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A Case of Cardiac Papillary Fibroelastoma - An Increasingly Described Cardiac Tumor with Fatal Consequences

Alvin C. Yiu MD; Ali Hussain MD; Uzoagu A. Okonkwo MD, PhD; and John-Paul O’Shea MD, PhD

Abstract

Papillary fibroelastomas (PFE) are rare primary cardiac tumors characterized by non-malignant, pedunculated, endocardial lesions with a significant risk of embolic potential and death. With improvements in the imaging quality and availability of transthoracic echocardiograms (TTE), the diagnosis of PFE has become more common in the last 2 decades. PFE is changing from a rare “zebra” diagnosis to one that community providers will encounter in their practice and must appropriately treat to prevent morbidity and mortality. Data shows that there are significant survival and morbidity benefit associated with surgical excision over non-operative management, with the benefit of anticoagulation remaining unclear at this time. We report a case describing the diagnostic workup and management of a 58-year-old woman who presented with an unidentified endocardial mass determined to be a PFE. Based on current literature, we favor a strategy of early surgical excision of PFE for an optimal reduction in mortality and thromboembolic sequelae associated with this pathology.

Keywords

Papillary Fibroelastoma, Primary Cardiac Tumor, Anticoagulation, Aortic Valve Mass

Abbreviations and Acronyms

AHA = American Heart Association
CMR = cardiac magnetic resonance
CVA = cerebrovascular accident
PET = positron emission tomography
PFE = papillary fibroelastoma
TEE = transesophageal echocardiogram
TTE = transthoracic echocardiogram

Introduction

Primary cardiac tumors are rare, only occurring in 0.02% of autopsies, based on the data from 22 autopsy series. Fortunately, 75% are non-malignant tumors. They are primarily diagnosed in older patients, with a mean age of 60 years old at the time of diagnosis. Fibroelastomas are the most common valvular tumor, with diagnosis becoming more common in the past 2 decades due to advances in resolution and availability in echocardiography. Histopathologically, PFE is a small, highly papillary, pedunculated, avascular tumor covered by a single layer of endothelium. The tumor consists of a hyaline stroma with variable amounts of elastic fibrils. The size of PFE can vary, though most are in the range of 8 mm to 16 mm on the valvular surfaces and 22 mm on the non-valvular right heart endocardium. Although the pathogenesis of PFE is not well understood; it is hypothesized that microscopic endocardial damage and subsequent dysregulated endothelial repair leads to the excessive formation of basal membrane material and formation of PFE. Risk factors for the development of PFE include a history of endocardial surgery, thoracic radiation therapy, history of rheumatic heart disease, and cardiac valvular disease causing trans-valvular pressure gradient. Associations have also been found with other comorbidities to include hypertension, hyperlipidemia, diabetes, and chronic obstructive lung disease. Morphologically, PFE most commonly occurs at valvular surfaces, with the aortic valve being the most common location, followed by the mitral, tricuspid, and pulmonary valves. Outside of valvular surfaces, the left ventricle is the most common endocardial surface of occurrence.

The clinical presentation of papillary fibroelastoma varies. When discovered early, papillary fibroelastomas can be asymptomatic. Still, sequelae range from disruption of cardiac valve function causing dyspnea or clinical heart failure syndrome to severe embolic complications such as ischemic stroke. In thromboembolic cases, the cerebral and retinal arteries are typically affected. With these possible sequelae in mind, we herein describe a case of a 58-year-old female with a history significant for mediastinal radiation therapy and moderate aortic regurgitation, who presented with an initially uncharacterized aortic valve mass later diagnosed as a fibroelastoma on transesophageal echocardiogram.

