



Oropharyngolaryngeal manifestations of Kaposi's sarcoma with airway obstruction and its management in patients with HIV/AIDS

Manifestaciones orofaringolaríngicas del sarcoma de Kaposi con obstrucción de las vías respiratorias y su manejo en pacientes con VIH/SIDA

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ABSTRACT

Introduction: Kaposi's sarcoma is one of the manifestations in HIV/AIDS patients which is found in the oropharyngolaryngeal region. This can be life-threatening because it can cause airway obstruction. Therefore, proper management is needed in handling HIV/AIDS patients with oropharyngolaryngeal Kaposi's sarcoma manifestations.

Objective: To characterize clinical characteristics, airway interventions, and clinical outcomes of oropharyngolaryngeal Kaposi's sarcoma in HIV-positive patients with airway obstruction.

Methods: This study was a systematic review followed PRISMA 2020 guidelines. Scopus, PubMed, Web of Science, and Cochrane Library were searched systematically. The JBI checklist was used for the quality assessment.

Results: Twenty-four studies were finally included. Patients were dominated by male with an adult age group. Symptoms that often appear include dyspnea, hoarseness, and stridor, with Kaposi's sarcoma sites most commonly found in the supraglottis. In airway management, tracheotomy proved to be the most effective in the airway obstruction management.

Conclusions: This review concludes that tracheotomy is an effective option for airway management in patients with oropharyngolaryngeal Kaposi's sarcoma. However, most patients' prognosis is poor due to the advanced stage of HIV infection and the accompanying complications.

Keywords: AIDS; airway obstruction, HIV; larynx; oropharynx; Kaposi's sarcoma.

RESUMEN

Introducción: El sarcoma de Kaposi es una de las manifestaciones en pacientes con VIH/SIDA que se encuentra en la región orofaringolaríngea. Esto puede ser potencialmente mortal, ya que puede causar obstrucción de las vías respiratorias. Por lo tanto, es necesario un manejo adecuado en pacientes con VIH/SIDA con manifestaciones de sarcoma de Kaposi orofaringolaríngeo.

Objetivo: Caracterizar las características clínicas, las intervenciones de las vías respiratorias y los resultados clínicos del sarcoma de Kaposi orofaringolaríngeo en pacientes VIH positivos con obstrucción de las vías respiratorias.



Métodos: Este estudio fue una revisión sistemática que siguió las pautas PRISMA 2020. Se realizaron búsquedas sistemáticas en Scopus, PubMed, Web of Science, y Cochrane Library. Para la evaluación de calidad se utilizó la lista de verificación del JBI.

Resultados: Finalmente se incluyeron 24 estudios. Los pacientes fueron predominantemente varones con un grupo de edad adulta. Los síntomas que aparecen con frecuencia incluyen disnea, ronquera, y estridor, y los sitios de sarcoma de Kaposi se encuentran más comúnmente en la supraglotis. En el manejo de la vía aérea, la traqueotomía demostró ser la más efectiva en el manejo de la obstrucción de la vía aérea.

Conclusiones: Esta revisión concluye que la traqueotomía es una opción eficaz para el manejo de las vías respiratorias en pacientes con sarcoma de Kaposi orofaringolaríngeo. Sin embargo, el pronóstico de la mayoría de los pacientes es desfavorable debido al estadio avanzado de la infección por VIH y las complicaciones asociadas.

Palabras clave: SIDA; obstrucción de las vías respiratorias, VIH, laringe, orofaringe, sarcoma de Kaposi.

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INTRODUCTION

According to UNAIDS/WHO estimates, the prevalence of individuals infected with HIV has reached 85.6 million, with approximately 40.4 million deaths attributed to HIV. By the end of 2022, around 39 million people worldwide were living with HIV, and about 630,000 deaths were caused by HIV or AIDS-related diseases.^(1,2)

HIV-infected patients often experience complications and manifestations in the form of diseases related to HIV infection, including complications in the head and neck region.^(3,4) Some manifestations of HIV/AIDS that can be found in the oropharyngolaryngeal area, which includes



the oral cavity, pharynx and larynx, are oral and esophageal candidiasis, periodontal diseases, Kaposi's sarcoma (KS), recurrent aphthous ulcers, hairy leukoplakia, oral melanotic hyperpigmentation, oral and perioral warts, non-Hodgkin's lymphoma, and squamous cell carcinoma.^(5,6,7,8,9) A systematic review found KS to be the most common manifestation among patients with HIV.⁽⁵⁾

KS is a tumor characterized by excessive vascular proliferation and is caused by infection with the herpes virus associated with KS, also known as human herpesvirus 8 (HHV8).⁽¹⁰⁾ KS can be classified into four distinct epidemiological subtypes, namely classic, African endemic, immunosuppression-associated KS, and AIDS-associated KS.⁽¹¹⁾ In the general population, the estimated global incidence rate of KS is 0.39 per 100,000 population, with approximately 34,270 new cases reported in 2020. Furthermore, KS caused around 15,086 deaths in 2020, corresponding to a mortality rate of 0.18 per 100,000 population.⁽¹²⁾ Meanwhile, among individuals with HIV infection, the overall incidence of KS was 418.54 per 100,000 person-years.⁽¹³⁾

KS has clinical manifestations in the form of multiple lesions that may present as macules, nodules, plaques, or exophytic, pigmented, raised or flat lesions, which are painless and do not change color when pressed.⁽¹⁴⁾ Based on its anatomical location, KS can be found in various mucocutaneous sites, including uncommon areas such as the oropharynx and larynx.^(15,16) In the oropharyngolaryngeal region, symptoms of KS may include dyspnea, hoarseness, cough and dysphagia. Apart from that, stridor, which indicates airway obstruction, is also a symptom of oropharyngolaryngeal KS which can be life-threatening if emergency treatment is not promptly administered.⁽¹⁷⁾

The high incidence of KS as a manifestation among individuals with HIV infection is of particular concern, especially in cases involving the oropharyngolaryngeal region. This is due to the presence of KS masses that may cause airway obstruction, which can be life-threatening if not properly managed. Systematic review regarding KS in the oropharyngolarynx with airway obstruction remains limited; therefore, the objective of this study is to describe the clinical characteristics, airway interventions, and clinical outcomes of oropharyngolaryngeal Kaposi's sarcoma in HIV-



positive patients with airway obstruction from previous published studies using a systematic review method.

METHODS

Registration

This study used a systematic review method following the PRISMA 2020 guidelines.⁽¹⁸⁾ This systematic review has been registered in the PROSPERO with the registration number of CRD42024548844 (available from:

<https://www.crd.york.ac.uk/PROSPERO/view/CRD42024548844>).

Focused question

The focused question in this study was "What evidence is available regarding the clinical presentation, airway involvement, and management approaches of oropharyngolaryngeal KS with airway obstruction in patients with HIV/AIDS?" This question was formulated to guide a descriptive synthesis of available evidence from published article, acknowledging the limited quality and heterogeneity of the data. To address this, authors used the Population, Intervention, Comparison, and Outcomes (PICO) model, with P: patients with HIV/AIDS positive status, I: oropharyngolaryngeal manifestations of KS with airway obstruction, C: -, and O: manifestation and management characteristics.

Eligibility criteria

The authors applied inclusion and exclusion criteria to screen the articles. The inclusion criteria in this study were clinical research involving HIV/AIDS patients with manifestations of oropharyngolaryngeal KS, written in English, and available in full text. Meanwhile, the exclusion criteria used were reviews, editorials, and commentaries; research involving HIV-negative patients, and article for which the full text was unavailable. All articles published up to April 2024 were considered for inclusion, with no limitation on the year of publication.



Search strategy

Scopus, PubMed, Web of Science, and Cochrane Library were used to comprehensively literature search. To avoid articles published but not found in the previously mentioned databases, we also conducted literature search through other methods, such as citation searches. In the literature search, authors used the following main keywords: oral, oropharynx, oropharyngeal, pharynx, pharyngeal, larynx, laryngeal, Kaposi's sarcoma, HIV, AIDS, and airway obstruction. Search using these keywords combined with the Boolean operators “AND” and “OR”. The detailed queries used in each database is available in the [supplementary file](#) (Table S1). The literature search was performed during March to April 2024.

Quality assessment

All articles obtained and included in this systematic review were then subjected to quality assessment using the Joanna Briggs Institute (JBI) critical appraisal checklist. A single adapted JBI form was applied uniformly across all studies to ensure consistency, as most of the included articles were case reports with similar reporting structures. The checklist covered eight domains based on the JBI checklist for case reports, including patient characteristics, previous history, current clinical conditions, diagnosis methods, treatment/management, post-treatment conditions, side effects, and important learning. This process was carried out by two independent researchers (A.S. and E.W.D.) and validated by a senior researcher (T.S.). All discrepancies of opinion during this quality assessment were resolved carefully through in-depth discussion.

Data extraction

Two authors (A.S. and F.M.R.) independently read the full text of the included studies, then data extraction was conducted. Authors used a table to describe and summarize all important information from the articles, including the author of the article, country of research, study design, number of cases, gender, age, HIV/AIDS status, specific location of KS, symptoms, treatment that has been given, management carried out, and final clinical results. All data extracted was subsequently validated by one author (N.D.K.).



RESULTS

Study selection

Selection: 101 potentially eligible articles were obtained from database searches after removing duplicates and seven from citation searches. After screening the title and abstract, 41 studies were eliminated due to incompatibility with the research topic, remaining 60 studies remaining. At this stage, a more in-depth review of the articles obtained was carried out by applying previously determined inclusion and exclusion criteria. Of the 60 studies from the database search and seven studies from the citation search, 43 studies were excluded for several reasons, including the full-text was not available, the methodology was not suitable, the location of KS was not appropriate to the topic, the patient was HIV negative, HIV status was unknown, and no association with airway disorders or obstruction was found. Thus, the total articles included in this systematic review study were 24 articles with details of 23 case reports and one retrospective study (Fig. 1).

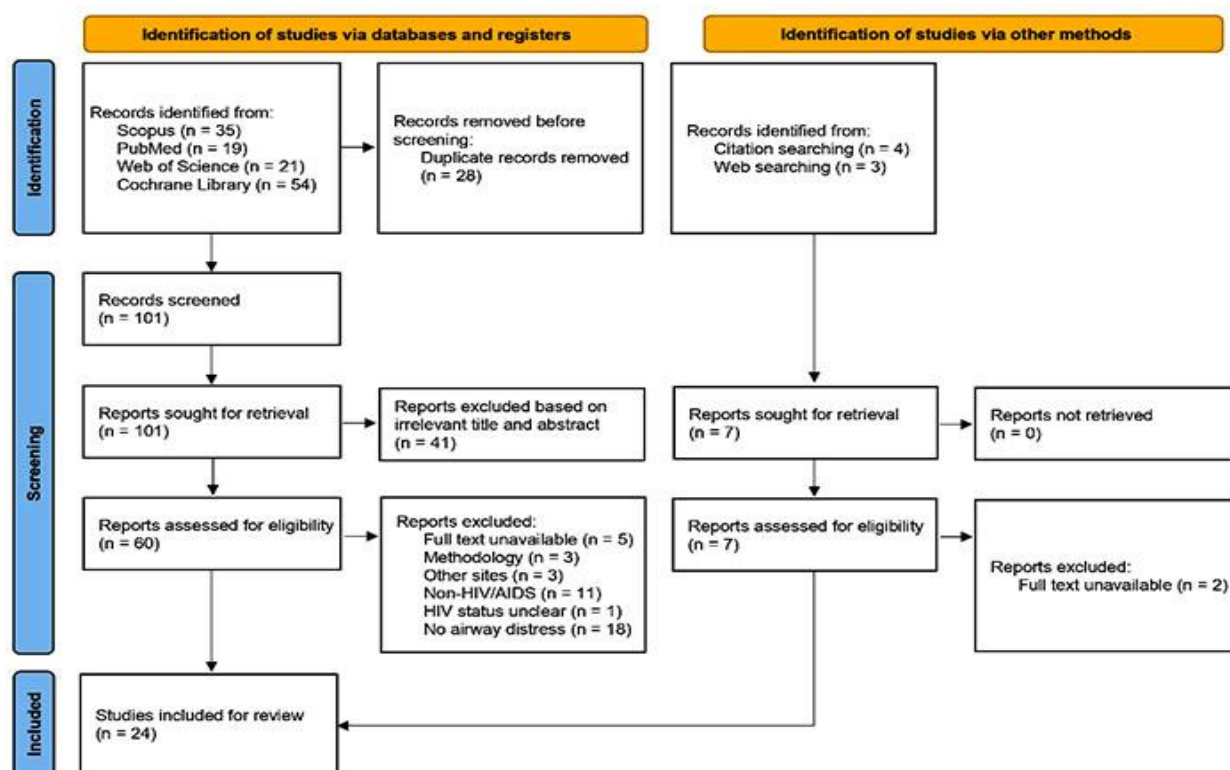


Fig. 1 – PRISMA flowchart.



Characteristics of included studies

This systematic review included twenty-four articles involving forty HIV/AIDS patients with manifestations of KS located in the oropharyngolarynx and symptoms of airway obstruction. A total of 11 studies were case report studies from USA,^(19,20,21,22,23,24,25,26,27,28,29,30) one case report from Spain,⁽³¹⁾ one case report from Switzerland,⁽³²⁾ one case report from UK,⁽³³⁾ one case report from Belgium,⁽³⁴⁾ one case report from Malaysia,⁽³⁵⁾ one case report from Portugal,⁽³⁶⁾ one case report from Tanzania,⁽³⁷⁾ one case report from South Africa,⁽³⁸⁾ one case report from Bhutan,⁽³⁹⁾ one case report from Japan,⁽⁴⁰⁾ one case report from China,⁽⁴¹⁾ and one retrospective study from UK.⁽⁴²⁾ A table summarizing study characteristics and summary results of all articles are available in the [supplementary file](#), table S2.

Patients characteristics

In this systematic review, 5% of patients were children and the remaining 95% of patients were adults. Meanwhile, according to gender, KS patients in patients with HIV infection and airway obstruction were dominated by 90% of male patients and the remaining 10% of patients were female. Based on the characteristics of the patients involved in this study, the most frequently found symptoms were dyspnea (22.86%), hoarseness (18.57%), stridor (15.71%), and dysphagia (14.29%). Meanwhile, symptoms that only a few patients experienced included cough (4.29%), sore throat (4.29%), lymphadenopathy (2.86%), weight loss (2.86%), and bleeding (2.86%). Other less common symptoms include hemoptysis, difficulty speaking, choking sensation, foul odor, hypersalivation, and unable to close mouth, each of which occurred in only 1.43%.

According to the anatomical site of KS patients, the most common sites were supraglottis (29.51%), glottis (18.03%), subglottis (11.48%), epiglottis (9.84%), and tongue (6.56%). Meanwhile, less common anatomical locations were the arytenoids, aryepiglottic folds, hard palate, soft palate, tonsils, and vocal cords, each occurring in 3.28%, and the gingiva and ventricular folds, each occurring in only 1.64%.

In the management of KS, chemotherapy was the most commonly performed (30%), followed by a combination of surgery and radiotherapy (17.5%), surgery and chemotherapy (15%),



radiotherapy (12.5%), and surgery alone (10%). While the rest was a combination of surgery, radiotherapy, and chemotherapy (5%), and a combination of radiotherapy and chemotherapy (2.5%). Finally, 7.5% of patients had not undergone KS treatment because the patient died from serious complications.

Regarding airway management, tracheotomy was the most common primary choice (73.9%), while other management such as steroids (8.7%), oxygen therapy (8.7%), nebulized adrenaline (4.3%), and intubation (4.3%) were rarely given to patients in treating airway obstruction in KS patients. Finally, the most frequently reported clinical outcomes by studies were died due to HIV/AIDS including other complications (55%). Meanwhile, patients who recovered or improved from airway obstruction symptoms were 27.5%. Meanwhile, the other 5% died due to airway obstruction (2.5%) and died of unknown causes (2.5%), and as many as 12.5% of patients did not have their outcomes or follow-up reported. All details regarding the characteristics of patients are available in the [supplementary file](#), table S3.

Study quality assessment

The quality of the included studies was then assessed using the JBI critical appraisal checklist adapted for case reports. The overall quality of the studies was generally low to moderate due to incomplete reporting of key methodological details. Among 24 included studies, 18 (75%) were rated as low risk of bias and 6 (25%) as moderate risk of bias. All studies provided patient details regarding previous history, current clinical conditions, treatment/management, and important learning outcomes, while none described side effects. One study failed to fully describe patient characteristics, and another did not clearly describe the diagnostic method. Furthermore, five studies failed to report post-treatment conditions.

The limited methodological rigor and heterogeneity of reporting reflects the descriptive nature of the available evidence, which primarily consists of case reports. This suggests that the findings in this study need to be interpreted with caution. Quality assessment results of included studies is available in the [supplementary file](#), table S4.



DISCUSSION

In this systematic review, KS patients with HIV/AIDS mostly occur in adult men. This is in accordance with the meta-analysis conducted in 71 different periods, where the results show that the incidence of KS in adults reached 539/100,000 population-years and in children it was 53/100,000 population-years. Meanwhile, according to sex, the homosexual male population dominates the incidence of KS in HIV/AIDS patients with an incidence of 1,397/100,000 and heterosexual men at 655/100,000, and in women at 172/100,000 population-years.⁽¹³⁾ Strengthened by research which stated that KS is dominated by men with a rate of 147/100,000 and in women it is 21/100,000. This number is higher in the 20-34 and 35-49 age groups.⁽⁴³⁾ Meanwhile, a meta-analysis also stated that KS in HIV sufferers generally occurs at around 40 years of age, whereas in the general population it is diagnosed at around 60 years of age, indicating that KS tends to occur at a younger age in patients with HIV compared to non-HIV patients.⁽¹¹⁾

The most common symptoms in patients of the included studies with oropharyngolaryngeal KS were dyspnea, hoarseness, stridor, and dysphagia. Meanwhile, the anatomical locations most frequently affected by oropharyngolaryngeal KS are the supraglottis, glottis, subglottis, epiglottis, and tongue. One patient may experience more than one symptom and anatomical location. Symptoms in patients involved in this study were dominated by symptoms suggestive of upper airway obstruction caused by a KS mass in the oropharyngolaryngeal region.

Airway obstruction, one of which is caused by the presence of a tumor mass, is divided into upper airway obstruction involving the oral or nasal region to the larynx, and lower airway obstruction, which affects the tracheobronchial area. Upper airway obstruction involving the oropharynx has symptoms including snoring, gurgling sounds, and swelling. At the base of the tongue and epiglottis, there will be symptoms such as dysphagia, stridor, and hypersalivation/drooling. Meanwhile, in the glottis area, symptoms such as stridor, dyspnea and changes in voice often appear.⁽⁴⁴⁾ In addition, patients with malignant conditions in the larynx or pharynx may also experience airway obstruction which usually has initial symptoms, including hoarseness or dysphagia.⁽⁴⁵⁾ *Costa L et al.*⁽⁴⁶⁾ stated that dyspnea and stridor are symptoms that are often found in patients who are given emergency airway management. These symptoms of upper airway



obstruction are often found in patients with oropharyngolaryngeal KS, where dyspnea, hoarseness, stridor, and dysphagia are the four most common symptoms found in patients involved in this systematic review.

In the management of airway obstruction, there is control management that can be carried out, such as administering high-flow oxygen, steroid therapy which reduces laryngeal edema and stridor, nebulized adrenaline to reduce symptoms of stridor, heliox, and continuous positive airway pressure. Meanwhile, management to secure the airway includes options such as fiberoptic intubation, videolaryngoscopy, tracheotomy, and general anesthesia with IV induction or inhalation, and cricothyroidotomy.⁽⁴⁴⁾ In this systematic review, 23 (57.5%) patients underwent airway management. Of the 23 patients, 73.9% of patients underwent tracheotomy, most of which were carried out urgently due to airway obstruction. The remainder, in the management of airway safety through control management, this systematic review found cases where patients were given steroids,^(19,40) oxygen therapy,^(26,37) and nebulized adrenaline.⁽³⁸⁾

On management to secure the airway, this systematic review found cases using steroids, oxygen therapy, and nebulized adrenaline. Steroids were given because there was an edematous lesion in the pharynx which triggered airway obstruction, and was stopped because the edema in the pharynx improved quickly,⁽⁴⁰⁾ as was the case in this case study.⁽¹⁹⁾ Steroid administration is indicated if the patient experiences edema or inflammation associated with potential airway obstruction. Meanwhile, nebulized adrenaline has a function as a vasoconstrictor which appears to be effective in reducing symptoms of stridor due to obstructive KS masses,⁽⁴⁴⁾ where this management was carried out in one of the patients in this systematic review but failed due to a KS mass on the tongue which manifested up to the oropharynx.⁽³⁸⁾

