Review Article

A Systematic Literature Review of Demographics, Clinical Presentation and Surgical Procedures Performed for the Appendico-Umbilical Fistula for the Past 100 Years

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Abstract

The persistent vitellointestinal duct (PVID) is the rarest of all the vitelline duct anomalies. The enteric end of the vitelline duct mostly connects to the terminal ileum. The appendico-umbilical fistula (appendiceal-umbilical fistula) in which the proximal end of the vitelline duct connects to the appendix is a rare clinical entity. This systematic literature review was carried out to review the demographics, clinical characteristics and operative procedures executed for the appendico-umbilical fistula. Literature/cases with incomplete desired details for the above-mentioned anomaly were excluded. The guidelines of the preferred reporting items for systematic reviews and meta-analyses are followed for presenting this manuscript. An online search was made for the retrieval of the published manuscripts. The search was performed for the previous 100 years from 1921 to December 2021. A total of $n = 21$ ($n = 12$ female and $n = 9$ males) cases of the appendico-umbilical fistula were included in the systematic review. All the cases, except one, were infants and $n = 10$ of the cases were neonates/newborn babies. Their ages ranged from 1 day to 5 years. Three-fourths ($n = 16$) of the children presented with the chief complaint of discharge of mucus/stool through the umbilicus. An appendicectomy with resection of the PVID/fistulous duct/prolapsed mucosa was executed as the standard operative procedure for the entire case. A peri-umbilical incision was preferred and frequently employed for surgical therapy. This review revealed only one (4.76%) post-operative death that was unrelated to the surgical procedure. PVID connected to the appendix, presented clinically as appendico-umbilical fistula is a rare entity. The standard surgical procedure performed for the appendico-umbilical fistula was an appendicectomy with resection of the PVID/fistulous duct/prolapsed mucosa.

Keywords: Appendicectomy, appendico-umbilical fistula, children, congenital anomaly, infants, persistent vitellointestinal duct

Introduction

The vitelline duct connects the embryonic yolk sac to the primitive midgut in the early intrauterine life.[1,2] The vitelline duct obliterates during 4–9 weeks of intrauterine life.[2,3] The vitelline duct fails to obliterate and persists in approximately 2%–4% of the human population.[2] Failure of the obliteration of the vitelline duct results in varieties of anomalies, depending on the part of the vitelline duct that persists.[1-3] The commonest of all vitelline duct anomalies is Meckel's diverticulum. Meckel's diverticulum occurs as the consequence of the failure of obliteration of the vitelline duct towards the enteric site.[1,2,4]

The complete failure of the involution of the vitelline duct results in the persistent vitelline duct. The patent vitelline duct is the rarest of all the vitelline duct anomalies.[1,3,5] The proximal part of the persistent vitelline duct is connected to the terminal ileum in most cases.[3,6,7] The patent vitelline duct connected to the vermiform appendix (appendico-umbilical fistula) is a rare clinical entity.[1,7] The preferred reporting items for systematic reviews and meta-analyses (PRISMA) guidelines are followed for the presentation of this systematic literature review.[8,9] A total of $n = 21$ cases (all children) who were operated upon for the appendico-umbilical fistula (patent vitelline duct connected

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to the vermiform appendix?) were included in this systematic literature review.[10-29]

Materials and Methods

This manuscript is a systematic literature review of the cases operated upon for the patent vitelline duct connected to the vermiform appendix and clinically presented as an appendico-umbilical fistula. This systematic review followed the PRISMA guidelines. For retrieval of the literature/manuscripts, an electronic search was performed. Various websites such as PubMed, PubMed Central, Scopus, ResearchGate and Google Scholar were searched. The keywords employed for the electronic search were 'appendico-umbilical fistula', 'appendico-umbilical fistula', 'appendiceal-umbilical fistula', 'appendiculo-umbilical fistula', 'umbilico-appendicular fistula', 'umbilical-appendicular fistula', 'appendicular-umbilical fistula', 'patent/persistent vitelline duct', 'persistent vitelline duct connecting to the appendix' and 'congenital umbilical fistula'. The persistent vitelline duct connected to the appendix and presented as appendico-umbilical fistula was first reported in the year 1921, and the relevant literature/manuscripts were retrieved from 1921 to December 2021, and preferably those were available in the English language. Two manuscripts/case reports available in a language other than English were also included for this review. Manuscripts/case reports with incomplete desired details were excluded from the review. Appendiceal-umbilical fistula of inflammatory and other causes were excluded. The titles, abstracts and full texts of the manuscripts were assessed for the selection of the manuscripts. This is a single-author review, and the selection of the manuscript, extraction of the desired details and all others was carried out independently by the author alone. The variables/details extracted from the manuscripts were age and sex, clinical characteristics, surgical procedures performed for the anomaly and the outcome. This article is a systematic review of the already published literature/manuscripts, and institutional ethical committee approval is not required.

