

Oral warts in an African female child; a rare pathology, diagnostic and therapeutic challenges in a resource limited setting: A case report.

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Abstract

Background: Warts are benign lesions that are dermatological manifestation of Human Papilloma viruses. They are unusual in infancy and early childhood with the lowest prevalence among children less than 6 years old, nevertheless the incidence increases as they approach school-age. The appearance of warts is determined by the type of virus and the location of the infection; ranging from common warts, palmer and planter warts to flat, filiform and genital warts. Oral warts however, are rare findings in adults and even more so in children and the diagnosis as well as management in resource poor settings is often challenging.

Case presentation: We report the case of an 8 year old black African girl with no relevant past history, who presented with a 6 month history on multiple painless oral lesions. She had no soreness, no fever nor any other relevant symptom. Oral examination revealed multiple hyperkeratotic, sharply margined, flat-topped papules covered by normal mucosa with no specific pattern of organization, lining the oral cavity excluding the soft palate. There were no signs of excoriations, swellings or other suspicious lesions on the face, hands, knees, genitalia and perianal areas. The rest of the examination was unremarkable. A diagnosis of oral warts was made and was managed by electrocauterization and subsequent mouth washing with salt and warm water.

Conclusion: Oral warts are a rare pathology especially in children. HPV infection is the most like aetiology. Investigating the origin is often a daunting task as well as method of management. Spoon sharing is a possible risk factor for oral warts especially in children and electrocauterization is an effective and less costly method of management in pediatric cases. We therefore recommend thorough oral exam for all individuals at risk and electrocautery for identified cases of oral warts especially in limited resource settings.

Keywords: Oral warts, Human papilloma virus, Children, Electrocauterization, Limited resource settings.

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Background

Warts are benign epidermal lesions that are dermatological manifestations of Human papilloma virus (HPV) infection [1]. The incidence and prevalence of warts is not well known, however 2 large population based studies showed prevalence rates of 0.84 percent in the United States and 12.9 percent in Russia [2]. In the United Kingdom, studies show a 30% prevalence in children [3]. Warts are unusual in infancy and early childhood but incidence increases as

they approach school-age to peak at 12-16 years [4]. As at the time of presentation of this case report, the prevalence of warts in Africa is not known though studies suggest that common warts are twice as frequent in whites as in blacks and Asians [5]. Trauma and maceration facilitates epidermal inoculation of HPV as such it can be transmitted by indirect contact with contaminated objects such as fomites but also by direct skin-to-skin contact such as sexually, perinatally or by autoinoculation as is mostly the case with pediatric patients [6-9]. Autoinoculation will

explain dissemination to different site [10]. Due to these routes of transmission, warts most commonly affect the hands, feet, face and the genitalia. Oral warts are a rare occurrence in adults and even more so in children given that its principal risk factor is oral sex thus making intraoral warts a very unusual finding in children [11-13]. The appearance of warts is determined by the type of virus and the location of the infection [2]. Over 189 distinct Human papillomavirus types had been described, of which 151 were found in humans Types 1, 2, 3, 4, 10, 27 and 57 are most often implicated in the etiology of cutaneous warts [3,14]. The presentations of warts are thus wide and varied, ranging from common warts, palmer and planter warts to flat, filiform and genital warts. The initial diagnosis is made after a thorough history and physical exam [15,16]. Investigations such as biopsy for histopathology or HPV DNA are not a common practice, as such the diagnosis is therefore clinical [16]. There are multiple recommended treatment modalities available but none is always effective. It is either by destructive methods (excision, cautery, cryotherapy, liquid nitrogen) or use of topical or injected agents [3,17-21]. The method of treatment will depend on the peculiarity of the patient but it is often recommended to begin with least painful, least expensive and least time consuming method [22]. As oral warts are a rare finding in adults and even more so in children, information about the occurrence of this pathology is scarce and its diagnosis as well as management in resource poor settings is often challenging. Here, we present the case of oral warts in an 8 year old female and the approach to her management.

Case Presentation

This is the case of an 8 year old black African girl, with a 6month history of multiple painless oral lesions. The lesions began on the tongue initially and were few but progressively increased in number and size to occupy most of the inner surface of the mouth. Initial consultation in a clinic resulted in a diagnosis of oral candidiasis which was managed with fluconazole which the patient took with no improvement; this then prompted a second consultation at our hospital. There was no associated odynophagia, no dysphagia, no soreness, no change in voice, no hematemesis and no loss of appetite, no diarrhea and no vomiting. She denies any contact with a person with warts and had no relative with warts. She occasionally plays barefooted but neither swims nor has access to community showers. In addition, the younger brother had started developing similar mouth lesions and here was the notion of spoon sharing during meals but not tooth brushes. The Mother and child denied any history of sexual abuse or exposure. The mother was HIV negative as of recent records and non-diabetic as well, but the child's HIV status was unknown. She had no history of medical illness or any other relevant past history.

On examination, she was stable and appropriate for her age. She was afebrile with a temperature of 37°C, pulse of 90beats per minute, blood pressure od 124/70 mm Hg

and respiratory rate of 18 breaths/min. She had a pink conjunctivae and anicteric sclerae. There were no signs of excoriations, swellings or other suspicious lesions on the face, fingers, hands, knees, genitalia and perianal areas. Oral examination revealed multiple hyperkeratotic, sharply marginated, flat-topped papules covered by normal mucosa with no specific pattern of organization, lining the oral cavity excluding the soft palate (Figure 1). Lesions were non-tender, pharynx was not erythematous and tonsils were not enlarged. There were no enlarged cervical, submandibular, sub mental and peri-auricular lymph nodes. Thoracic and abdominal examinations were not contributory. The rest of the examination was unremarkable.

Investigations were done with their Results as Follows

HIV serology (Determine): Negative

Complete blood count: White blood cell count of $4.8 \times 10^3/\mu\text{l}$, Lymphocyte of $2.3 \times 10^3/\mu\text{l}$, granulocyte of $2.0 \times 10^3/\mu\text{l}$, Haemoglobin (Hb) 11.1 g/dl, Mean Corpuscular Volume (MCV) of 92 fl, Mean Corpuscular Hemoglobin Concentration (MCHC) of 32 g/dl Haematocrit of 34.5% and platelet count of $261 \times 10^3/\mu\text{l}$.

HPV DNA by PCR: It was not done because the test was very expensive (100,000 CFA) and samples can only be sent out of the country for analysis which is a drawback in resource limited settings like ours.

No biopsy was taken for cytopathology analysis.

Based on all of the above, she was diagnosed of oral warts most likely caused by Human Papilloma Virus. After receiving consent from the child's mother, the dental surgeon managed the oral wart lesions by electrocauterization under 2% lidocaine local anesthesia (Figures 2a and 2b).

She was then sent home on the same day and asked to continue regular mouth rinsing with salt and warm water. She was booked for a follow-up visit in 3 week's time which the patient did not respect despite futile reminder through phone calls.



Figure 1. Picture showing oral warts on the tongue and buccal mucosa



Figure 2a. Picture showing oral cavity immediately after electrocauterization



Figure 2b. Picture showing oral cavity immediately after electrocauterization

Discussion

Human papillomaviruses (HPV) are capable of causing mucosal and cutaneous infections in adults as well as children [11]. Warts are benign epidermal lesions that are dermatological manifestation of this infection [1]. Warts are rare in early childhood and infants with the prevalence lowest among children less than 6 years old. However, the incidence of warts increases as they approach school-age to peak at 12-16 years with a total prevalence as high as 30% [4,11,23]. In our case, the patient was 8 years old which falls between the age groups of the least and peak prevalence, nonetheless she is of school going age where the incidence begins to rise, thus explaining her predisposition.

Some identified risk factors for the transmission of HPV orally include, kissing and oral-genital contact (oral sex or sexual abuse in the case of children). Other risk factors may include poor oral hygiene and immunodepression, such as those with HIV [23]. Spoon sharing (poor hygiene) as was the case in this patient and her younger brother, who also was having an onset of similar warts, could be the possible risk factor for transmission of oral warts in our patient though literature on this has not been established.

The presentation of warts is often a function of or determined by the type of virus and the infection site [1] is the commonest location in descending order include the hands, then the feet, followed by the face and genitalia, unlike the atypical location in our patient which was the oral cavity [11]. Oral warts can appear anywhere inside

the mouth or on the lips and are generally painless, except if they are traumatized or irritated [23]. In this patient, she had multiple painless oral warts, spanning the tongue, the lips and the entire buccal mucosa except the soft palate. Usually, oral warts are small and well demarcated and may occur singly or a small number of them may be present at any point in time. In our patient, the warts were numerous at presentation and only partly discrete. Furthermore, they can have a rough and lumpy appearance in a variety of different guises; as dome-shaped growths that may be white or the color of normal mucosa, flat-topped elevations that have the color of normal mucosa as was the case in this patient though with no regular pattern of arrangement, or thickened frond-like growths [12].

The diagnosis of oral warts is often made after a thorough history and physical exam [15,16]. Laboratory investigations such as biopsy for histopathology or HPV DNA are not common practices as the diagnosis is mainly clinical. The place of biopsy in patient management of oral warts is for the confirmation of the diagnosis in situations where it is not clear (obvious) or in the case of suspected malignancy. Neither PCR for HPV DNA nor biopsy for histopathology was done in this patient as they are very costly, not readily available in our setting and were not extremely needed for the management of our patient. Other investigations necessary include HIV serology and a fasting blood sugar. Our patient's HIV serology, which was negative was investigated given that immune suppressive states are a risk factor for malignant changes in HPV infection (like oral cancer) [13,16]. Other causes of immune suppression such as malnutrition and diabetes were also ruled out in this patient i.e. she was well nourished with a FBS of 98 mg/dl.