The main treatment for acute airway obstruction is transoral endotracheal intubation via laryngoscopy.⁽⁴⁷⁾ This was done successfully in one patient in this review,⁽³⁸⁾ but failed in one case of a patient who had a KS mass on the tongue that filled the oral cavity and oropharynx.⁽³⁷⁾ Furthermore, *Panda N et al.*⁽⁴⁷⁾ added that in patients with airway obstruction that does not allow endotracheal intubation, surgical tracheal intubation can be performed, including percutaneous or open tracheotomy and transtracheal needle ventilation. Emergency tracheotomy can be performed



in patients with acute airway obstruction who have difficulty intubating. This procedure is considered effective and safe with a low complication rate, and must be conducted immediately on the patient to prevent the patient's status from developing into a surgical emergency.⁽⁴⁶⁾ In this systematic review, of the 73.9% of patients who underwent tracheotomy, four patients underwent emergency tracheotomy.^(21,26,28,35) Apart from the cases of HIV/AIDS patients included in this systematic review, the tracheotomy procedure is also used in HIV/AIDS negative oropharyngolaryngeal KS patients, who also do not allow endotracheal intubation, as preventive efforts or management of airway obstruction.^(48,49,50)

Patients may have a better prognosis after emergency tracheotomy.⁽⁴⁶⁾ Of the 17 patients who underwent tracheotomy, 94.1% of patients had airway obstruction treated, while one patient died due to airway obstruction due to worsening of the patient's condition with clinical stage 4 HIV infection, oxygen saturation dropping to 40%, failed intubation, and blockage of the oropharynx by KS mass.⁽³⁷⁾ In conclusion, tracheotomy is the most effective management in treating the patient's airway in conditions where the patient experiences airway disturbance or obstruction caused by a malignancy, in this case KS in the oropharyngolaryngeal region. Endotracheal intubation can still be performed if possible, as in one case we encountered; however, because KS masses are fragile and can bleed, tracheotomy is an option to prevent aspiration from bleeding.

Clinical outcomes for patients showed that 27.5% of patients' condition improved and recovered, the remainder died due to worsening HIV infection and complications, airway obstruction, and died of unknown causes, as well as 12.5% of patient outcomes were not reported. However, in terms of the cause of patient death, more than half of the total patients involved in this systematic review had worsening HIV infection or complications related to HIV/AIDS. Meanwhile, one patient died due to airway obstruction caused by a large KS mass that blocked the oropharynx, and the cause was unknown for the rest and there were no clinical outcome reports.

Medical personnel must increase awareness of symptoms of airway obstruction in patients with oropharyngolaryngeal KS. Training in emergency management of airway obstruction, including the use of tracheotomy should be intensified. A multidisciplinary approach involving many related specialties is highly recommended for comprehensive management. In addition, regular monitoring



and follow-up to assess the patient's response to therapy and detect complications early should be carried out to ensure timely treatment adaptation. One of the instruments that medical personnel can use to monitor and detect patient deterioration is the Early Warning Score (EWS) instrument. This systematic review characterizes the clinical symptoms, airway interventions, and clinical outcomes of oropharyngolaryngeal KS with airway obstruction in people living with HIV/AIDS. The reviewed cases predominantly involved adult male patients presenting with dyspnea, hoarseness, stridor, and dysphagia. Lesions were most frequently located in the supraglottis, glottis, subglottis, epiglottis, and tongue areas. Airway management varied across the included studies depending on the severity of obstruction, with tracheotomy consistently reported as the most effective and immediate intervention for securing the airway in emergency situations. Despite these consistent clinical patterns, the evidence base remains limited to descriptive case reports. Consequently, the findings should be interpreted with caution, as comparative efficacy cannot be reliably established.

Further research can be carried out regarding multicenter prospective studies to collect more comprehensive data regarding the manifestations of oropharyngolaryngeal KS and its management in HIV/AIDS patients. The development of evidence-based clinical guidelines is urgently needed to support early detection, emergency treatment, and long-term therapy, especially in patients with airway disorders or obstruction. In addition, research into specific risk factors that increase the likelihood of airway obstruction may be warranted.

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Conflicts of interest

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Data availability

Supplementary file: Research results. PDF. Available from:

<https://revmedmilitar.sld.cu/index.php/mil/libraryFiles/downloadPublic/104>