Results

The results of the literature/manuscripts search for the systematic literature review for the appendico-umbilical fistula/patent vitelline duct connected to the vermiform appendix are provided as a PRISMA flow chart in Figure 1. A total of $n = 20$ manuscripts were recruited for the systematic review. All the manuscripts were full-text articles. One of the manuscripts was an original article and $n = 19$ were case reports. This review included a total of $n = 21$ cases of appendico-umbilical fistula, and all of them were children. The present review included $n = 9$ (42.85%) boys and $n = 12$ (57.14%) girls. The demographics, clinical features, clinical findings, surgical procedures performed and outcome of the children operated upon for the above-mentioned anomaly are detailed in Table 1. The summary of the results obtained from the systematic review are detailed in Table 2. Twenty (95.23%) were infants, and $n = 10$ (47.61%) of the cases were newborn babies. Three-fourths ($n = 16$) of them presented with a chief complaint of discharges of the mucus/faecal matter through the umbilicus. Clinical examination revealed that three-fourths ($n = 16$) of the children delineated an opening/aperture in the umbilicus. The standard surgical procedure carried out for the entire case was an appendicectomy and resection of the persistent vitelline duct/fistulous duct connected to the umbilicus. This review evidenced that $n = 1$ (4.76%) of the children died during the post-operative period.

Discussion

Patent vitellointestinal duct occurs due to the complete failure of obliteration of the vitellointestinal duct.[1,3,5,30] The patent vitellointestinal duct is normally-connected to the terminal ileum.[3,6,7] Persistent vitellointestinal duct (PVID) opening/connected to the vermiform appendix and presenting as a congenital appendico-umbilical fistula is very rare.[10-29]

Summary of Evidence

A total of twenty manuscripts were recruited for the systematic literature review on the management of appendico-umbilical fistula. All the manuscripts included were full-text articles.[10-29] Nineteen of the manuscripts were case reports.[10-12,14-29] Only one of the manuscripts was published as an original article.[13] A total of $n = 23$ cases were retrieved through the online literature search for the management of the appendico-umbilical fistula.[7,10-30] Two of the cases were excluded from review due to the incomplete desired details.[7,30] A total of $n = 21$ cases, all children, were recruited for review and were retrieved from the $n = 20$ of the manuscripts.[10-29] Eighteen of the manuscripts were published and are available in the English language.[10-13,15-18,20-29] Two of the manuscripts that were published and are available in a language other than English were also recruited for review.[14,19]

The PVID occurs 1 in 15,000–16,000 live births and accounted for 0.0063%–0.0067% of hospital births.[31] Literature/manuscripts on the management of appendico-umbilical fistula are scanty.[32] Only $n = 23$ cases of appendicoumbilical fistula were published during the past one century from the 1921 to December 2021.[7,10-30]

The PVIDs are most frequently detected during the neonatal period, similarly, half ($n = 10$) of the cases of the appendico-umbilical fistula were detected during the neonatal period.[13,14,16,18,19,21,23,25,26,28] All the cases ($n = 21$) included for review were children and $n = 20$ of them were detected/evidenced during the infancy.[10,12-29] Among $n = 21$ cases recruited for review, $n = 10$ of them were neonates.[13,14,16,18,19,21,23,25,26,28] The boys-to-girls ratio was 1:1.3 for the children who were surgically treated for the appendicoumbilical fistula.[10-29]

The clinical presentation of the appendico-umbilical fistula was much like that of the cases of PVID and varies from mucus/flatus/feces discharge to the prolapse of the bowel mucosa

