

ORIGINAL RESEARCH

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Hymen-saving hymenotomy of imperforate hymen in neonates and adolescents: tertiary medical center experience

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Abstract

Background: Imperforate hymen (IH) is the most common obstructive anomaly of female genital tract. Hymen saving has a great importance for both cultural and religious reasons. Conventional surgical treatment for this condition is cruciate incision hymenotomy or hymen-saving central annular hymenotomy (HSCAH). This study aimed to review presentations and management of neonate and adolescents with IH at one tertiary hospital over 12 year's period. The aim also was to stress upon importance of hymen saving to our Muslim community and improve knowledge, enable targeted education, and reduce unnecessary tests and diagnostic delay for this condition.

Methods: Twenty-five patients with IH were diagnosed and treated in this retrospective cohort study over 12 years, from January 2010 till December 2021. Demographic characteristics of the patients, their symptoms and signs, investigations, type of operations, recurrence, and infections were recorded and analyzed.

Results: Saudi accounted for the majority of IH cases (88%). There were fifteen adolescents, and ten were neonates. Bulging imperforate hymen, pelvic/abdominopelvic mass, constipation, acute urine retention, abdominal discomfort, and lower abdominal distension were among the signs and symptoms. All 25 patients had HSCAH, 21 of whom had primary HSCAH and four of whom had secondary HSCAH due to recurrence. To avoid recurrence, HSCAH was performed with suturing of the inner vaginal mucosa to the exterior vestibular mucosa.

Conclusion: Early diagnosis of imperforate hymen for neonate and adolescent girls and prompt definitive HSCAH could have positive impact on treatment outcome and decrease complications and fulfill integrity of hymen as virginity of high importance in our Muslim community and other similar culture, where hymen-sparing procedure is preferred by most of these patients and families.

Keywords: Adolescent, Hematometrocolpos, Imperforate hymen, Hymen saving, Infection, Neonate, Surgical correction, Recurrence, Urine retention, Virginity

Background

Imperforate hymen (IH) is the commonest obstructive congenital abnormality of the female genital tract with an incidence between 0.01% and 0.05% in newborns [1]. One in 2000 females, the hymen fails to perforate during

intrauterine life leading to formation of hematometrocolpos post pubertal [2]. Imperforate hymen usually an isolated anomaly, but it should be differentiated from other malformations in the genitourinary tract as vaginal or uterine septum, atresia, renal agenesis, duplicated ureter, anorectal, and vertebral abnormalities [3].

Clinical presentation of IH can occur in infancy with inter-labial swelling and mucocolpos or, more commonly, in adolescent girls presenting with primary amenorrhea, a bulging hymenal membrane, cyclical abdominal pain,

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and/or a pelvic mass. It is usually asymptomatic until the age of 11–15 years (post-puberty) [4], rarely diagnosed in the neonatal period [5]. A review of the literature demonstrates that there are numerous case reports of “rare” presentations, as well as other presenting symptoms including urinary retention [4, 6], acute kidney injury, constipation, tenesmus [7], back pain [8] and dyspareunia [9], sciatica, and lumbar radiculopathy [10]. Late discovery of imperforate hymen may lead to pain, retrograde menstruation, infection, hydronephrosis, hemoperitoneum, urine retention, endometriosis, and infertility [5]. By inspection of the vaginal introitus, IH can easily be recognized by visualizing the bulging, imperforated hymen. Ultrasound (US) evaluation usually shows hematometrocolpos, which further confirms the diagnosis [7].

Simple incision of IH with loss of virginity lead to social stigma and emotions of guilt as this conventional surgical procedure is frequently misunderstood by religious and orthodox cultures, resulting in emotional, psychological, and even bodily harm [11]. The preservation of hymenal tissue may have a role in the perception of genital “integrity” and “completeness.” As a result, anyone with an imperforate hymen may benefit from a simple, low-risk surgical technique that preserves hymenal tissue [12].

This study aimed to review presentations and management of 25 patients with imperforate hymen whom been operated by HSCAH during neonatal and adolescent period, to stress that HSCAH have positive impact on treatment outcome and decrease complications and fulfill integrity of hymen as the virginity of high importance in our Muslim community and other similar culture. The aim also was to improve knowledge, enable targeted education, reduce unnecessary delay for this condition, and delineate more on the neonatal menstruation and its consequences.