Case Description

A 58-year-old female with a past medical history significant for mediastinal radiation therapy in 1983 for a diagnosis of Hodgkin’s lymphoma and moderate aortic regurgitation (American Heart Association [AHA] Stage B) underwent her routine yearly surveillance echocardiogram for moderate aortic regurgitation. Significantly, TTE performed one year ago did not detect evidence of a PFE. The current TTE revealed a new, calcified, 9-mm mobile density on the non-coronary cusp of the aortic valve. She denied any symptoms of recent fevers, dyspnea, headaches, vision changes, or focal neurological deficits. She denied any risk factors for infective endocarditis, which can mimic the appearance of PFE, including a history of intravenous drug use, chronic infusion therapy, or hemodialysis. Laboratory evaluation revealed no evidence of significant abnormalities,
and 3 sets of blood cultures from multiple sites were negative for any evidence of bacteremia. She was admitted for further imaging and differentiation of her newly discovered cardiac mass. A repeat TTE was not able to discern the etiology of the mass. Therefore, she underwent further evaluation with a transesophageal echocardiogram (TEE). The TEE revealed good visualization of a tricuspid aortic valve with a 9-mm mobile, pedunculated, non-obstructing, calcified mass on the non-coronary cusp consistent with a calcified PFE with no other masses observed on the remaining valves or visible endocardial surfaces (Figure 1 and Figure 2). Given clear visualization of the PFE with TEE, further imaging studies were not pursued. The patient was counseled that elective surgical removal is the definitive management of PFE to reduce the risk of thromboembolism and was given the option for surgical referral. However, the patient desired to delay surgical intervention for a second opinion with her primary outpatient cardiologist. Given her election for nonsurgical management at that time, as well as the lack of other comorbidities, including heart failure, hypertension, previous embolic events, diabetes, and vascular disease, she was started on a low dose aspirin 81 milligrams (mg) by mouth daily and discharged home with yearly TTE follow-up. The patient ultimately decided to continue pursuing medical management of her PFE with aspirin and continued yearly TTE studies. She remains compliant on aspirin 81 mg daily and asymptomatic to date.
Discussion

This case illustrates the recognition of a PFE in a patient with aortic valve regurgitation and a history of thoracic radiation therapy. The patient’s chronic valvular regurgitation is a risk factor for the development of PFE by causing turbulent transvalvular blood flow resulting in microscopic valvular injury. The patient’s history of mediastinal radiation may also have been a predisposing factor by inducing cardiac endothelial damage.

When a cardiac valvular mass is initially incidentally visualized on imaging, other etiologies of valvular mass, based on clinical history and morphology, should also be considered. TTE evaluation should be followed with TEE evaluation to elucidate the mass both descriptively and quantitatively. TEE is the imaging modality of choice, and cardiac magnetic resonance (CMR) imaging is not required for diagnosis. In our case, the TEE adequately characterized the size and morphology of the PFE, so CMR was not necessary. However, CMR and positron emission tomography (PET) have been described in the literature as other viable modalities to investigate cardiac masses.

CMR demonstrates high accuracy in differentiating cardiac thrombi from other tumors. PET imaging may also be used to distinguish between benign cardiac mass from malignant masses via evaluation of metabolic activity. Furthermore, additional studies have also shown that CMR may detect intra-cardiac masses missed on initial echocardiography.

Patients with PFE are at significantly elevated risk for death when compared to age-matched healthy controls. Significantly, a retrospective review by Tamin et al showed that all-cause mortality at 5 years is twice as high in PFE patients as in age and sex-matched controls. Patients with PFE are at significantly higher risk of cerebrovascular accident (CVA) than age and sex-matched counterparts, even with medical and surgical treatment. Tamin and colleagues reported that the rate of observed CVA at 10 years was 2.4 times and 3.4 times higher in matched PFE patients who were treated with surgery and who were treated medically, respectively. Tumor mobility has also been shown as an independent predictor of PFE-related death and non-fatal embolization.