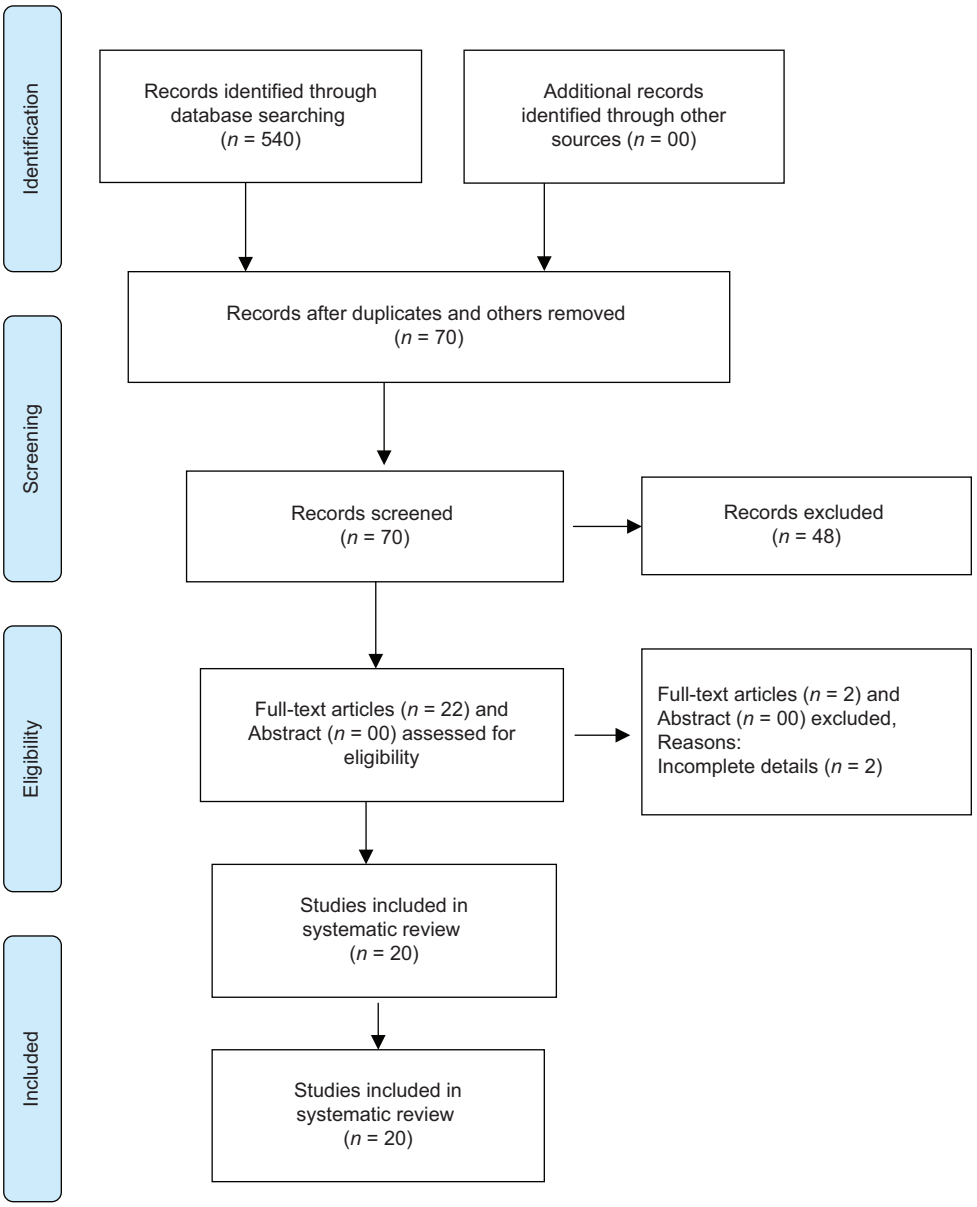


Figure 1: Preferred Reporting Items for Systematic Reviews and Meta-Analyses flow diagram for literature search to the selection for the management of the appendico-umbilical fistula/persistent vitellointestinal duct connected to the vermiform appendix

through the umbilicus. Major prolapses of the ileum/bowel through the PVID/umbilicus is one of the serious complications of the PVID, but it was not detected in any of the cases of the appendico-umbilical fistula. Three-fourths (n = 16) of the children presented with a chief complaint of discharge of mucus/faecal matter through the umbilicus.[12,13,15-22,24-29] Bowel prolapse was not detected in any of the cases, but prolapsed of the mucosa via umbilicus was documented in three-fourths (n = 16) of the children.[10,11,14-17,19,22-29] This finding is probably due to the fact of the smaller lumen diameter documented in the cases of the appendiculo-umbilical fistula. During the clinical examination in n = 18 (85.71%) of the children, a fistulous opening/meatus/apparatus (umbilical fistula) was also evidenced at the umbilicus.[10,12-27,29] A fistulogram was carried-out for delineation of the umbilical fistula/fistulous tract, that was performed in n = 9 (42.85%) of the children. Fistulogram detected that the umbilical fistula was connected to the caecum/colon, but none detected that it was connected to the appendix.[12,14,16-18,20-23]

The pre-operative/provisional diagnosis of either a PVID or enteric-umbilical fistula/umbilico-enteric fistula was made for the entire children. Surgical therapy was offered for the entire children. During the abdominal exploration, the proximal end (enteric side) of the umbilical fistula was connected to the vermiform appendix in all the children. An appendicectomy and resection of the PVID/fistulous duct/prolapsed mucosa was the standard surgical procedure performed for the entire children.[10-29] Peri-umbilical incision (infraumbilical/supraumbilical incision) was preferred for four-fifths (n = 17)

Table 1: Demographics, clinical features and operative procedures performed for the cases of the appendico-umbilical fistula (*n*=21)

Author publication year [reference number]	Age and sex	Clinical presentation and findings	Fistulogram	Surgical procedures performed	Mobile cecum	Remark
Crymble, 1921[10]	5 months male	Mucus/faecal discharge-No Prolapse-Yes	Not done	Appendicectomy and resection of the persistent vitelline duct	Yes	Well
Singleton and King, 1951[11]	5 years male	Mucus/faecal discharge-No Prolapse-Yes	Not done	Appendicectomy and excision of the prolapsed mucosa	Yes	Well
Sandborn and Shafer, 1967[12]	4 months female	Mucus/faecal discharge-Yes Prolapse-No	Yes	Appendicectomy and excision of the umbilical granuloma	NA	Well
Rao *et al.*, 1979[13]	1 month male	Mucus/faecal discharge-Yes Prolapse-No	Not done	Appendicectomy and resection of the patent duct	NA	Death
Hasemura *et al.*, 1987[14]	Neonate female	Mucus/faecal discharge-No Prolapse of mucosa-Yes	Yes	Appendicectomy and resection of the prolapsed mucosa	NA	Well
Kadzombe and Currie, 1988[15]	6 months female	Mucus/faecal discharge-Yes Prolapse of mucosa-Yes	Not done	Appendicectomy and resection of the prolapsed mucosa	NA	Well
Borgna-Pignatti *et al.*, 1995[16]	Neonate male	Mucus/faecal discharge-Yes Prolapse of mucosa-Yes	Yes	Appendicectomy and resection of the prolapsed mucosa	Yes	Well
Zaidenstein *et al.*, 1995[17]	2 months male	Mucus/faecal discharge-Yes Prolapse of mucosa-Yes	Yes	Appendicectomy and resection of the prolapsed mucosa	NA	Well
Crankson *et al.*, 1998[18]	Neonate female	Mucus/faecal discharge-Yes Prolapse of mucosa-No	Yes	Appendicectomy and adhesiolysis	NA	Well
Kim *et al.*, 2004[19]	Neonate female	Mucus/faecal discharge-No Prolapse of mucosa-Yes	Not done	Appendicectomy and resection of the prolapsed mucosa	NA	Well
Pal and Singh, 2006[20]	2 months male	Mucus/faecal discharge-Yes Prolapse of mucosa-No	Yes	Appendicectomy and resection of the fistulous duct	NA	Well
Fuijkschot *et al.*, 2006[21]	Neonate female	Mucus/faecal discharge-Yes Prolapse of mucosa-No	Yes	Appendicectomy and resection of the fistulous duct	Yes	Well
Salah *et al.*, 2009[22]	2 months female	Mucus/faecal discharge-Yes Prolapse of mucosa-Yes	Yes	Appendicectomy and resection of the fistulous duct	Yes	Well
Cevik *et al.*, 2011[23]	Neonate male	Mucus/faecal discharge-No Prolapse of mucosa-Yes	Yes	Appendicectomy and resection of the fistulous duct	Yes	Well
Montes-Tapia *et al.*, 2012[24]	2 months male	Mucus/faecal discharge-Yes Prolapse of mucosa-Yes	Not done	Appendicectomy and resection of the prolapsed mucosa	NA	Well
Guerron *et al.*, 2013[25]	Neonate female	Mucus/faecal discharge-Yes Prolapse of mucosa-No	Not done	Appendicectomy and resection of the fistulous duct	Yes	Well
Sani *et al.*, 2015[26]	Neonate female	Mucus/faecal discharge-Yes Prolapse of mucosa-Yes	Not done	Appendicectomy and resection of the fistulous duct	NA	Well
OlaOlorun *et al.*, 2015[27]	2 months female	Mucus/faecal discharge-Yes Prolapse of mucosa-Yes	Not done	Appendicectomy and resection of the persistent vitelline duct	Yes	Well
Ghoroubi *et al.*, 2018[28]	1 month female	Mucus discharge-Yes Prolapse of mucosa-Yes	Not done	Appendicectomy and resection of the fistulous duct	Yes	Well
Ghoroubi *et al.*, 2018[28]	45 days male	Mucus discharge-Yes Prolapse of mucosa-Yes	Not done	Appendicectomy and resection of the fistulous duct	Yes	Well
Delforge *et al.*, 2019[29]	2 months female	Mucus discharge-Yes Prolapse of mucosa-Yes	Not done	Appendicectomy and resection of the fistulous duct	NA	Well