Methods

This retrospective cohort study included 25 females diagnosed with imperforate hymen during neonatal and adolescent period and treated with HSCAH, 21 as primary procedure and 4 as secondary reoperation post previous cruciate hymenotomy, between January 2010 and December 2021. Females with other forms of menstrual irregularities or genital tract anomalies were excluded from the study. The research was conducted at Obstetrics and Gynecology Department, adolescent gynecology section and pediatric surgery section, at King Abdulaziz University hospital, Jeddah, Saudi Arabia.

Demographic and clinical details of the patients were gathered from both electronic medical records and written case records. The collected data included the demographic characteristics and their presenting symptoms and signs as well as investigations, type of operation

done, recurrence, and occurrence of infections (Table 1). All patients had a genital checkup before being diagnosed with imperforate hymen. Neonate diagnosis was made by vaginal examination at birth. Adolescent diagnosis was made by typical history that included cyclic monthly painful episodes without concomitant menstrual flow, lower abdomen pain, abdominal mass, and urine retention. Visual assessment of a completely closed hymenal structure, bulging of the hymen out of the vagina, and transabdominal and/or transrectal ultrasonographic detection of hematocolpos were used to diagnosis imperforate hymen. Urinary retention was also diagnosed via ultrasonography and treated initially with a urinary catheter. Under general anesthesia, patients were put in the lithotomy position. A central hymenal large pore needle puncture was made to aspirate the retained vaginal content; then, central hymenal sufficient annular membrane marked and excision was done while protecting the intact peripheral hymenal membrane ring; then, suturing of the inner vaginal mucosa to the external vestibular mucosa to maintain patency and prevent recurrence by complete epithelialization of edge of the hymenal ring was done (Figs. 1C and 2C, F). Suprapubic manual pressure was used to drain blood from the uterus and vagina. Foley catheters were used in patients with acute urine retention. Patients were admitted to day care unit with an average stay of 6–8 h for observation before being discharged. All patients’ neonate or adolescents had optimal period of follow-up to re-evaluate the complete healing without cross healing or stenosis; all were seen 1 week, then monthly for 6 months, and then annually for 2 years.

Statistical analysis

Data analysis was conducted using the Statistical Package for the Social Sciences, Version 20 (IBM Corp., Armonk, New York, USA). Results were presented as frequencies and percentages.

Results

Most of cases were Saudi (88%). Fifteen cases (60%) were diagnosed as adolescent imperforate hymen and 10 (40%) as neonate imperforated hymen. Manifestations were mostly bulging imperforate hymen (56%), pelvic/abdominopelvic mass (40%), constipation (40%), acute urine retention (28%), abdominal pain (28%), and lower abdominal distension (16%); operative procedures were HSCAH in all the 25 patients; of them, 21 patient (84%) made primary HSCAH, and the remaining 4 patients (16%) made secondary HSCAH as they previously had cruciate incision in other hospitals. Those 4 patients presented with recurrence hymenal stenosis with purulent discharge in 3 patients (12%) and complete menses obstruction in one patient (4%). Vaginal discharge was

Table 1 Characteristics of patients ($n=25$)

Characteristics	Frequency (%)
Nationality	
Saudi	22 (88%)
Sudanese	1 (4%)
Indian	1 (4%)
Yemeni	1 (4%)
Age category at diagnosis	
Neonates	10 (40.0%)
Adolescent	15 (60.0%)
Manifestations/presentations	
Bulging of imperforated hymen	14 (56.0%)
<i>Neonates</i>	4 (16.0%)
<i>Adolescents</i>	10 (40.0%)
Acute urine retention (all adolescence)	7 (28.0%)
Pelvic /Abdominopelvic mass (all adolescence)	10 (40.0%)
Abdominal pain (all adolescence)	7 (28.0%)
Constipation (all adolescence)	10 (40.0%)
Lower Abdominal distension (all adolescence)	4 (16.0%)
Recurrence stenosis with purulent discharge (all adolescence)	3 (12.0%)
Recurrence with amenorrhea (adolescence)	1 (4.0%)
Discharge per vagina	
Chocolate material	13 (52.0%)
<i>Neonates</i>	1 (4.0%)
<i>Adolescents</i>	12 (48.0%)
White mucous (all neonates)	9 (36.0%)
Purulent (all adolescents)	3 (12.0%)
Interventions	
Foley catheter as initial measure upon acute urinary retention	7 (28.0%)
Definitive surgical treatment	
Hymen-saving central annular hymenotomy	25 (100.0%)
Primary hymen-saving central annular hymenotomy	21 (84.0%)
Secondary hymen-saving central annular hymenotomy after recurrence	4 (16.0%)
Follow-up	
Yes	23 (92.0%)
Lost	2 (8.0%)
Menstrual history	
Regular	13 (52.0%)
Not yet	10 (40.0%)
Not known	2 (8.0%)

