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Address for Correspondence:Editorial Room Andalas Obstetrics and Gynecology Journal, 3rd floor of KSM of Obstetrics and Gynecology, RSUP DR. M. Djamil Padang, Jl. Perintis Kemerdekaan Padang, Sumatera Barat 25127**Website:**<http://jurnalobgin.fk.unand.ac.id/index.php/JOE>**CASE REPORT****Hygroma Colli**Vaulinne Basyir¹, Try Genta Utama²,

Affiliation author: 1. Sub Division of Maternal Fetal Medicine, Obstetrics and Gynecology Department, Faculty of Medicine, Andalas University, Dr. M. Djamil Central General Hospital Padang, West Sumatera, Indonesia; 2. Obstetrics and Gynecology Department, Faculty of Medicine, Andalas University, Dr. M. Djamil Central General Hospital Padang, West Sumatera, Indonesia

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Abstract

Background : Hygroma coli is a malformation of the lymphatic system in the form of a membrane cyst filled with fluid, limited by the epithelium that is located in the anterolateral or occipito-cervical region. The prenatal diagnosis of cystic hygroma coli by ultrasound is based on an apparently bilateral, mostly symmetrical, and sometimes unilateral cystic structure located in the occipitocervical region. Large hygroma coli can cause pressure on the respiratory tract and digestion, so it requires management as soon as possible. The main treatment modality is surgical excision to remove the cystic lesion. The prognosis of a hygroma coli cyst depends on its size and the action taken because it is rare for cases to experience spontaneous regression.

Destination : Reported a case of hygroma colli

Methods : Case Report

Case Report : Case 24 years old women with preterm G1P0A0L0 26-27 weeks + Hygroma colli + IUFD + Suspected COVID-19. On ultrasound examination, it was found that BPD = 4,71; AC = 15,91; FL = 2,89; EFW = 330 gr; FHR = (-); Cyst = 5,06 x 3,26. The presence of head presentation, IUFD, hygroma colli was found. The patient was planned for labor induction and the progress of labor was followed. Patient provided informed consent that baby was death during pregnancy and need to be labored. The baby was born, weight 300 gr, body length 14 cm and A/S 0/0. Postmortem physical examination revealed findings of hygroma colli infants such membrane cyst filled with fluid that located in the occipito-cervical region.

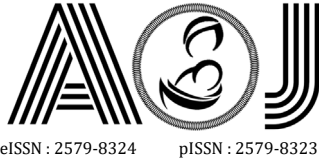
Conclusion: Hygroma colli is a malformation of the lymphatic system and the prognosis or complications depends on the size of cyst. Careful prenatal examination is required in diagnosis and termination of pregnancy

Keywords: Hygroma Colli, prenatal diagnosis

INTRODUCTION

Hygroma or also called cystic hygroma comes from the word hygro moist, and -oma = tumor, so in Greek it means a cyst filled with clear fluid. Hygroma is a congenital abnormality of the lymphatic system. The hygroma was first described by Wernher in 1843 as a lymphatic cystic lesion that can affect various anatomical areas of the human body. However, most of them affect the head and neck area (75%), with a left predilection.¹⁻⁵

Hygroma coli is a malformation of the lymphatic system in the form of a membrane cyst filled with fluid, limited by the epithelium that is located in the anterolateral or occipito-cervical region. The hygroma can be small, simple and transient or large, perceptual and



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persistent. This cystic hygroma is caused by obstruction of the jugular lymph flow which fails to develop at 40 days' gestation. If the connection between the jugular vein and jugular lymph is not formed, a progressive peripheral lymphedema will occur to hydrophy the fetal and cause fetal death.⁵⁻⁹

