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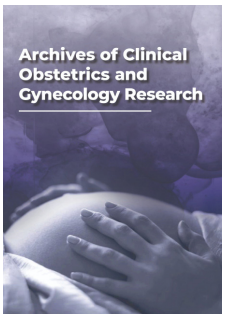
Case Report




# Late and Fortuitous Diagnosis of Two Cases of Uterine Malformations in Senegal and Review of The Literature

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## Abstract

*The frequency of uterine malformations is difficult to assess. Their detection requires specific explorations (hysterosalpingography, MRI, hysteroscopy and laparoscopy).*
*The bicornuate uterus is the best known of the uterine malformations and represents about half of the uterine malformations. They are also characterized by latency and clinical polymorphism. However, it is necessary to think about it in front of any accident of the pregnancy and any infertility of the couple. We report two cases of uterine malformations (Pseudo-unicorn uterus) discovered late and fortuitously intraoperatively.*

## Introduction

The frequency of uterine malformations is estimated at 0.5-4% in the general population and 3-4% in the female population. Spontaneous fertility may be impaired depending on the type of uterine abnormalities. All these anomalies can have repercussions on the evolution of the design. Their diagnosis is difficult because of their polymorphism and their clinical latency. However, they should be considered in all patients with a history of repeated miscarriages, late miscarriages and premature births; in adolescents who consult for primary amenorrhea, dysmenorrhea, dyspareunia and in patients followed in reproductive medicine..

## Comment

### Observation 1

We report an observation of a 29-year-old patient, sixth step, third pare, having in her history 3 vaginal deliveries with 3 healthy living children and 3 abortions who was evacuated from the maternity ward of the Darou Marnane health center ( Touba) for excruciating abdominopelvic pain on a 02 month delay in menstruation.

On admission, the general condition was preserved with slightly colored and anicteric conjunctival mucous membranes, blood pressure at 100/80 mmHg, temperature at 37°C and weight at 57 kg.

The gynecological examination found normal breasts, a supple abdomen, the vaginal examination found a median cervix, short, softened, dehiscent with finger streaked with blood.

Pregnancy test done, came back negative.

An ultrasound performed, was in favor of a right ovarian cyst of organic appearance. (Figure 1).

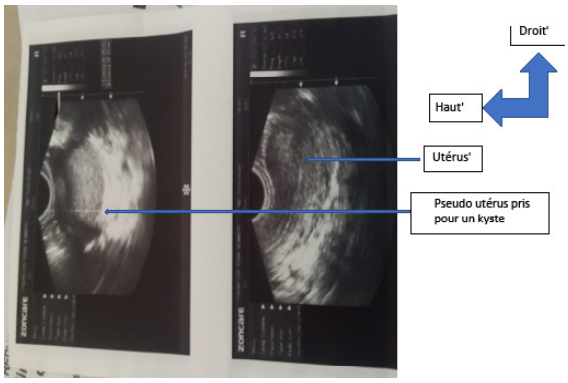


***Figure 1:*** *Ultrasound Images Showing an Empty Uterus and Pseudosac Image*

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Thus an indication for laparotomy was given for suspicion of adnexal torsion.

During the laparotomy, the exploration had found a right non-communicating canaliculated pseudounicorn uterus with a coiled right fallopian tube adhering to the ipsilateral ovary. (Figure 2)