either chocolate material (52%), white mucous (36%), or purulent (12%); HSCAH was done with suturing of the inner vaginal mucosa to external vestibular mucosa to prevent recurrence. Menstrual history was regular in 13 (52%) cases, while 10 cases (40%) did not reach puberty and 2 cases (8%) lost follow-up. None of the patients had recurrence after HSCAH over the period of 2 years follow-up. Hymen-saving central annular hymenotomy in adolescent was shown in Fig. 1. Neonatal IH was shown

in Fig. 2. Adolescent with recurrent IH with purulent discharge was shown in Fig. 3.

Discussion

Imperforate hymen is the most common genital tract malformation in females with an incidence between 0.01% and 0.05% in newborns [1]. The exact incidence and prevalence remained uncertain. In this study, during 12 year's period, only 25 cases were managed successfully

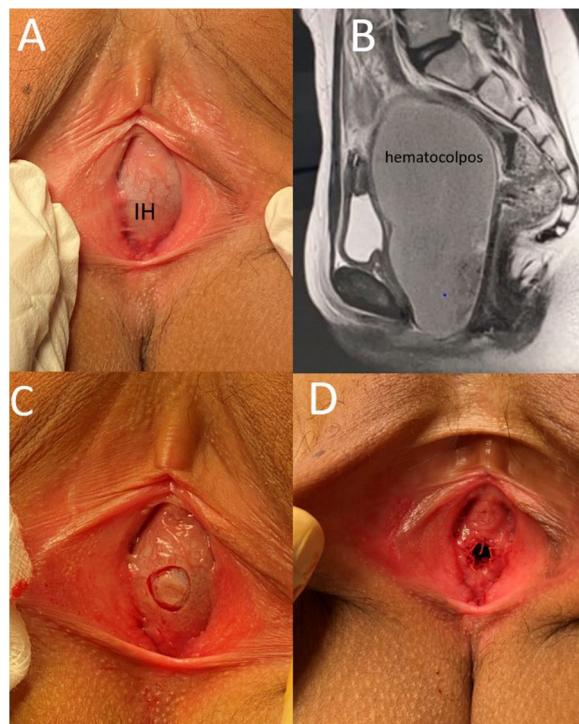


Fig. 1 Hymen-saving central annular hymenotomy (HSCAH) in adolescent. **A** Imperforated hymen (IH) with Valsalva maneuver showing increased bulging of imperforate hymen membrane. **B** Magnetic resonance image (MRI) showing massive hematocolpos occupying the pelvic cavity. **C** Demarcation of central annular incision. **D** Completed primary central annular excision of the hymenal membrane then suturing of the inner vaginal mucosa to external vestibular mucosa

with HSCAH. McCann et al. reported an incidence up to 1.2% in a descriptive study [13]. IH is a sporadic condition [14]. It is, however, occasionally linked to the Bardet-Biedl or McKusick-Kaufman syndromes [14] and reported in non-syndrome families [15]. Patients are typically diagnosed during the neonatal period or commonly at puberty. Because of recent advancements and extensive use of prenatal ultrasonography, the condition can be diagnosed prenatally [16].

