

FETAL BILATERAL ADRENAL HEMORRHAGE (case report)

J. S. Randhawa¹, *R. Nagamahendran², R. Shankaran³

1 – INDIAN NAVAL HOSPITAL SHIP KALYANI, VISAKHAPATNAM, INDIA

2 – INDIAN NAVAL HOSPITAL SHIP SANDHANI, MUMBAI, INDIA

3 – INSTITUTE OF NAVAL MEDICINE, MUMBAI, INDIA

Background. Fetal supra renal mass revealed incidentally by routine antenatal ultrasound is a great challenge for diagnosis and management by a surgeon. This is a matter of parental anxiety and diagnostic dilemma to a physician. Indeed, such masses turn out to be complicated by an intra-tumor hemorrhage in neuroblastoma or antenatally diagnosed adrenal hemorrhage. The first one needs intensive management and the latter needs watchful observation.

Objective. A case of bilateral fetal adrenal mass revealed by routine fetal ultrasound examination at 28th week of gestation which turned out to be adrenal hemorrhage is presented. This is aimed to make awareness to ensure that clinicians always keep benign etiologies first and thoroughly investigate in case of incidentally detected fetal adrenal mass.

Methods. The study is a single case report of incidentally revealed supra renal mass. This case report encompasses differentiating features between the two and investigations that aid the surgeon to avoid unnecessary intervention in a benign hemorrhage.

Results. The baby was kept on follow up with serial ultrasound scans in the postnatal period and by the second scan in a month, the hemorrhage had resolved completely.

Conclusion. In cases of benign looking masses like adrenal hemorrhage or spontaneously resolving neuroblastoma, appropriate antenatal assessment and close monitoring with serial ultrasound scans can avoid surgery.

KEYWORDS: bilateral fetal adrenal hemorrhage; neuroblastoma; adrenal mass.

Introduction

The supra renal glands were discovered in 1522 by Eustachius and described by him in 1563. The adrenal glands are responsible for essential physiologic functions including stress response, electrolyte balance, metabolism, immune function, blood pressure regulation and sexual development. Supra renal masses have been a longstanding source of fascination for both physicians and surgeons especially in neonatal period. Adrenal glands are relatively larger in size with high vascularity during fetal and neonatal period. This predisposes to its hemorrhage in early life. Neonatal adrenal hemorrhage is rare and is seen in 0.2-0.5% of cases [1]. With the recent advancement in imaging modalities, diagnosis and management of supra renal masses has become more efficient. Assessment of hemodynamic stability, adrenal insufficiency and exclusion of a hormonal active adrenal tumor proves to be a topical issue in management of such suprarenal

masses [2]. Association of adrenal hemorrhage with thrombosis of major blood vessels is also reported in the literature [3]. Thus, a rare case of antenatally diagnosed bilateral adrenal mass which turned out to be benign adrenal hemorrhage is presented. This case report envisages the need for watchful observation and importance of short interval follow up imaging in management of benign disorders of adrenal gland before any surgical intervention.

Case report

Rare presentation of neonatal adrenal masses is a diagnostic dilemma in differentiating benign adrenal fetal hemorrhage from neuroblastoma. This case report encompasses differentiating features between the two and investigations that aid the surgeon to avoid unnecessary intervention in a benign hemorrhage. Following is the case report of a 27 years old Gravida 3, Para 2, Live 2 woman, who was referred to our pediatric surgery center at 28 week of gestation for evaluation of bilateral supra renal cystic mass. Her previous medical history was unremarkable. On evaluation with sonography, bilateral cystic mass was located

*Corresponding author: Dr. R. Nagamahendran, Assistant Professor of the Department of General Surgery, Indian Naval Hospital Ship Sandhani, Mumbai, 400704, India.
E-mail: nagaa.mahendran@gmail.com

superior to both kidneys measuring 15×10 mm on the right side and 14×9 mm on the left side.

