahara M. (1972). Fetus in fetu located in the scrotal sac of a newborn infant: a case report. *J Uro*, 107, 506-508.
17. Sanal M, Kucukcelebi A, Abasiyanik F, Erdogan S, Kocabasoglu U. (1997). Fetus in fetu and cystic rectal duplication in a newborn. *Eur J Pediatr Surg*, 7, 120-121.
18. Braimoh KT, Abdulkadir AY, Balogun RO. (2008). Oroccervical foetus-in-foetu with prenatal sonographic diagnosis: a case report. *J Med Case Rep*, 4, 362.
19. Massad MG, Kong L, Benedetti E, et al. (2001). Dysphagia caused by a fetus in fetu in a 27 year old man. *Ann Thorac Surg*, 71, 1338-1341.
20. Aoki K, Matsumoto Y, Hamazaki M, et al. (2004). MRI reveals fetus in fetu in the mediastinum. *Pediatr Radiol*, 34, 2004, 1017-1019.
21. Chua, JHY; Chui CH, Sai Prasad. (2005). Fetus-in-fetu in the pelvis: Annals of the Academy of Medicine Singapore, 34(10), 646-9.
22. Goldstein I, Jakobi P, Goisman G, Itskovitz-Eldor J. (1996). Intracranial fetus in fetu. *Am J Obstet Gynecol*, 175, 1389-1390.
23. Afshar F, King TT, Berry CL. (1982). Intraventricular fetus in fetu. Case report. *J Neurosurg*, 56(6), 845-9.
24. Tsai CH, Lin JS, Tsai FJ. (1993). Intraventricular fetus in fetu: report of one case. *Acta Paediatr Sin*, 34, 143-150.
25. Jeyanti NN, Mohan A. (2014). Fetus-in-fetu: a pediatric rarity. *Journal of surgical case reports*, 22-24.
26. Kapoor V, Flom L, Fitz CR. (2004). Oropharyngeal fetus in fetu. *Pediatr Radiol*, 34(6), 488-91.
27. Chitra Y, Zorn B, Scart G, et al. (1990). Adrenal fetus infetu. *J Gynecol Obstet Biol Reprod*, 19, 1019-1022.
28. Elizabeth Borges, Jennifer E. Lim-Dunham, Aruna Vade. (2005). Fetus in fetu appearing as a prenatal neck mass. *J Ultrasound Med*, 24, 1313-1316.
29. Alpers V, Harrison MR. (1985). Fetus in fetu associated with an undescended testis. *Pediatr Pathol*, 4, 37-46.
30. Shin JH, Yoon CH, Cho KS, Lim SD, Kim KS, Pi SY, Auh YH. (1999). Fetus-in-Fetu in the scrotal sac of newborn infant: imaging, surgical and pathologicval findings. *Eur Radiol*, 9(5), 945-7.
31. Al-Baghdadi R. (1992). Fetus-in-Fetu in the liver: case report and rewiw of literature. *J Pediatr Surg*, 27, 1491-1493.
32. Magnus KG, Miller AJ, Sinclair-Smith CC, Rode H. (1999). Intrahepatic fetus-in-fetu: a case report and review of literature. *J Pediatr surg*, 34(2), 1861-1864.
33. Dagradi AD, Mangiante GL, Serio GF, Musajo FG, Mesentrina FV. (1992). Fetus-in-fetu removal in 47- year-old man. *Surgery*, 112, 598-602.
34. Knox JS, Webb AJ: The clinical features and treatment of fetus in fetu: two cases reports and review of literature. *J Pediatr Surg*, 10, 1975, 483-489.
35. Kaufman D, Du L, Velcek FT, Alfonso AE. (2007). Fetus in Fetu. *J Am Coll Surgeons*, 205, 378-9.
36. Potter EL. (1961). Pathology of fetus and the newborn. In Pathology of the fetus and the newborn, Edited by Potter EL, 183-187.
37. Saito K. (2007). Fetus in fetu: Parasite or Neoplasm. *Japan; Fetal Diagn*, 22(5), 383-8.
38. Eng HL, Chuang JH, Tze-Yu L, Chen WJ. (1989). Fetus in fetu: a case report and review of literature. *J Pediatr Surg*, 24, 296-299.
39. Grant P. & Penna JH. (1969). Fetus in fetu. *Med J Australia*, 1, 1010-9.
40. Kumar AN, Chandak GR, Rajshekhar A, Reddy NCK, Singh L. (1999). Fetus-in-fetu: a case report with molecular analysis. *J Pediatr Surg*, 34, 1999, 641-4.
41. Kim OH, Shinn KS. (1993). Postnatal growth fetus in fetu. *Pediatr Radiol*, 23, 411-2.
42. Brand A, Alves MC, Saraiva C, Loio P, Goulao J, Malta J, et al. (2004). Fetus in fetu-Diagnostic criteria and differential diagnosis- A case report and literature review. *J Ped Surg*, 39(4), 616-618.
43. De Lagausie P, de Napoli Cocci S, Stempfle N, Truong QD, Vuillard E, Perkarji L, et al. (1997). Highly differentiated teratoma and fetus in fetu: a single pathology? *J Pediatr Surg*, 32, 115-6.
44. Bader D, Riskin A, Vafsi O, Tamir A, Peskin B, Israel N, et al. (2004).  $\alpha$ -fetoprotein in the early neonatal period- a large study and review of literature. *Clin Chim Acta*, 349, 2004, 13-23.
45. Mohta A, Shrivastva UK, Sodhin P, Upreti L. (2003). Fetus-in-fetu. *Pediatr Surg Int*, 19, 499-500.