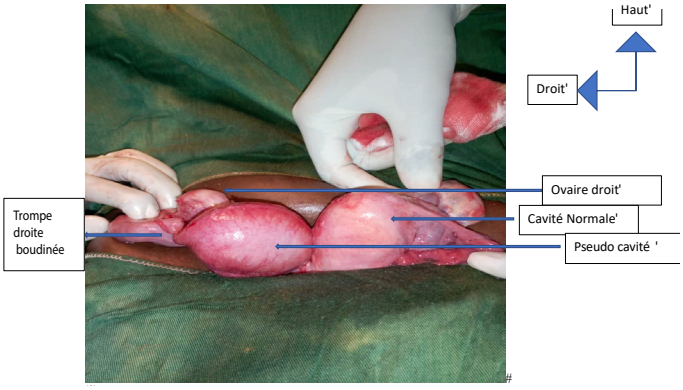


***Figure 2:*** *XXXXX*

We proceeded to adhesiolysis perforating the fallopian tube with chocolate liquid, a resection of the pseudo uterus preserving the homolateral ovary. The macroscopic examination of the operating specimen with incision of this one, had found that the latter had a cavity which contained a chocolate liquid corresponding to the accumulation of menses over the years after obliteration of the fallopian tube and the absence of other escape routes. Indeed, there was no communication allowing the passage of menses into the real uterus. Thus, the periods accumulated in the pseudo uterus causing pain. (Figures 3 and 4).

***Figure 3:*** *XXXXX*

***Figure 2:*** *XXXXX*

The postoperative course was simple and the patient was discharged 3 days after admission.

## Observation 2

It was an eighth seventh gesture of 33 years with 3 living children, with a history of a spontaneous abortion, 2 premature deliveries and 1 fetal death in utero in retention. She had no particular medical history, neither smoking nor contraception.

The patient was evacuated from the King Baudouin hospital center for a pregnancy of 32 weeks and 01 day with fetal death in utero. Her prenatal check-up was unremarkable.

The clinical examination had shown a patient in good general condition, apyretic, her conjunctivae were normally colored and her blood pressure was 118/71 mm Hg. The abdomen was enlarged with a fundal height of 30 cm. The uterus was contractile with poor interphase relaxation. Fetal heart sounds were absent. On vaginal examination, the cervix is short, admitting 2 fingers, felt membranes, mobile cephalic presentation and finger cot returns stained with red blood with clots.

In total, it was a ROM (Retained dead egg) complicated by a HRP (Retroplacental Hematoma) in a 33-year-old patient

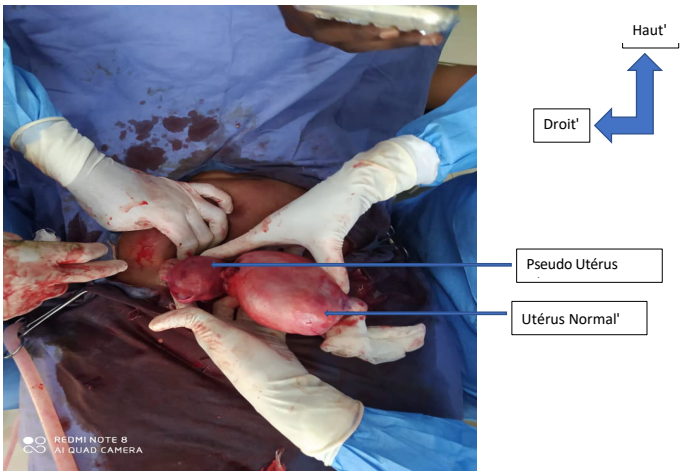


***Figure 3:*** *XXXXX*

with a history of with a heavy history (1 abortion, 2 premature deliveries with children who died a few days after their birth and 1 fetal death in utero in detention).

Conduct maintained: Assessment (NFS, TP/TCK, Creatininemia, etc.) Emergency cesarean section indicated for maternal salvage

On exploration, a non-communicating pseudo-uniform uterus without a cavity was observed. (Figure 5).

Thus, a low transverse hysterotomy was performed allowing the extraction of a stillborn fetus and delivery.

The Pseudo uterus was left in place initially.

Two weeks after the caesarean, the patient was called in for an ultrasound (Figure 6) and at the same time to propose a cure for this malformation by removal of the pseudo uterus.

This cure was done four months later and the postoperative course was simple.

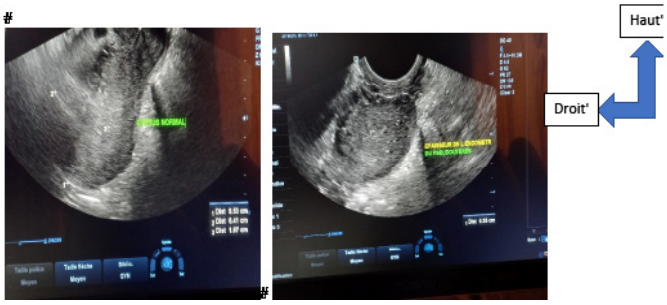


***Figure 6:*** *XXXXX*

## Discussion

### Prevalence

The incidence of congenital uterine malformations in the female population is 3-4% [1,2]. Their frequency is 5-10% in women consulting for recurrent miscarriages and 25% in women with late miscarriages or premature deliveries [2,3]. However, it is difficult to determine the exact prevalence since many of these malformations are asymptomatic and imaging techniques such as Ultrasound, 3D Hysterosonography and MRI are not available in all countries or are not accessible for some populations. The septate uterus is the most common uterine malformation accounting for 30 to 50% of cases followed by malformations of the bicornuate uterus type and unicornuate uterus [2,4].

In our series, we had two cases of pseudounicorn uterus.

### Diagnostic

First of all, it is important to remember that more than 50% of uterine malformations will remain asymptomatic during pregnancy. For others, the malformation will be a source of high-risk pregnancy and obstetric complications. If congenital uterine malformations are present in 3-4% of the fertile and/or infertile female population, their frequency rises from 5-

10% in women consulting for recurrent miscarriages and to 25% in women with miscarriages. late childbirth or premature deliveries [2,3]. The problem in these patients is not that of conceiving, but of carrying the pregnancy to term. Several factors explain this: uterine malformations are associated with a reduced uterine cavity, less efficient musculature, inability to distend, myometrial and cervical dysfunction, inadequate vascularization and poorly developed endometrium. These abnormalities contribute to a higher rate of recurrent miscarriages, premature deliveries, obstructed presentations, intrauterine growth retardation and caesarean sections [2,4,5].

In our series, the patients all presented obstetrical complications such as miscarriages, fetal death in utero and premature deliveries.

In our first observation, the patient consulted for acute abdominal pain. Which is an atypical circumstance of discovery: Most of the patients presenting with a uterine malformation consulted for infertility or obstetric accidents.

The different techniques used in the assessment of uterine malformations are ultrasound, hysterosonography, MRI, hysteroscopy [6-8]. These different techniques can be combined with each other. 3D ultrasound and MRI are currently the techniques showing the best results in terms of sensitivity and specificity [9,10].

Evaluation of uterine malformations should be supplemented by renal imaging to detect frequently associated urinary tract malformations.

In our series, the discovery was fortuitous intraoperatively despite the multiple ultrasounds performed during the pregnancies and the obstetrical complications suffered by these patients. This testifies to the diagnostic difficulty of these pathologies due to their clinical polymorphism or the lack of diagnostic means in our regions.

### Management and treatment of uterine malformations

AThe management of uterine malformations before pregnancy includes surgical treatment whenever indicated and possible. Septate uteri are the only uterine malformations for which surgical treatment is relatively simple and consists of resection of the septum by hysteroscopy. This treatment is only indicated in symptomatic patients who have had obstetric complications in their history [10-12]. Given the simplicity of the procedure and the low morbidity rate, some authors recommend treating this malformation as soon as it is diagnosed with the intention of reducing the rate of late miscarriages [13].

For unicorn or bicervical bicornuate uterus, the reunifying surgery of the two hemi-uteri described by Strassmann [14] in 1952 did not show any real benefit.

For unicornuate uteri with a contralateral rudimentary horn, the main risk is to see a pregnancy develop in the rudimentary horn with the risk of rupture of the blind hemi-uterus. Therefore, a rudimentary horn resection is recommended when an endometrium is present [15].

In partial bilateral uterine agenesis, no surgery can be proposed to allow pregnancy. The creation of a new vagina must be proposed.

For DES uteri, enlargement surgery can provide an improvement when there is mid-cavitary stricture and recurrent miscarriages [16,17].