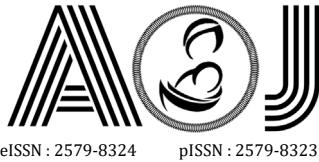
The prenatal diagnosis of cystic hygroma coli by ultrasound is based on an apparently bilateral, mostly symmetrical, and sometimes unilateral cystic structure located in the occipitocervical region. Coli cystic hygroma is frequently associated with chromosomal abnormalities such as Turner syndrome (45, X0), and Trisomies 21, 18, and 13. It is also associated with cardiac anomalies and nonimmune fetal hydrops. Large hygroma coli can cause pressure on the respiratory tract and digestion, so it requires management as soon as possible. The main treatment modality is surgical excision to remove the cystic lesion. The prognosis of a hygroma coli cyst depends on its size and the action taken because it is rare for cases to experience spontaneous regression.¹⁰⁻¹⁶

CASE REPORT

Reported a 24 years old female patient diagnosed with preterm G1P0A0L0 26-27 weeks + Hygroma colli + IUFD + Suspected COVID-19. From history, it was found that the fetal movements have not been felt since 3 days, bleeding from vagina was absent, amenorrhea since + 6 months ago. First date of last menstrual on April 1st 2020 with estimation of delivery on Januari 20th 2021. During ANC examination, patient control with midwife 2 times at 2 month and 4 month pregnancy, never control to an obstetrics. Menstrual history menarche at the age of 13 years, regular menstrual cycles 1 time per month, 5-6 days of duration, 3-4 times changing pads/day, menstrual pain

From the physical examination, the general condition was moderate, composmentis cooperative awareness; TD 120/70 mmHg; Pulse 83 x/min; Rr 20 x/min. From the obstetric examination it was seen that the abdomen was enlarging accordance to preterm pregnancy, striae gravidarum and hyperpigmented linea mediana were present. The fundal height was 20 cm. Pelvic examination obtained wide pelvis.

Labor results show within normal limits, Hb = 11.6 gr/dl; leukocytes = 10,430/mm³ Ht 35%; Platelets 451.000/mm³. On ultrasound examination, it was found that BPD =4,71; AC = 15,91; FL = 2,89; EFW = 330 gr; FHR = (-); Cyst = 5,06 x 3,26. The presence of head presentation, IUFD, hygroma colli was found. The patient was planned for labor induction and the progress of labor was followed. Patient provided inform consent that baby was death during pregnancy and need to be labored. The baby was born, weight 300 gr, body length 14 cm and A/S 0/0. Postmortem physical examination revealed findings of hygroma colli infants such membrane cyst filled with fluid that located in the occipito-cervical region.



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A 24 year old woman came to Dr. Fetomaternal clinic. M. Djamil Padang. After taking anamnesis, physical examination and supporting examinations, the patient was diagnosed with preterm G1POAOHO gravid 26-27 weeks + hygroma coli + IUFD + suspected COVID19.

The patient came to the polyclinic with the complaint that the child's movement was not felt since 3 days ago. After an ultrasound examination, it turned out that the fetal heartbeat was no longer found, which indicated that the fetus had died intrauterine. Gestational age is based on LMP and fetal size from ultrasound examination.

During the pre-natal ultrasound examination, a cervical cyst was also found with a size of 5.06 x 3.26 cm. This cyst was previously unknown because of the patient never went to a specialist doctor and never had an ultrasound examination before. Based on the characteristics of the cysts found on ultrasound, it is suspected that these cysts are hygroma coli. In 80% of cases, the location of the hygroma is found in the cervico-facial region. The prenatal ultrasound image of hygroma coli is a multi-septal and thin-walled cystic mass, according to the description in this patient.

Hygroma can occur as a single finding or it can be found in conjunction with other defects. The causes also vary involving environmental factors such as maternal viral infections such as parvovirus or maternal substance abuse such as alcohol consumption during pregnancy. However, 60-70% of prenatal diagnoses of hygroma are associated with chromosomal anomalies such as Turner syndrome, Down syndrome or Knifelter syndrome. The hygroma on a single finding can be inherited as an autosomal recessive disorder from the silent carrier parent.¹⁷⁻²⁰