Fetus was evaluated further by Doppler examination of umbilical cord, biophysical profile and fetal biometry and were in norm. Evaluation with antenatal MRI scan on 28 weeks of gestation was suggestive of bilateral fetal adrenal hemorrhage. The antenatal mother was kept on serial ultrasound follow up, scans at 32 weeks and 35 weeks revealed multicystic lesions of both adrenals. The right adrenal measured 21×12 mm and the left one – 20×13 mm without any abnormal vascularity. Mother underwent elective lower segment caesarean section at 38 weeks of gestation. The surgery and postoperative period was uneventful. A 2.815 kg, healthy female child was delivered with normal peripartum period.

Discussion

Ultrasonography of Abdomen on the 2nd day of life revealed mild resolution of bilateral suprarenal areas to 15×15 mm in the right and 13×14 mm in the left with hypo echoic foci within. Further biochemical evaluation of serum aldosterone, renin, cortisol and plasma ACTH levels were in norm. The baby was kept on follow up with serial ultrasound scans and by the second scan in a month, the hemorrhage had resolved completely.

Spencer, in 1892 first described adrenal hemorrhage in stillborn infants. Based on extensive autopsy reports, the incidence of adrenal hemorrhage is estimated to be about 1.7 per thousand births and the incidence of antenatal detection is not established due to its rarity [2]. Bilateral adrenal hemorrhage is found in 5-15% of cases with right side preponderance with right and left side ratio of 3:1 [4].

The increased use of perinatal ultrasound has led to early detection of an increasing number of neonatal suprarenal masses [5]. In

ultrasound, the adrenal glands typical looks like wishbone appearance in the form of cap or an inverted V over the kidneys [6]. Adrenal hemorrhage is rarely associated with adrenal insufficiency. Even in cases with severe bilateral adrenal hemorrhage, it is hardly ever encountered as less than 10% availability of functional glandular component that is enough to prevent adrenal insufficiency [7].

Adding to the existing knowledge of management of supra renal masses, which are detected antenatally, it is prudent from our study that serial radiological monitoring is crucial. Masses with radiological reduction in size should be given full consideration for conservative management [8]. Rare occurrence of fetal adrenal hemorrhage should be kept in mind in those cases where surgery can be avoided.

Conclusion

Differentiation between fetal adrenal hemorrhage and congenital neuroblastoma is difficult and urgent. In cases of radiologically benign looking supra renal masses, appropriate antenatal assessment and close monitoring with serial ultrasound scans can avoid unnecessary surgical intervention as they resolve spontaneously.

Limitations

Single case report in a tertiary care center.

Conflict of Interests

Authors declare no conflict of interest.

Acknowledgements

Acknowledgements if any.

Author's Contributions

Dr. Jaskiran Singh Randhawa – conceptualization, methodology, formal analysis, writing – reviewing and editing; *Dr. R. Nagamahendran* – investigation, writing – original draft, writing – reviewing and editing; *Dr. R. Shankaran* – investigation, formal analysis, data curation.

ДВОСТОРОННЯ КРОВОТЕЧА З НАДНИРКОВИХ ЗАЛОЗ У ПЛОДА (клінічний випадок)

J. S. Randhawa¹, R. Nagamahendran², R. Shankaran³

1 – INDIAN NAVAL HOSPITAL SHIP KALYANI, VISAKHAPATNAM, INDIA

2 – INDIAN NAVAL HOSPITAL SHIP SANDHANI, MUMBAI, INDIA

3 – INSTITUTE OF NAVAL MEDICINE, MUMBAI, INDIA

Вступ. Утворення в наднирниках плода, випадково виявлені під час звичайного допологового ультразвукового дослідження, викликають занепокоєння батьків і створюють діагностичну проблему для лікуючого лікаря. Такі утворення можуть виявитися ускладненим внутрішньопухлинним крово-

виливом у нейробластому або антенатально діагностованим крововиливом у наднирковій залозі. Перший потребує інтенсивного лікування, а другий – уважного спостереження.