When the diagnosis of uterine malformations is made in early pregnancy, treatment will only be preventive (rest, lung maturation and ultrasound monitoring of fetal growth and cervical competence) [18].

Cervical cerclage should only be offered in cases of proven cervical incompetence, which is observed in 25-30% of cases of uterine malformations [3,19,20].

## Conclusion

Uterine malformations are relatively common and often asymptomatic. Their direct impact is difficult to assess. They can be manifested by gynecological disorders or have

an impact on reproduction and thus they must be sought in the presence of certain gynecological disorders and certain accidents of pregnancy. Their management largely depends on the type of malformation. Psychological care must be undertaken especially for malformations incompatible with pregnancy or compromising sexuality.

## References


1. Nahum GG. Uterine abnormalities. How common are they, and what is their distribution among subtypes? J Reprod Med 1998;43:877-87.
2. Raga F, Bauset C, Remohi J, et al. Reproductive impact of congenital Mullerian anomalies. Hum Reprod 1997;12:2277-81.
3. Acien P. Reproductive performance of women with uterine malformations. Hum Reprod 1993;8:122-6.
4. * Poncelet C, Aissaoui F. Uterine malformations and reproduction. Gynecol Obstet Fertil 2007;35:821-5.
5. Rackow BW, Arici A. Reproductive performance of women with mullerian anomalies. Curr Opin Obstet Gynecol 2007;19:229-37.
6. Troiano RN, McCarthy SM. Mullerian duct anomalies: Imaging and clinical issues. Radiology 2004;233:19-34.
7. * Deutch TD, Abuhamad AZ.The role of 3-dimensional ultrasonography and magnetic resonance imaging in the diagnosis of Mullerian duct anomalies: A review of the literature. J Ultrasound Med 2008;27:413-23.
8. Braun P, Grau FV, Pons RM, Enguix DP. Is hysterosalpingography able to diagnose all uterine malformations correctly? A retrospective study. Eur J Radiol 2005;53:274-9.
9. Salim R, Woelfer B, Backos M, Regan L, Jurkovic D. Reproductibility of three- dimensional ultrasound diagnosis of congenital uterine anomalies. Ultrasound Obstet Gynecol 2003;21:578-82.
10. Woelfer B, Salim R, Banerjee S, et al. Reproductive outcomes in women with congenital uterine anomalies detected by three-dimensional ultrasound screening. Obstet Gynecol 2001;98:1099-103.
11. Heinonen PK. Complete septate uterus with longitudinal vaginal septum. Fertil Steril 2006;85:700-5.
12. Lourdel E, Cabry-Goubet R, Merviel P, et al. Partitioned uterus: place of hysteroscopic hysteroplasty. Gynecol Obstet Fertil 2007;35:811-8.
13. Grimbizis GF, Camus M, Tarlatzis BC, Bontis JN, Devroey P.Clinical implications of uterine malformations and hysteroscopic treatment results. Hum Reprod Update 2001;7:161- 74.
14. Strassmann EO. Plastic unification of double uterus; A study of 123 collected and five personal cases. Am J Obstet Gynecol 1952;64:25-37.
15. Canis M, Wattiez A, Pouly JL, et al. Laparoscopic management of unicornuate uterus with rudimentary horn and unilateral extensive endometriosis: Case report. Hum Reprod 1990;5:819-20.
16. Garbin O, Ziane A, Castaigne V, Rongieres C. Do septum sections and enlargement plasties really have an obstetrical impact? Gynecol Obstet Fertil 2006;34:813-8.
17. 8 Aubriot FX, Hamou J, Dubuisson JB, Frydman R, Fernandez H. Enlargement hysteroplasty: about the results. Gynecol Obstet Fertil 2001;29:888-93.
18. Airoldi J, Berghella V, Sehdev H, Ludmir J. Transvaginal ultrasonography of the cervix to predict preterm birth in women with uterine anomalies. Obstet Gynecol 2005;106:553-6.
19. Golan A, Langer R, Neuman M, et al. Obstetric outcome in women with congenital uterine malformations. J Reprod Med 1992;37:233-6.
20. Golan A, Langer R, Wexler S, et al. Cervical cerclageits role in the pregnant anomalous uterus. Int J Fertil 1990;35:164-70.