IH management needs proper understanding and interpretation with consideration of the age, manifestations at presentation, and associated risks. Pressure effects had two categories: firstly internal pressure effects on the genital tract and secondly the external pressure effects on other structures. The effect of vaginal obstruction leads to increase intraluminal pressure, dilatation, and damage of genital tract, when further neglected and in accordance of nature of contained material may lead to retrograde menstruation and endometriosis or ascending infection with pelvic inflammatory diseases or acute abdomen caused by rupture of hematosalpinx and

hemoperitoneum [17]. The nature of vaginal content had significant meaning in particular the chocolate material. Fifty-two percent of our patients, one neonate and twelve adolescents, were discharging chocolate material upon hymenotomy, 36% neonates discharged white mucous, while 12% were adolescents presented with recurrence with hymenal stenosis with purulent discharge due to stasis and improper drainage of vaginal secretion and menstruation.

The serious long-term morbidity of IH and other genital tract outflow obstruction with accumulated secreted mucous and menstrual blood is retrograde menstruation and endometriosis which starts manifesting in adolescents at menarche or in neonatal period in phenomenon known as neonatal menstruation or neonatal uterine bleedings (NUB) leading to what is called early onset endometriosis (EOE) caused by early neonatal migration of menstrual material into peritoneum [18]. Neonatal uterine bleedings, which mostly observed in first week, is an ignored sign of EOE. Prevalence of NUB varies considerably with overt which ranged from 3.3 to 53.8% and occult from 25.4 to 96.7%. Future research should focus on prospective cohort studies in order to attempt to correlate NUB cases to EOE. Bianchi et al. [19] elaborated more on the hypothesis of neonatal menstruation as a cause of increased risk of EOE indicating the genital tract outflow obstruction with patent fallopian tubes carries significant increased risk of endometriosis, suggesting that in children and young adolescents, endometriosis may originate from retrograde uterine bleeding soon after birth as endometrial cells shed during NUB remain dormant till thelarche when the rising estrogen allow their proliferation to establish the ectopic lesions. Both studies stated that NUB is not “a perfectly physiological phenomenon of any clinical relevance”; on the contrary, it is an important sign of fetal distress and cause of early onset of endometriosis. On the other hand, the external pressure effects on other system structures related to intra-pelvic mass of the distended genital tract inside the limited none expandable bony pelvic cavity are causing pressure effects and obstructive symptoms of urinary and low bowel [20].

In this study, manifestations were mostly bulging from the imperforated hymen followed by acute urine retention, abdominal pain, lower abdominal distension, pelvic/abdominopelvic mass, and constipation. The girls with IH usually remain clinically quiescent until puberty age. When the girl starts to menstruate, they are with delayed diagnosis [21]. The clinical presentation can be primary amenorrhea, recurrent periodic abdominal pain [22, 23], constipation [10], acute urinary retention, and dysuria that occur in up to 58% of hematocolpos [24]. Acute urinary retention occurred in 7 (28%) cases in this study. In