The pathophysiology of hygroma coli is due to a defect during the development of the lymphatic system. Lymph channels are formed at the sixth week of gestation. From this channel, a sac is formed which will provide drainage to the venous system. Failure to drain into the venous system will result in dilatation of the lymph channels, and if they are large, a hygroma will result. In the embryo, the lymphatic system drains into the jugular lymphatic sac. The relationship between the primitive structures of the lymphatic system and the jugular vein is formed at 40 days of gestation. Failure to form this structural relationship causes lymph flow stasis and the jugular lymphatic sac will widen, forming a cyst in the neck area. If the drainage system to the venous system is not established at this time, progressive peripheral lymphoedem will develop and can lead to intrauterine death.²⁰⁻²³



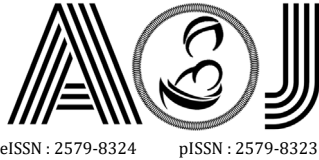
Figure 1. Ultrasonography of a fetus that show Higroma colli

The treatment consisted of vaginal termination of pregnancy in this patient because the fetus had died intrauterine and on pelvic examination found a wide pelvis allowing for vaginal delivery. In some cases of hygroma coli where the development of the fetus occurs during the 3rd trimester, termination of pregnancy is necessary at a health service that has complete facilities to be aware of neonatal complications. If the hygroma is large, terminate abdominally and cooperate with a neonatologist.



Figure 2. Postmortem examination: hygroma colli found in posterior neck of the fetus

If a case of hygroma coli is found while the fetus is still alive, further evaluation of the mother and fetus is necessary. Diagnosis that can be done in prenatal hygroma coli includes complete ultrasound to see other types of anomalies accompanying hygromas, complete



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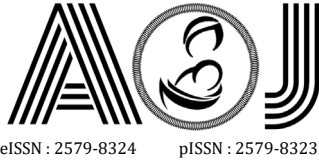
family history to determine indications of hereditary syndromes, amniosynthesis to see chromosomal abnormalities or specific genetic syndromes, assessment of viruses in the amniotic fluid if indicated the occurrence of hydrops and periodic ultrasound evaluation is urgently needed to see the resolution of the cyst and / or the development of other anomalies.

CONCLUSION

Hygroma coli is a malformation of the lymphatic system in the form of a membrane cyst filled with fluid. The prenatal diagnosis of cystic hygroma coli by ultrasound is based on an apparently bilateral, mostly symmetrical, and sometimes unilateral cystic structure located in the occipitocervical region. Coli cystic hygroma is frequently associated with chromosomal abnormalities such as Turner syndrome (45, X0), and Trisomies 21, 18, and 13. It is also associated with cardiac anomalies and nonimmune fetal hydrops. This case reported is a case of Hygroma colli based on prenatal ultrasound findings and post natal physical examination.

REFERENCES

1. Sjahmuhidajat, R. et al. 2010. Buku Ajar Ilmu Bedah-de Jong Edisi 3. Jakarta : EGC. Hal 461 – 465.
2. Acevedo L. Jason. 2011. Cystic Hygroma. Diunduh dari <http://www.emedicine.medscape.com/article/994055-overview#a0101> pada tanggal 21 Oktober 2020.
3. Kamble V., Bhatia T., Patil Shaifali. 2014. Cystic hygroma with hydrops fetalis : a rare case report. International Journal of Reproduction, Contraception, Obstetric and Gynecology. September : 3 (3): 847 – 850. Diunduh dari www.ijrcog.org pada tanggal 21 Oktober 2020.
4. Bilal Mirza, et al .2011 .Cystic Higroma. Department of Pediatric Surgery, The Children's Hospital and The Institute of Child Health, Lahore, Pakistan. Diunduh dari www.jcasonline.com pada tanggal 21 Oktober 2020.
5. Sabih, Durre. 2011. Cystic Hygroma Imaging. Diunduh dari www.medicine.medscape.com/article/402757-overview pada tanggal 21 Oktober 2020.
6. Turkington, et all. Neck Masses in Children. In British Journal Radiologi (2005) 78, 75-88. British Institute of Radiology. Diunduh dari www.bjr.birjournals.org/cgi/content/full/78/925/75 pada tanggal 21 Oktober 2020.
7. Nyberg, A., David, M.D., Mc Gahan P John, M.D., Pretorius H. Dolores, M.D, Pilu Gianluigi, M.D. 2003. Diagnostic Imaging of Fetal Anomalies. Lippincott williams & wilkins. 530 Walnut street. Philadelphia,PA 19106. USA.