***Figure 5**. Summary receiver operator characteristic curve of sFlt-1 (a), PlGF (c) and sFlt-1/PlGF (c).*

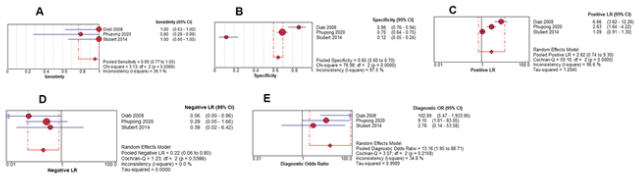

***Figure 6**. Forest plot of sFlt-1 predicting summary sensitivity (a), specificity (b), positive likelihood ratio (c), negative likelihood ratio (d), and diagnostic odds ratio (e) of EO-PE.*

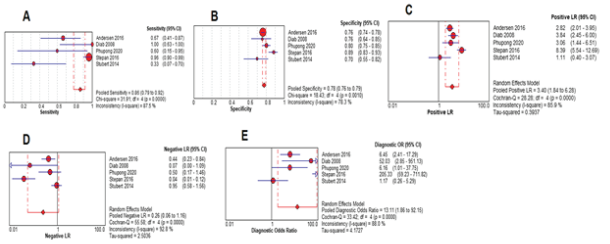

***Figure 7**. Forest plot of PlGF predicting summary sensitivity (a), specificity (b), positive likelihood ratio (c), negative likelihood ratio (d), and diagnostic odds ratio (e) of EO-PE.*

***Table 2.** Summary of meta analysis results*

| Indicator | Index | Merger value | 95% CI | I2(%) | Cochran-Q | P |
|---|---|---|---|---|---|---|
| sFlt-1 | Sen | 0.811 | 0.783–0.837 | 86.6 | 111.71 | <0.001 |
| | Spe | 0.786 | 0.769–0.802 | 96.1 | 388.12 | <0.001 |
| | PLR | 5.097 | 3.498–7.426 | 92.5 | 199.92 | <0.001 |
| | NLR | 0.265 | 0.164–0.430 | 92.5 | 199.81 | <0.001 |
| | DOR | 21.092 | 10.857–40.976 | 84.9 | 99.16 | <0.001 |
| subgroup | Sen | 0.826 | 0.780-0.866 | 51.3 | 4.11 | 0.128 |
| | Spe | 0.691 | 0.649-0.730 | 52.9 | 4.25 | 0.119 |
| | PLR | 2.661 | 2.316-3.056 | 0.0 | 1.64 | 0.440 |
| | NLR | 0.256 | 0.199-0.329 | 3.1 | 2.07 | 0.356 |
| | DOR | 11.251 | 7.872-16.081 | 0.0 | 0.11 | 0.947 |
| PLGF | Sen | 0.735 | 0.713–0.757 | 83.1 | 159.78 | <0.001 |
| | Spe | 0.731 | 0.721–0.741 | 96.1 | 693.94 | <0.001 |
| | PLR | 4.053 | 3.150–5.214 | 90.6 | 287.00 | <0.001 |
| | NLR | 0.341 | 0.275–0.423 | 83.1 | 159.84 | <0.001 |
| | DOR | 14.150 | 8.972–22.315 | 86.2 | 195.03 | <0.001 |
| sFlt-1/PLGF | Sen | 0.779 | 0.763–0.795 | 86.4 | 295.18 | <0.001 |
| | Spe | 0.885 | 0.881–0.889 | 98.6 | 2855.25 | <0.001 |
| | PLR | 6.385 | 4.847–8.410 | 96.5 | 1136.54 | <0.001 |
| | NLR | 0.241 | 0.192–0.303 | 88.4 | 345.59 | <0.001 |
| | DOR | 31.431 | 19.681-50.197 | 91.5 | 470.98 | <0.001 |

Sen: sensitivity;Spe:specificity;PLR:positive likelihood ratio;NLR:negative likehood ratio;DOR:diagnostic odds ratio;95%CI:95% confidence interval.

***Table 3.*** *Summary of EO-PE and LO-PE meta analysis results*

| Indicator | Index | Merger value | 95% CI | I2(%) | Cochran-Q | P |
|---|---|---|---|---|---|---|
| sFlt-1 (EO-PE,n=3) | Sen | 0.955 | 0.772-0.999 | 36.1 | 3.13 | 0.209 |
| | Spe | 0.652 | 0.603-0.698 | 97.5 | 78.98 | <0.001 |
| | PLR | 2.615 | 0.735-9.304 | 96.6 | 59.1 | <0.001 |
| | NLR | 0.217 | 0.059-0.798 | 0 | 1.23 | 0.54 |
| | DOR | 13.16 | 1.952-88.713 | 34.8 | 3.07 | 0.216 |
| PlGF (EO-PE,n=5) | Sen | 0.862 | 0.788-0.917 | 87.5 | 31.91 | <0.001 |
| | Spe | 0.776 | 0.758-0.793 | 78.3 | 18.43 | 0.001 |
| | PLR | 3.401 | 1.844-6.275 | 85.9 | 28.28 | <0.001 |
| | NLR | 0.259 | 0.058-1.160 | 92.8 | 55.58 | <0.001 |
| | DOR | 13.108 | 1.865-92.146 | 88 | 33.42 | <0.001 |
| PlGF (LO-PE,n=2) | Sen | 0.776 | 0.714-0.830 | 94.4 | 17.73 | <0.001 |
| | Spe | 0.682 | 0.662-0.703 | 47.9 | 1.92 | 0.166 |
| | PLR | 2.331 | 1.974-2.752 | 54.5 | 2.2 | 0.138 |
| | NLR | 0.273 | 0.082-0.911 | 92.1 | 12.61 | <0.001 |
| | DOR | 8.572 | 2.254-32.603 | 90.4 | 10.37 | 0.001 |
| sFlt-1/PlGF (EO-PE,n=12) | Sen | 0.944 | 0.921-0.961 | 69.1 | 35.64 | <0.001 |
| | Spe | 0.805 | 0.790-0.819 | 96.5 | 317.71 | <0.001 |
| | PLR | 13.751 | 4.948-38.216 | 96.9 | 358.78 | <0.001 |
| | NLR | 0.084 | 0.048-0.147 | 55 | 24.44 | 0.011 |
| | DOR | 230.24 | 63.956-828.82 | 79.2 | 52.93 | <0.001 |
| sFlt-1/PlGF (LO-PE,n=7) | Sen | 0.72 | 0.680-0.757 | 91.6 | 71.8 | <0.001 |
| | Spe | 0.72 | 0.702-0.737 | 97.9 | 280.66 | <0.001 |
| | PLR | 6.148 | 2.717-13.912 | 96.2 | 156.43 | <0.001 |
| | NLR | 0.318 | 0.194-0.522 | 92.7 | 81.89 | <0.001 |
| | DOR | 20.997 | 5.947-74.132 | 93.7 | 95.25 | <0.001 |

EO-PE:early onset preeclampsia;LO-PE:late onset preeclampsia;Sen: sensitivity;Spe:specificity;PLR:positive likelihood ratio;NLR:negative likelihood ratio;DOR:diagnostic odds ratio;95%CI:95% confidence interval.