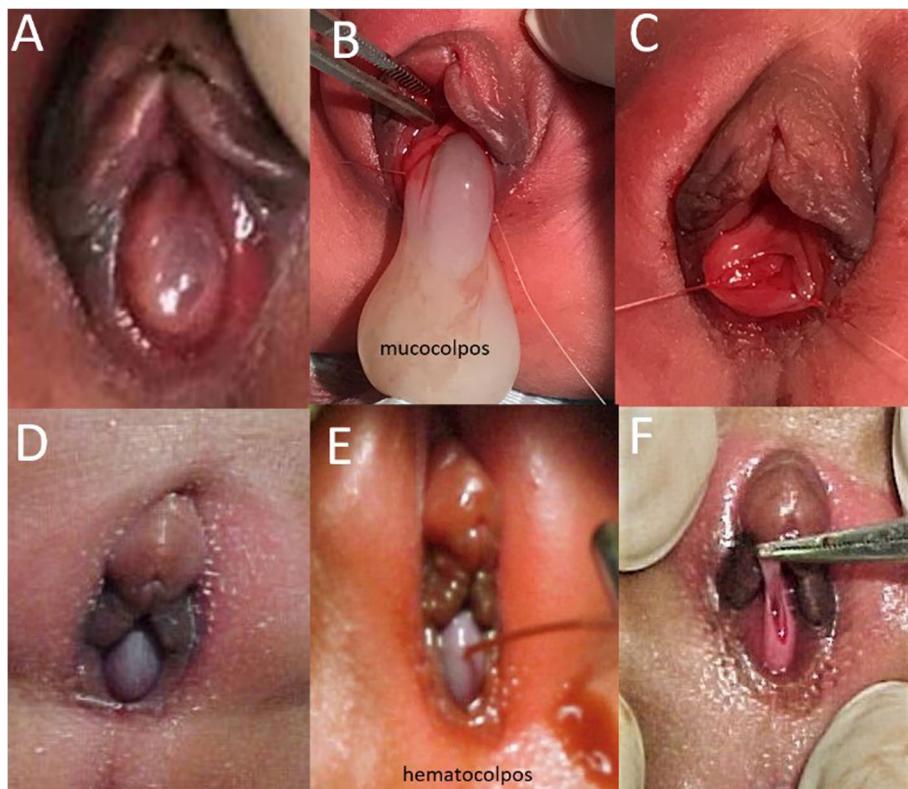


Fig. 2 Neonate imperforate hymen (IH). **A** White bulging IH pre-incision. **B** Drained thick white mucous upon hymenotomy. **C** Completed primary HSCAH. **D** Blue bulging IH pre incision. **E** Drained retained neonatal menstruation blood upon hymenotomy showing the high intraluminal genital pressure. **F** Primary hymen-saving central annular hymenotomy (HSCAH)



Fig. 3 Adolescent imperforate hymen (IH). **A** IH with the scar of stenotic previous incision seen with tiny opening that shows pus dripping. **B** Drained pus from vagina. **C** completed secondary HSCAH

a study on the causes of acute urinary retention in children, the incidence of imperforate hymen was found to be 10.7% [25]. Therefore, emergency physicians should

have a high index of suspicion of imperforate hymen as a cause for pubescent girls presenting with acute retention of urine.

On physical examination, IH can be diagnosed at any age by inspection of the external genitalia. Lui et al. [26] reported that classical findings are lower abdominal tenderness with a pelvic mass, associated with a bluish bulging mass through the introitus. Digital rectal examination may reveal an extrinsic mass corresponding to the vagina. The girl usually has attained Tanner stage 3 pubertal development with the absence of menarche [27].

Imaging may be necessary in some cases; in others, we utilized computer tomography (CT) and ultrasound which showed a fluid filled mass posterior to the bladder that might compress the bladder additionally magnetic resonance imaging (MRI) and transabdominal, or transperineally and transrectal US can differentiate IH from Mullerian malformations such as Mullerian agenesis, obstructing vaginal septa, and associated renal malformation as renal agenesis or duplication of the renal system [6, 28]. The differential diagnoses of a pubescent girl presenting with primary amenorrhea, lower abdominal pain, and a pelvic mass include transverse vaginal septum, longitudinal vaginal septum, vaginal agenesis, and cervical atresia [29]. IH can be differentiated from low type transverse vaginal septum by Valsalva maneuver, in which bulging of the hymen should occur with IH but not with septate vagina we had; this hymenal bulge was evident in the majority of our patients on Valsalva maneuver or applying pressure on the abdomen in adolescents and on careful observation of crying in neonates.

The exact function of the hymen is not known. It may act as a barrier to infections during prepubertal period in which vaginal self-defense mechanisms are not yet fully developed. It may be an example of exaptation that involves positive selection of women with intact hymen despite an embryologic remnant [24]. Although an intact hymen is generally mentioned in connection with Islam, actually, desire for virginity is also seen in many non-Islamic cultures [24, 30]. In Western countries, as a part of practices of culturally sensitive health care or personalized treatment alternatives involving cosmetic and personal demands, conservative management might be an alternative surgical option for all patients [30]. So, as virginity is cherished by many religions, cultures, and families, in this report, we describe an annular central hymenal membrane excision with preservation of hymenal ring of the membrane peripherally with suturing of the inner vaginal mucosa to external vestibular mucosa in order to keep the hymen intact and of sufficient caliber to drain the vaginal secretions and menstruation after the procedure.

We obtained a favorable surgical outcome in all 25 patients in our research, with 21 instances (84%) performed primarily by HSCAH and 4 cases (16%) operated by secondary HSCAH due to recurrence after prior

cruciate incision, all without recurrent obstruction or stenosis throughout long-term follow-up.