NA=Not available/details not available

of the cases for the surgical correction of the above-mentioned anomaly.[10,11,14,15,18-29] For the entire children, the resected specimen was submitted for histological examination and all were reported as appendix.[10-29] The major post-operative surgical complications were not documented/not-mentioned for any of the cases reviewed. This systematic review documented only n = 1 (4.76%) death, which was observed during the post-operative period. The infant died due to aspiration. Death was not related to the surgical procedure performed.[13]

In general, the appendicular fistula is grouped/categorised into two: external and internal fistula.[33] In the external appendicular fistula, the connection of the appendix is to the exterior/anterior abdominal wall and most frequently to the right lower quadrant of the abdomen.[33,34] In the internal appendicular fistula, the connection of the appendix is to the bowel/gastrointestinal tract (GIT), urinary bladder, female genital tract and rarely to the vascular system.[33,34] In bowel fistulae, the appendiculo-ileal fistula was the most common, and it was

Table 2: Summary of the results of the management of appendico-umbilical fistula

Description	Variables	Number of cases, *n* (%)
Cases included	Total	21 (100)
Sex distribution	Boys	9 (42.85)
	Girls	12 (57.14)
Age distribution	Neonates	10 (47.61)
	Infants (excluding neonates)	10 (47.61)
	13 months and onwards	1 (4.76)
Age of presentation	Median	1 month
	Range	1 day–5 years
	Presented within 2 months of life	17 (80.94)
Chief complaints	Mucus/faecal discharges through umbilicus	16 (76.19)
	umbilical granuloma/mass	16 (76.19)
Clinical findings	Mucus/faecal discharges through umbilicus	16 (76.19)
	Prolapsed mucosa/umbilical granuloma/umbilical mass	16 (76.19)
	Fistulous opening (umbilical fistula)	18 (85.71)
Investigation	Fistulogram	9 (42.85)
Operative procedure executed	Appendicectomy with resection of the patent vitelline duct/fistulous duct	21 (100)
Incision (operative)	Peri-umbilical	17 (80.95)
	Right transverse	2 (9.52)
	Details not available	2 (9.52)
Mortality	Post-operative	1 (4.76)
Histology	Excised specimen	Appendix (*n*=21; 100)

due to their proximity to each other.[33,34] McDonald (2020), in a systematic review of the literature relating to the fistulae involving the appendix (n = 301) and found that the most common site of the fistulae was the urinary bladder. Other sites were the GIT, skin/abdominal wall, umbilicus, vascular system and female genital tract.[34] On further review, they found that in half of the cases, appendicitis was the cause of the appendicular fistulae.[34] Due to the possibilities of fistulous connection of the appendix either to the bowel, urological system, female genital tract, vascular system, external abdominal wall, or umbilicus, it is always better to have pre-operative knowledge of the etiopathogenesis. For the suspected congenital appendicular/umbilical fistula, pre-operative knowledge of its embryological origin is helpful for planning an appropriate surgical procedure.