As regards the management of oral warts, over the counter treatments which are great for warts on your hands and feet are definitely not a good call for oral warts and no particular therapy has so far been confirmed to be most effective at achieving complete remission in every patient [11]. Thus, the choice of therapy is often adapted for each patient by patient basis, however suggested recommendations is to commence with the least time consuming, least costly and least painful method [22]. As of now, there are 2 major approaches to management, i.e., a medical (topical or injectable agents) or a surgical (by destructive methods) management. As regards to the medical management, salicylic acid is a first-line therapy and has a reported cure rate of 70-80%. Other topical agents such as Podophyllum, Cantharidin, Podofilox, podophyllin, 5-Fluorouracil and Trichloroacetic acid are effective in the management of cutaneous warts but have a high evidence of being toxic and thus are not recommended for the treatment of oral warts. Neither of these were used in the management of our patient because the available topical agent in our hospital was podophyllin, used principally in the management of genital warts but it is toxic and inappropriate for use in the mouth of an 8

year old [3,24]. The antiviral agent Cidofovir, has been effective in the management of recalcitrant wart, but this was at the onset of oral warts, as such was not viewed as an option for this patient with long standing warts [25-27]. Intralesional injections are used only when warts are resistant and refractory to topical agents. This can be in the form of intralesional immunotherapy such as Mumps, Candida or Trichophyton skin test antigens which reports a success rate of up to 74% of cases or in the form of chemotherapeutic agents such as Bleomycin. Interferon-alfa could also be used but there was no indication in our patient for the use of these therapies, as such they were not considered [28,29].

Standard surgery (excision) or cryotherapy are often the recommended methods of treatment when it comes to warts even oral warts [30]. Cryosurgery with liquid nitrogen (-196°C) is the most effective method of cryosurgery and it is often used as first line surgical technique for wart management but in our hospital what was available was cryotherapy using carbon dioxide for the treatment of genital warts [31]. It is equally as effective as topic salicylic acid therapy [3,11,24,32]. Furthermore, electrodesiccation and curettage is another method of treatment and may be more effective than cryosurgery but it is painful and more likely to scar. An alternative and effective surgical destructive method of management is electro cauterization under local anesthesia, which was the method of choice in this patient based mainly on the site of the lesion, the cost, the range of discomfort and the materials available in our setting [33]. Electrocautery was also chosen because it has a shorter duration of treatment as compared to topical agents, injection and cryotherapy. Nevertheless, cryotherapy in combination with salicylates has been shown to be very effective in the eradication process [11]. Laser which is another method of management is an expensive treatment and is reserved only for large or refractory warts. It should also be of note that oral benign warts have the tendency of relapse after treatment and can also carry with them a risk of mouth and throat cancer [30,34].

A limitation of our case study is the loss of follow-up and thus the risk of recurrence as the patient and her guardian failed to respect their follow up visit despite attempts to contact them by phone. This greatly underscores the difficulty in managing patients in our setting as there is often discontinuity in the follow-up of patients probably because of the difficulties to meet the high cost of medical care or poor health education.

Key Summary Points

- This case report underscores oral warts as a very rare pathology in children due to its main means of transmission (oral sex or kissing) which is an uncommon practice in children.
- It also provides information or awareness on the possible occurrence of oral warts in children in

Cameroon which so far is very scarce or non-existent.

- It highlights the diagnostic challenges faced in the diagnosis of oral warts in resource limited settings as the lab investigations involved such as HPV DNA are very costly and unavailable for diagnostic purposes in our setting.
- It lays emphasis on the many management possibilities of oral warts of which none has been proven to be 100% effective for the treatment of all cases of oral wart without a possibility of recurrence.
- It highlights the limitations in the management options in resource limited as a results of unavailable of certain modes of management like cryotherapy using liquid Nitrogen which are more effective methods.
- Finally it talks about the difficulty in following-up patients in developing countries such as Cameroon due to non-compliance by the patients themselves or their guardians in the case of minors.

Conclusion

Oral warts in children are very rare and its most likely aetiology is HPV. Establishing it's the possible aetiologies and management can be challenging. Lack of respect of follow up visits by patients in developing countries can make management more difficult. Spoon sharing is a possible risk factor for oral warts especially in children and electro cauterization is an effective and less costly method of management in pediatric cases.

Consent for Publication

Informed consent was obtained from the patient's Guardian for publication of this case report and any accompanying images.

Ethical Guidelines

All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1964, as revised in 2013.

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