Мета. Представлено випадок двостороннього утворення в надниркових залозах плода, виявленого під час планового ультразвукового дослідження плода на 28-му тижні вагітності, які виявилися крововиливом у наднирковій залозі. Випадок представлено з метою загострення уваги та настороженості клініцистів щодо доброякісної патології та проведення ретельного обстеження у випадку випадкового виявлення новоутворень надниркових залоз плода.

Методи. Представлено випадок випадково виявлених утворень надниркових залоз. Описано відмінності між ними та дослідження, які допомагають хірургу уникнути непотрібного втручання при доброякісному крововиливі.

Результати. У післяпологовому періоді дитину спостерігали за допомогою серійних ультразвукових досліджень. До другого УЗД через місяць крововилив повністю розсмоктався.

Висновки. У випадках доброякісних новоутворень, таких як крововилив у наднирковій залозі або нейробластома, що спонтанно розсмоктується, відповідна антенатальна оцінка та ретельний моніторинг за допомогою серійних ультразвукових досліджень можуть допомогти уникнути операції.

КЛЮЧОВІ СЛОВА: двостороння фетальна надниркова кровотеча; нейробластома; утворення в наднирниках.

Information about the authors

Dr. Jaskiran Singh Randhawa – Associate Professor and Head, Department of Surgery, Indian Naval Hospital Ship Kalyani, Vishakapatnam, Andhra Pradesh, India

<https://orcid.org/0000-0002-0081-7382>, e-mail: moshesingh2000@yahoo.co.in

Dr. R. Nagamahendran – Assistant Professor, Department of General Surgery, Indian Naval Hospital Ship Sandhani, Mumbai, India

<https://orcid.org/0000-0002-9854-7236>, e-mail: nagaa.mahendran@gmail.com

Dr. R. Shankaran – Dean and Professor, Department of General Surgery, Institute of Naval Medicine, Mumbai, India

<https://orcid.org/0000-0002-0105-2270>, e-mail: drshankaran@gmail.com

References

1. Izbizky G, Elias D, Gallo A, Farias P, Sod R. Prenatal diagnosis of fetal adrenal carcinoma bilateral. *Ultrasound Obstet Gynecol.* 2005; 26:669-71. <https://doi.org/10.1002/uog.2623>
2. Alabsi SY, Layland T. Adrenal Hemorrhage in Neonates: Unusual Presentation. *Neonatal Netw.* 2015; 34(4):220-6. <https://doi.org/10.1891/0730-0832.34.4.220>
3. Peruri G, Suthar R, V, Bhatia A. Thrombosis of Bilateral Renal Veins, Inferior Vena Cava, and Superior Sagittal Sinus with Adrenal Hemorrhage in a Neonate. *J Postgrad Med Edu Res* 2019; 53(2):89-90. <https://doi.org/10.5005/jp-journals-10028-1320>
4. Shin SI, Yoo JG, Park IY, Cheon JY. Prenatal diagnosis – fetal adrenal hemorrhage and endocrinologic evaluation. *Obstet Gynecol Sci.* 2016; 59(3):238-40. <https://doi.org/10.5468/ogs.2016.59.3.238>
5. Maki E, Oh K, Rogers S, Sohaey R. Imaging and differential diagnosis of foetal suprarenal masses. *J Ultrasound Med.* 2014;33:895-904 <https://doi.org/10.7863/ultra.33.5.895>
6. Spencer HR. On visceral hemorrhages in stillborn children. *Trans Obstet Soc London.* 1892; 33:203.
7. GLENN JF. Neonatal adrenal hemorrhage. *J Urol.* 1962 May;87:639-42 [https://doi.org/10.1016/S0022-5347\(17\)65019-5](https://doi.org/10.1016/S0022-5347(17)65019-5)
8. Comline RS, Silver M. Catecholamine secretion by the adrenal medulla of the foetal and new-born foal. *J Physiol.* 1971 Aug; 216(3):659-82 <https://doi.org/10.1113/jphysiol.1971.sp009546>

Received 5 September 2022; revised 20 October 2022; accepted 1 November 2022.

This is open access article distributed under the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited.