Case Report

First case report of horseshoe appendix in Morocco according according to SCARE guidelines

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\section*{ABSTRACT}

\textbf{Introduction:} The appendiceal duplication is a sporadic malformation in which the horseshoe form is the uncommon described variant. To our knowledge, we report the first Moroccan case of a horseshoe appendix in a girl admitted to managing of pain at the right iliac fossa.

\textbf{Case presentation:} Through this article, we present a very rare case of appendicular duplication. It has not been objectified in radiological exploration and discovered by chance during the operation. Resection then closure of the appendicular bases allowed our patient to heal.

The objectives of this work is threefold: i) to report this sporadic case of horeshoe appendix, ii) to emphasize the importance of suspicion of appendicular duplication in appendicular syndrome and iii) to recommend the exploration of the ileocecal region to avoid surgical complications and medicolegal problems.

\textbf{Conclusion:} Our case report shows that we have to take into consideration this sporadic presentation of appendicular syndrome and this even in the absence of radiological signs. Our work brings enriched the literature by a new case of horseshoe appendicitis highlighting the importance of surgical treatment.

\section{1. Introduction}

Duplication of the vermiform appendix is a sporadic malformation [1]. It is associated with other appendix malformations described in the literature such as a congenital absence of the vermiform appendix and the intracecal appendix [2,3]. The horseshoe appendix is the rarest and the most recently described type of this entity [4]. Several classifications are available to guide surgeons to diagnose this variation [5,6]. Radical surgery is the treatment of choice, enables a successful management, and avoids recurrence, as well as medico-legal problems [7–9]. To date, only few cases describing this anatomical variation were published. We report in this paper the first Moroccan case of a horseshoe appendix according to SCARE guidelines [10] (see Table 1).

\section{2. Clinical case}

A 26-year-old married woman without children from eastern Morocco and under insulin treatment for type I diabetes for 6 years was admitted to the emergency department for ketoacidosis and presented with acute onset of abdominal pain and vomiting. The clinical examination found a patient with fever at 39°C with a positive urine test strip and tenderness in the right iliac fossa (Alvarado score > 6). We performed an infectious assessment that showed hyperleukocytosis (16,000/mm\(^3\)), an increase of C-reactive protein (CRP) at 66 mg/L with slight metabolic acidosis on arterial gasometry. Ultrasound examination was in favor of acute appendicitis. After discussion with the patient, the consent for surgical exploration was obtained and showed a horseshoe appendix (Fig. 1). An appendectomy with control of both appendicular bases was performed by the head of the digestive surgery department who was called upon in the operating room by his team in the face of this anomaly. The intervention went well, tolerated by the patient, without any unwanted events. The patient was discharged three days later after the control of ketoacidosis and penicillin-based oral therapy (amoxicillin 3g/day) for 6 days was prescribed. The histopathological examination of the surgical specimen was in favor of suppurrative appendicitis and our patient was satisfied with the medical and surgical management.
### Table 1
Summary of published case reports on horseshoe appendix.

| Author/Year          | Country | Diagnosis                                      | Anatomical location | Surgical procedure       |
|----------------------|---------|------------------------------------------------|---------------------|--------------------------|
| Mesko et al., 1989   | USA     | Appendicitis                                   | Unclear             | Appendectomy             |
| Dong et al., 1994    | China   | Bowel occlusion                                | Cecum-cecum         | Appendectomy             |
| Dasgupta et al., 1994 | England | Appendicular mass                              | Cecum-cecum         | Appendectomy             |
| Zhu et al., 2000     | China   | Appendicitis                                   | Cecum-cecum         | Appendectomy             |
| Liu et al., 2006     | China   | Appendicitis                                   | Cecum-cecum         | Appendectomy + enterotomy|
| Takabatake et al., 2010 | Japan | Tubulovillous adenoma in ascending colon       | Cecum-ascending colon | Ileocecal resection      |
| Singh et al., 2016   | India   | Appendicitis                                   | Cecum-cecum         | Appendectomy             |
| Cai and Yu, 2011     | South Africa | Appendicitis                                  | Cecum-hepatic flexure of colon | Appendectomy |
| Li and Yu, 2012      | China   | Appendicitis and bowel occlusion               | Cecum-cecum         | Appendectomy + enterotomy|
| Bulut et al., 2013   | Turkey  | Appendicitis                                   | Cecum-cecum         | Appendectomy             |
| Calota et al., 2010  | Romania | Bowel occlusion                                | Cecum-cecum         | Appendectomy             |
| Ninos et al., 2010   | Greece  | B Cell non Hodgkin’s lymphoma                  | Cecum-cecum         | Appendectomy + chemotherapy |
| Dube et al., 2011    | South Africa | Appendicitis                                  | Cecum-cecum         | Appendectomy             |
| Li and Liu, 2019     | China   | Appendicitis and bowel occlusion               | Cecum-cecum         | Appendectomy + enterotomy|
| Oruç et al., 2016    | Turkey  | Appendicitis                                   | Cecum-cecum         | Appendectomy             |
| Liu et al., 2017     | China   | Appendicular mass                              | Cecum-cecum         | Appendectomy             |
| Zhu et al., 2019     | China   | acute appendicitis and endometriosis           | Cecum-cecum         | Appendectomy, oophorectomy and partial resection of the small intestine |

**Fig. 1.** Horsesho appendix with two different bases

### 3. Discussion

Duplication of the vermiform appendix is an extremely rare malformation with an estimated incidence of 0.004% [1]. It was first described by Picoli in 1892 [25]. It also described other rare anatomical variations such as acute appendicitis with absence of appendix [2] and appendicular ectopia [3]. Cave classified appendix duplication in 1936 according to the anatomical location [5]. Since then, this classification was updated by Wallbridge in 1963 [6] and was named “Cave-Wallbridge”. In 2010, Calota et al. added the horseshoe shape to the old classification [4]. Recently other variations such as the triple appendix [26] have been described. We reported a new case of a horseshoe appendix that is a very rare morphological entity in addition to the fifteen reported cases described in the literature.

Several hypotheses have explored the genesis of the a horseshoe appendix such as fusion of the two appendicular points, appendicocolocolic fistula and division of the appendicular mesoappendix explained by the presence of a single vessel in the mesoappendix [22]. The intraoperative fortuitous discovery often dominates diagnosis circumstances for suspicion of appendicitis or during surgery for other indications [27,28]. In our patient, appendicitis was strongly suspected with an Alvarado score greater than 6. Preoperative radiology rarely helps in the diagnosis of appendicular duplication. However, one reported case of fortuitous discovery on a barium radiological examination was described in a patient admitted for other medical conditions [29]. Liu et al. [4] reported two cases of a horseshoe appendix diagnosed preoperatively on ultrasound examination with 3D reconstruction in addition to other cases after re-reading the CT scan after postoperative discovery of this very rare variation during surgery. In our case, the radiology team suspected acute appendicitis, and our surgical exploration found horseshoe appendicitis with a single vessel on the mesoappendix. The evaluation of the cecum must be rigorously performed to avoid any forensic problems if a new inflammation affects the remaining annex [7,8]. It also allows better assessment of differential diagnosis of duplication with the cecal diverticulitis as well as colon tumors [30,31]. Removal of both appendices is important to avoid confusion and additional re-interventions [9]. Moreover, an additional dissection of the cecum should be performed to eliminate duplication at the retrocecal area [16].

### 4. Conclusion

Appendicular duplication is a sporadic malformation that often escapes preoperative radiological imaging. Surgeons must rigorously examine the ileocecal region in order to avoid missing any appendicular duplication in case of symptomatic recurrence, and we insist on the radical surgical treatment which alone allows the cure.

### Patient perspective

The procedure of surgery was explained to the patient with all advantages and possible complications. He agreed on the procedure and informed consent was taken from her.

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Ethics approval

not applicable.

Consent of patient

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author’s contribution

Jabi Rachid: Writing, review and editing of the manuscript.
Siham Elmir: Contributed for diagnose and treatment of the patient.
Mohamed Bouziane: Review, Supervision and surgeons of the patient.

Registration of research studies

1 Name of the registry:
2 Unique identifying number or registration ID:
3 Hyperlink to your specific registration (must be publicly accessible and will be checked):

Guarantor

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References

[1] A.A. Kothari, K.R. Yagnik, V.P. Hathila, Duplication of veriform appendix, J. Postgrad. Med. 50 (4) (2004) 285–286.
[2] A. Sarkar, Congenital absence of the veriform appendix, Singap. Med. J. 53 (9) (2012) e189–e191.
[3] S. Chauhan, S. Anand, Intracecal appendix: an extremely rare anatomical variation. A case report and review of literature, Surg. Radiol. Anat. 40 (3) (2018) 111–114.
[4] J. Liu, C. Dong, H. Wang, et al., One type of duplex appendix: horsehoe appendix, Therapeut. Clin. Risk Manag. 14 (2018) 1987–1992, https://doi.org/10.2147/TCRM.S179929, Published 2018 Oct 12.
[5] A.J. Cave, Appendix vermiformis duplex, J. Anat. 70 (Pt 2) (1936) 283–292.
[6] P.H. Wallbridge, Double appendix, Br. J. Surg. 50 (1962) 346–347, https://doi.org/10.1002/bjs.1800502214.
[7] S. Tutcu Şahin, Y. Erhan, H. Aydede, Double acute appendicitis in appendical duplication, Ulus Travma Acil Cerrahii Derg 19 (1) (2013) 83–85, https://doi.org/10.5505/itj.2013.80557.
[8] J.R. Travis, J.L. Weppner, J.C.baugh 2nd, Duplex veriform appendix: case report of a ruptured second appendix, J. Pediatr. Surg. 43 (9) (2008) 1726–1728, https://doi.org/10.1016/j.jpedsurg.2008.04.023.
[9] A.A. Kothari, K.R. Yagnik, V.P. Hathila, Duplication of veriform appendix, J. Postgrad. Med. 50 (4) (2004) 285–286.
[10] A. Agba, T. Franchi, C. Sohrabi, G. Mathew, A. Kerwan, SCARE group. The SCARE 2020 guideline: updating consensus surgical case REport (SCARE) guidelines, Int. J. Surg. 84 (2020 Dec) 226–230, https://doi.org/10.1016/j.ijsu.2020.10.034. Epub 2020 Nov 9, PMID: 33181358.
[11] T.W. Mesko, R. Lugo, T. Breitholtz, Horsehoe anomaly of the appendix: a previously undescribed entity. Surgery 106 (3) (1989) 563–566.
[12] Z. Dong, X. Fu, H. Luo, Horsehoe appendix induced intestinal obstruction, Chinese J Clinical Anatomy 12 (4) (1994) 309–310.
[13] R. DasGupta, P.U. Reber, A.G. Patel, Horsehoe appendicitis, Eur. J. Surg. 165 (11) (1999) 1095–1096, https://doi.org/10.1002/ejso.5901651108.
[14] C. Li, J. Yu, A horsehoe appendix, Guangdong Med J 21 (11) (2000) 982–983.
[15] S. Cai, M. Lin, A case of annular appendix with acute appendicitis, J. Med. Imaging 17 (1) (2017) 27–28.
[16] F. Calot, I. Vasilis, S. Mogoantă, et al., Horsehoe appendix: A extremely rare anomaly, Chirurgia (Bucur) 105 (2) (2010) 271–274.
[17] A. Ninos, G. Douridas, E. Papakonstantinou, et al., A horsehoe appendix double positioned on a non-Hodgkin lymphoma, Hellenic J Surgery 82 (1) (2010) 73–76.
[18] B. Dube, G.R. Manoharan, M. Daya, Anomalous origin of the veriform appendix, S. Afr. J. Surg. 49 (2011) 100.
[19] J. Liu, D. Li, Man xing huan Zhuzhang lan wei yan yi xi xia qian dian xing hui chang nei zuo yi li, Chin J Gastrointest Surg 15 (6) (2012) 786–787.
[20] C. Oruc, O. Ipş, O. Ureyen, O.S. Kahyaoglu, A. Koseoglu, An extremely rare appendiceal anomaly: horsehoe appendicitis, Ulus Travma Acil Cerrahii Derg 19 (4) (2013) 385–386, https://doi.org/10.5505/itj.2013.67424.
[21] S.P. Bulut, N. Calbuga, M. Akunc, Perforated double appendicitis: horsehoe type, Ulus Cerrahii Derg 32 (2) (2015) 134–136, https://doi.org/10.5152/UCD.2015.2857. Published 2015 Jun 19.
[22] ChG. Singh, K.T. Nyuuri, R. Rangaswamy, Y.S. Ezung, H.M. Singh, Horsehoe appendix: An extremely rare appendiceal anomaly, J. Clin. Diagn. Res. 10 (3) (2016) PD25–PD26, https://doi.org/10.7860/JCDR/2016/16509.7494.
[23] K. Takabatake, J. Ikeda, H. Furuke, et al., A case of a horsehoe appendix, Surg Case Rep 2 (1) (2016) 140, https://doi.org/10.1186/s40792-016-0261-3.
[24] M.V. Zhu, F.M. Fei, J. Chen, Z.C. Zhou, B. Wu, V.Y. Shen, Endometrosis of the duplex appendix: A case report and review of the literature, World J Clin Cases 7 (15) (2019) 2094–2102, https://doi.org/10.21997/wjcc.v7.i15.2094.
[25] A.K. Khanna, Appendix veriformis duplex, Postgrad. Med. 59 (687) (1983) 69–70, https://doi.org/10.1136/pgmj.59.687.69.
[26] H. Nageswaran, U. Khan, F. Hiii, A. Maw, Appendiceal duplication: A comprehensive review of published cases and clinical recommendations, World J. Surg. 42 (2) (2018) 574–581, https://doi.org/10.1007/s00268-017-4178-1.
[27] Ayoub K., Kayali S., Dabbagh M.F., Banjaby B., Acute single appendicitis in a female with a duplicated appendix, J. Surg. Case Rep. 2018 (6) ry132. Published 2018 Jun 19, doi:10.1093/jscr/ry132.
[28] B. Sobhian, M. Mostegel, C. Kunc, J. Karner, Appendix veriformis duplex - eine seltene Überraschung [Appendix veriformis duplex - a rare surprise], Wien Klin. Wochenschr. 117 (13–14) (2005) 492–494, https://doi.org/10.1007/s00508-005-0390-3.
[29] P. Pedu, P.S. Sidhu, Appearance of a type B duplex appendix on barium enema, Br. J. Radiol. 77 (915) (2004) 248–249, https://doi.org/10.1259/bjr/65763477.
[30] E.A. Griffiths, F.G. Bergin, J.A. Henry, A.M. Mudawi, Acute inflammation of a congenital cecal diverticulum mimicking appendicitis, Med Sci Monit 9 (12) (2003) CS107–CS109.
[31] M.K. Bluet, S.A. Halter, K.E. Salhany, J.P. O’Leary, Duplication of the appendix mimicking adenocarcinoma of the colon, Arch. Surg. 122 (7) (1987) 817–820, https://doi.org/10.1001/arch Surg.1987.01400190083016.