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8. Varma, Thangam R. Cystic hygroma, colli. London: St. George's Hospital & Medical School. Diunduh dari www.sonoworld.com/fetus/page.aspx?id=202 pada tanggal 21 Oktober 2020.
9. Trager, Jochen, Seidensticker, Peter. 2008. Head and Neck in Paediatric Imaging Text Book. Chapter 3:39-40.
10. Wiley, John. 2003. Prenatal Diagnosis of a Huge Cystic Hygroma Colli. Journal Ultrasound Obstet Gynecol; 22: 323–324. Published online in Wiley Inter Science. Diunduh dari www.onlinelibrary.wiley.com/doi/10.1002/uog.219/pdf pada tanggal 21 Oktober 2020.
11. Sandhyarani, Ningthoujam. Anatomy of neck. Diunduh dari www.buzzle.com/articles/anatomy-of-neck.html pada tanggal 21 Oktober 2020.
12. Pameijer, Frank et al. 2009. Neck spaces - Infrahyoid Neck ;Normal Anatomy and Pathology. Radiology Department of the University Medical Centre of Utrecht, the Rijnstate Hospital in Arnhem and the Rijnland hospital in Leiderdorp, the Netherlands. Diunduh dari www.radiologyassistant.nl/en/49c603213caff pada tanggal 21 Oktober 2020.
13. Ellis, Harrold. 2006. The Vein of The Head and Neck. In Clinical Anatomy Text Book. UK: Blackwell publishing. Part 5::304.
14. Departemen of Human Genetics of Medicals Genetics. Cystic Higroma. Emory University of Human Genetics. Diunduh dari www.genetics.emory.edu/.../Emory_Human_Genetics_Cystic_Hy... - Amerika Serikat pada tanggal 21 Oktober 2020./
15. Wilson, JW. 1995. Neonatal Respiratory Obstruction due to Hygroma Colli Cysticum. Hospitals Group, Northern Ireland, City and County Hospital, Londonderry.
16. Domansky, Mark, et al. 2007. Pediatric Neck Masses. Diunduh dari www.utmb.edu/otoref/grnds/pedi-neck-mass.../pedi-neck-mass-071021.pdf pada tanggal 21 Oktober 2020
17. Estroff, JA. 2001. Nuchal translucency in Turner syndrome. In: Cohen HL, Sivit CJ, eds. Fetal & Pediatric Ultrasound. Columbus, OH: McGraw-Hill; 36-8.
18. Amin, Umar et al. 2007. Cystic Hygroma an Unusual Cause of Induced Abortion in Journal: J Ayub Med Coll Abbottabad 2007; 19(1) : 61. Diunduh dari www.docpdf.info/articles/hygroma+report+a+case.html pada tanggal 21 Oktober 2020.
19. Graesslin, et al. 2007. Characteristics and Outcome of Fetal Cystic Hygroma Diagnosed In the First Trimester. Acta obstet Gynecol Scand. 86(12):1442-6.
20. Chervenak,FA, et al. 1983. Fetal Cystic Higroma. Cause ang Natural History. N ; J Med, Oct 6; 309(14):822-5.



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21. Rasidaki M, Sifakis S, Vardaki E, et all. Prenatal diagnosis of a fetal chest wall cystic lymphangioma using ultrasonography and MRI: a case report with literature review. *Fetal Diagn Ther.* Nov-Dec 2005; 20 (6):504-7.
22. Cohen HL. Ascites and pleural effusion in hydrops. In: Cohen HL, Sivit CJ, eds. *Fetal and Pediatric Ultrasound.* New York, NY: McGraw-Hill;2001:79-82.
23. Mota R, Ramalho C, Monteiro J, et all. Evolving indication for the exit procedure: usefulness of combining ultrasound and fetal MRI. *Fetal Diagn Ther.* 2007; 22(2):107-11.