The exact aetiology/embryogenesis for the congenital appendico-umbilical fistula is not clearly understood due to the paucity of literature. Collins (1955) in a study of 50,000 specimens of the human vermiform appendix reported that only three of the cases had fistula of the umbilicus to the appendix. For the above three cases, the author did not describe their aetiology as congenital.[33] Eckstein (1963) in a review of 100 consecutive cases of exomphalos reported that in two of the patients the appendix was the content within the exomphalos sac.[35] McDonald *et al*. in a systematic literature review on fistulae involving the appendix also included n = 16 cases of appendico-umbilical fistulae of congenital origin and expressed that their aetiology remains unclear.[34] Two of the explanation provided/expressed by various authors for the above-described anomaly. One of the explanations is that it is a variant of the persistent vitelline duct where the proximal end of the duct was connected to the appendix, rather than the terminal ileum.[10,11,17,18-20,23,27] Another explanation was that the tip of the appendix was entrapped at the umbilical ring or presented as a content of the small unrecognisable omphalocele/hernia of the cord and either ruptured or accidentally clamped/ligated at the umbilical cord and later presented as an appendiculo-umbilical fistula.[12,14-16,21,22,25] Some of the authors of the above cases thought that it was a case of simply appendico-umbilical fistula rather than a variant of the PVID connecting to the appendix. Amongst the above n = 21 cases reviewed; a mobile caecum was documented in n = 10 of the cases.[10,11,16,21-23,25,27,28] Whether the presence of mobile cecum plays any major role in the causation of appendico-umbilical fistula is also not clearly defined.

An acquired appendico-umbilical fistula is also a rare clinical condition and mostly it was documented as a consequence of appendicitis/appendicular perforation.[20,36,37] Congenital appendiculo-umbilical fistula is most frequently detected during infancy. In the present systematic review of the literature, 95.23% were infants and 47.61% of the cases were newborn babies. The age of presentation is not necessarily during the infancy/neonatal period for the acquired appendico-umbilical fistula.[20,36,37] Clinically, the cases of the acquired appendiculo-umbilical fistula presented mostly as purulent umbilical discharge.[20,36,37] The cases of congenital appendiculo-umbilical fistula presented with a chief complaint of discharges of the mucus/faecal matter through the umbilicus. For congenital appendico-umbilical fistula, the clinical examination also delineated an opening/aperture in the umbilicus in three-fourths of the children. In three-fourths of the cases, prolapse of the mucosa via the umbilicus was also documented for congenital appendiculo-umbilical fistula. For the acquired appendico-umbilical fistula, it is not necessarily to have mucosal prolapse through the umbilicus. During the operative procedure for the acquired appendiculo-umbilical fistula cases, intra-abdominal/bowel adhesions and features of inflammation of the appendix were evidenced.[20,36] A perforated appendix with multiple intraabdominal abscesses has also been documented in a case of an acquired appendiculo-umbilical fistula.[37] Histological evaluation of the operative specimen (appendix and fibrous tract/tissues) of the above cases also supported the acquired nature of the appendicular-umbilical fistula.[20,36]

Limitation

The congenital appendico-umbilical fistula is a rare clinical entity and only n = 23 cases were published during the past 100 years. Due to the paucity of published cases/available literature, the exact cause/embryogenesis is not well understood. There is no uniformity in describing the anomaly.

Various terminology used to describe this congenital anomaly are appendico-umbilical fistula, appendicoumbilical fistula, appendiceal-umbilical fistula, appendiculo-umbilical fistula and appendicular-umbilical fistula.[12,14,19-21,23-26,34,36]

Conclusion

PVID connected to the vermiform appendix and presented as a congenital appendico-umbilical fistula is a rare clinical entity. Most of the cases were detected and surgically treated during infancy. An appendicectomy and resection of the persistent duct/fistulous duct was the standard surgical therapy offered for the appendico-umbilical fistula. Due to the paucity of literature, the exact aetiology/embryological explanation is not clearly understood.

Informed consent

Review article: Informed consent not required for this manuscript.

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Conflicts of interest

There are no conflicts of interest.

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