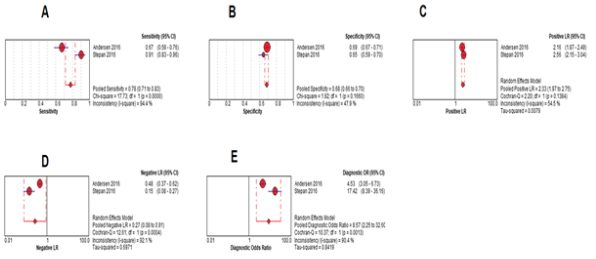


***Figure 8:*** *Forest plot of PlGF predicting summary sensitivity (a), specificity (b), positive likelihood ratio (c), negative likelihood ratio (d), and diagnostic odds ratio (e) of LO-PE.*

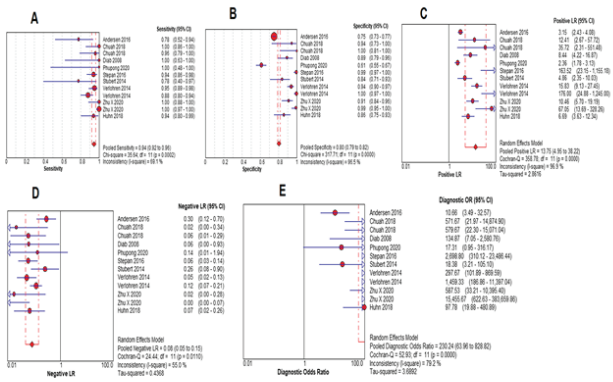


***Figure 9:*** *Forest plot of sFlt-1/PlGF predicting summary sensitivity (a), specificity (b), positive likelihood ratio (c), negative likelihood ratio (d), and diagnostic odds ratio (e) of EO-PE..*

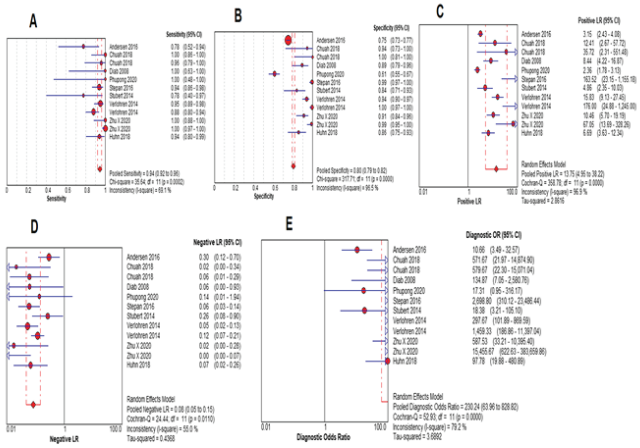


***Figure 10:*** *Forest plot of sFlt-1/PlGF predicting summary sensitivity (a), specificity (b), positive likelihood ratio (c), negative likelihood ratio (d), and diagnostic odds ratio (e) of LO-PE.*

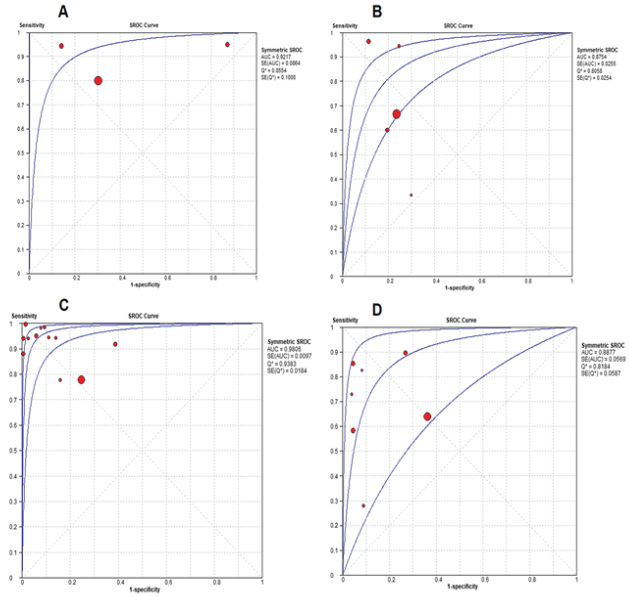


***Figure 11:*** *Symmetric receiver operator characteristic curve of sFlt-1 predicting EO-PE (a), PlGF predicting EO-PE (b), sFlt-1/PlGF predicting EO-PE (c) and sFlt-1/PlGF predicting LO-PE (d)..*