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Neonatal adenomyomatosis of the gallbladder: An incidental finding at 12 hours of life

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Adenomyomatosis of the gallbladder, although relatively common in adults, is a rare entity in the pediatric age group. Controversy still exists as to whether or not to perform followup ultrasound, as there is a questionable increased risk of gallbladder carcinoma in adults. We present a case of neonatal adenomyomatosis that was diagnosed at 12 hours of life in a term newborn.

Case report

A 12-hour-old term newborn female was delivered at 38 weeks without any complications. The patient was suspected to have heterotaxy syndrome with balanced complete AV canal on fetal echocardiography. Additional findings included interrupted IVC with azygous continuation to the right SVC and bilateral vena cava; these were confirmed by neonatal echocardiography. A routine abdominal ultrasound demonstrated a speckled appearance of the gallbladder with numerous echogenic foci that caused the classic "ring down artifact" (Fig. 1). These findings were consistent with adenomyomatosis of the gallbladder, which is a rare entity in the realm of pediatric radiology. The only positive maternal history was genital herpes, which was treated with acyclovir from 36 weeks. Ultimately, no definite etiology was established, and this was felt to be idiopathic in origin. Followup ultrasound, approximately 8 months later, demonstrated apparent resolution of the adenomyomatosis.

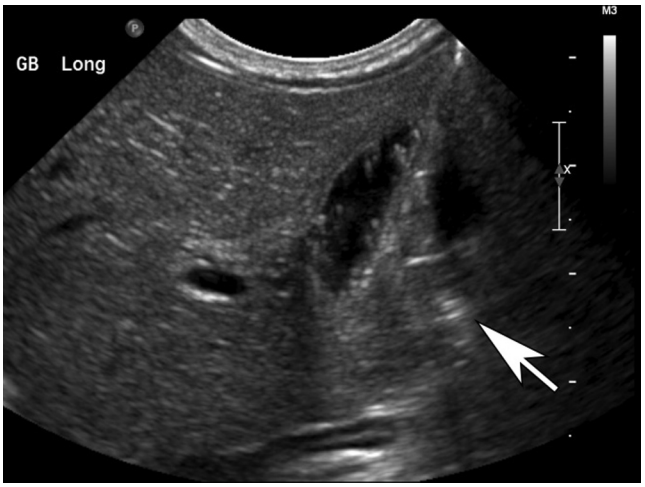

Figure 1. Ultrasound image shows gallbladder with comet-tail reveration artifact (arrow), pathognomic for adenomyomatosis of the gallbladder.

Discussion

Adenomyomatosis of the gallbladder is a relatively common and benign condition in adults. The incidence varies between 0.6% and 33.3% of all gallbladder pathology (1). It is thought to constitute 1-9% of all cholecystectomy specimens (1). The incidence increases with age; the peak age is the 5th decade (1). It is extremely rare in the pediatric population. Only three cases have been reported so far in the literature (2-6).

The majority of the affected patients are asymptomatic. The diagnosis is associated with gallstones in 25.75% and cholesterolosis in 33% of the cases (1). Occasionally, the condition gives rise to a symptom complex similar to that of cholelithiasis and chronic cholecystitis (6).

Adenomyomatosis is described as hyperplastic cholecystosis of the gallbladder wall, characterized by proliferation

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of the mucosa of the wall. This forms invaginations and diverticula penetrating a thickened muscular layer; these are termed Rokitansky-Aschoff sinuses. Precipitated cholesterol crystals from bile get trapped in these sinuses.

Radiographically, three morphologic types are identified: generalized (diffuse), segmental (annular), and fundal (localized), which is also called adenomyoma. The typical sonographic appearance is mural thickening associated with the more specific V-shaped comet-tail reverberation artifact (Fig. 1). This is secondary to the acoustic signature of cholesterol crystals within the lumina of Rokitansky-Aschoff sinuses (7). CT demonstrates nonspecific wall thickening and enhancement. The rosary sign has been described, which is formed by enhancing epithelium within intramural diverticula surrounded by the relatively unenhanced hypertrophied gallbladder muscularis (8). On MRI, the pearl-necklace sign is so called because of the characteristic curvilinear arrangement of multiple, rounded, hyperintense, intraluminal cavities on T2 and MRCP (9).

The differential diagnosis includes gallbladder carcinoma, phyrgian cap, cholesterol polyp, and cholelithiasis. The most problematic situation arises when carcinoma (in segmental and focal cases) cannot be excluded.

Usually, no treatment is recommended. Followup ultrasound on this patient demonstrated apparent resolution, which has not been reported in the literature to date. Cholecystectomy is indicated in patients who are symptomatic with right-upper-quadrant pain, and when appearances make it difficult to distinguish the condition from malignancy. The majority of the pediatric patients reported in the literature had cholecystectomy (2-5). However, there is a reported increased risk of malignancy (only in adults), and no substantial data is available in pediatric population to show that it is true in this subset of the population.

References

1. Weerakkody Y, Yoshiharu R et al. Adenomyomatosis of the gallbladder. Radiopaedia.org
2. Akçam M, Buyukyavuz I, Ciriş M, Eriş N. Adenomyomatosis of the gallbladder resembling honeycomb in a child. *Eur J Pediatr*. 2008 Sep; 167(9):1079-81. Epub 2007 Oct 19. [PubMed]
3. Zani A, Pacilli M, Conforti A, Casati A, Bosco S, Cozzi DA. Adenomyomatosis of the gallbladder in childhood: report of a case and review of the literature. *Pediatr Dev Pathol*. 2005 Sep-Oct; 8(5):577-80. Epub 2005 Oct 5. [PubMed]
4. Alberti D, Callea F, Camoni G, Falchetti D, Rigamonti W, Caccia G. Adenomyomatosis of the gallbladder in childhood. *J Pediat Surg*. 1998 Sep; 33(9):1411-2. [PubMed]
5. M. P. Bethune, M. Pahuja. Adenomyomatosis causing an echogenic fetal gallbladder – a new association, Article first published online: 30 APR 2002, Wiley online library.
6. JC Balaguera et al. Diffuse Adenomyomatosis of the gallbladder: An infrequent disease with difficult preoperative diagnosis. *The Internet Journal of Surgery* ISSN: 1528-8242.
7. Boscak AR, Al-hawary M, Ramsburgh SR. Best cases from the AFIP: Adenomyomatosis of the gallbladder. *Radiographics*. 26 (3): 941-6. [PubMed]
8. Ching BH, Yeh BM, Westphalen AC et-al. CT differentiation of adenomyomatosis and gallbladder cancer. *AJR Am J Roentgenol.* 2007; 189 (1): 62-6. [PubMed]
9. Haradome H, Ichikawa T, Sou H et-al. The pearl necklace sign: an imaging sign of adenomyomatosis of the gallbladder at MR cholangiopancreatography. *Radiology*. 2003; 227 (1): 80-8. [PubMed]