Alternative hymen-saving surgery had also been proposed. Acar et al. [31] described a procedure in which the hymenal membrane was opened with a knife in a 0.5-cm central oval incision, and then an inflated 16F Foley catheter was inserted and remained in place for 2 weeks to avoid re-adhesion of the margins, but maintaining a catheter in a cavity filled with retained menses for an extended amount of time is extremely dangerous and can lead to pelvic inflammatory disorders [32]. Furthermore, using a scalpel to make an incision can result in an irregularly shaped opening [33]. Darwish [34] reported hymen-saving approach in 36 cases; he performed a circle hymenotomy with a 10-mm laparoscopic trocar tip and sleeve to produce a new annular hymen, resulting in wide hymenal aperture while preserving the annular hymen, but this approach is not applicable in neonates and young adolescents with small-size hymenal membrane.

Surgical treatment of imperforate hymen has been reported in a variety of ways in the literature. The surgical drainage of the trapped blood via a hymenotomy, in which a vertical, cross, or T-shaped incision is created in the hymenal membrane, followed by suturing of the margins to maintain patency and prevent restenosis, is the standard treatment for this condition [35]. Excision of hymenal tissue and superfluous mucosa is usually required to address clinically severe hymenal abnormalities [32]. However, because of the risk of ascending infection and sepsis, simple incision and drainage of hematocolpos in a patient with an IH should be avoided [36]. Other study suggested that thicker hymenal membranes be surgically resected, while thinner membranes be treated with a vertical incision [32]. Fixing the vaginal wall to the introitus also helps to prevent restenosis and post-operative problems [32]. Lee et al. [37] reported that majority of IH patients (83.5%) were treated surgically, with the most common procedures being hymenotomy (35.2%) and hymenectomy (36.4%), with no significant differences in outcomes. However, previous research indicated possibility of recurrent restenosis following a hymenectomy [38]. In this study, 4 (16%) adolescents presented to our hospital with recurrence after previous cruciate hymenotomy, 3 (12%) of them suffered from secondary infections, and one patient presented with complete obstruction; meanwhile, all cases that reach puberty had regular menstrual cycle after benign re-operation with HSCAH. Liang et al. [39] reported that women who had their imperforate hymen surgically corrected had irregular menstrual cycles in the majority of cases. The majority of these women were concerned about their future fertility, and six of them had dysmenorrhea. Two of the 11 women who started having sexual

relations were pregnant, and none of them complained of sexual problems. Women with imperforate hymen were reported to have much less cryptomenorrhea after hymenectomy, as well as improved micturition and stooling.

Conclusions

IH should be suspected and diagnosed in neonates with local hymenal bulge and in adolescent girls with primary amenorrhea presenting with lower abdominal pain, acute urinary retention, pelvic mass, constipation, or low back pain. Physical examination and bedside ultrasonography are the key to correct management and prevention of complications. Early diagnosis of imperforate hymen in adolescents and thorough examination of the neonate at birth is of paramount importance and associated with good prognosis. Virginity-sparing surgery constitutes a good treatment option because of cultural and religious reasons. In this study, we presented our experience in the management of 25 patients with IH by HSCAH with satisfactory surgical outcome with maintaining vaginal patency and preserving the virginity. The key of success to perform the HSCAH without recurrence is sufficient caliber central annular hymenal membrane excision then suturing of the inner vaginal mucosa to external vestibular mucosa that result in complete epithelialization of the hymenal ring.

Abbreviations

CT: Computer tomography; EOE: Early onset endometriosis; HSCAH: Hymen-saving central annular hymenotomy; IH: Imperforate hymen; MRI: Magnetic resonance imaging; NUB: Neonatal uterine bleedings; US: Ultrasound.

Acknowledgements

Not applicable

Authors' contributions

All authors contributed equally in writing and revision of manuscript. E.A.A. contributed specifically in the patient selection, statistical analysis, operative technique, and writing of the manuscript. M.K. contributed in designing the manuscript, selection cases, collecting of the data, shared operative technique, and in writing the manuscript. All authors have read and approved the manuscript for publications.

Funding

None.

Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate

Obtained from Unit of Biomedical Ethics Research Committee (Reference No 411–20).

Consent for publication

Agree to consent for publication.

Competing interests

The authors declare no competing interests.

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Received: 5 March 2022 Accepted: 15 June 2022

Published online: 01 August 2022

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