Surgical resection is the gold standard of treatment for papillary fibroelastoma, even for those patients who are asymptomatic. In surgical removal, the roof of the pedicle and full thickness of endocardium is removed. The resulting defect may be closed by either primary closure or pericardial patch. Recurrence of PFE after surgical excision is rare, noted in 0.04% of PFE cases. Patients who undergo surgical excision have 30% higher overall survival rates in the first 7 years post-procedure than those who opt for non-operative management. Furthermore, among those who underwent surgical excision, only 8% experienced neurologic sequelae at 5-year follow-up, versus 13% in a pooled group of those who did not undergo surgical
excision, regardless of anticoagulation decision. However, the optimal medical management of PFE is unknown. There are currently no randomized controlled trials directly comparing anticoagulation versus antiplatelet treatment versus surgical excision. In 2 separate retrospective reviews, Tamin et al and Gowda et al describe anticoagulation regimens with one of the following regimens: (1) warfarin, (2) heparin, (3) aspirin and clopidogrel, (4) aspirin alone, or (5) clopidogrel alone. There are currently no large-scale studies describing anticoagulation with direct oral anticoagulants. Even within cohorts, the decision to anticoagulate and the results of treatment were variable. In a case series of 725 patients identified via literature search, Gowda et al described only 57 patients anticoagulated with either heparin or warfarin. Tamin et al described a group of 121 PFE patients who experienced neurologic events despite being on anticoagulant therapy, of whom 27 were on warfarin (22%), 57 on aspirin (47%), 1 on clopidogrel (1%), and 2 on dual antiplatelet therapy (2%). This report was not sufficiently powered to detect a difference in outcomes between the different treatment modalities. Therefore, optimal anticoagulation regimen remains unclear, although non-operative management is inferior to surgical excision of PFE.

For those patients who undergo medical therapy alone, current literature agrees that serial TTE should be performed for surveillance. However, there is no clear consensus in recommendations for imaging intervals. Imaging intervals in case reports have ranged from every 2 months to more than 1 year. Muthu et al pursued a close interval follow-up with TTE at 2-month intervals until 6 months, and then biannually after that for a PFE patient who presented with dyspnea. Others have followed asymptomatic patients at closer intervals. Seol et al pursued TTE at 2-month intervals for 3 years for asymptomatic patients. In contrast, Ayabe et al followed patients with less than 1 TTE a year to average 7 over 10 years. In our case, we considered the patient’s AHA Stage B aortic regurgitation when proposing a surveillance schedule. We chose to pursue a yearly TTE surveillance schedule to adequately screen for both progression of her PFE and progression in her aortic regurgitation as recommended by the American College of Cardiology.

We conclude that all patients with PFE should be considered for surgical excision of the tumor as a first-line intervention. This procedure should be done regardless of symptoms to reduce incidence of thromboembolic events or death. If the patient is not a surgical candidate, medical management of PFE should focus on therapeutic anticoagulation or antiplatelet therapy. Clinical factors known to increase the risk of arterial thromboembolism, age, sex, and pertinent past medical history such as heart failure, hypertension, previous embolic events, diabetes, and history of coronary artery disease should be taken into consideration when crafting an individual risk-benefit assessment. It may be reasonable for patients with elevated risk factors for thromboembolism to undergo systemic anticoagulation with heparin or warfarin.

Conversely, those with less risk factors may warrant only aspirin or clopidogrel alone, as in our patient. Any decision to initiate systemic anticoagulation should be weighed against potentially fatal bleeding. Clinical risk factors for bleeding include hypertension, renal disease, liver disease, history of hemorrhagic stroke, prior bleeding, age, and concomitant anticoagulant use. These should be evaluated to assess a risk-benefit analysis before anticoagulation.

Papillary fibroelastoma is a histologically benign tumor that causes significant morbidity and mortality from thromboembolic events. Patients should undergo surgical excision of the fibroelastoma for survival benefit. If patients decline surgery and opt for medical management with anticoagulation therapy alone, the optimal regimen is unclear. The choice between administration of heparin, warfarin, aspirin, or clopidogrel should be made with consideration of thrombotic risk factors and factors for major bleeding events. In the future, more research is needed to compare varying regimens of anticoagulation treatment, especially the use of direct oral anticoagulants, as well as more thorough comparison between the long-term outcomes of anticoagulation instead of surgical excision in the treatment of PFE.

Conflict of Interest

None of the authors identify a conflict of interest.

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Human Fascioliasis (Liver Fluke Disease) in Hawai‘i: Case Report and Review of Human Fascioliasis Acquired in the United States

Joel D. Brown MD, FIDSA, DTM&H

Abstract

Fascioliasis is a foodborne zoonotic infection caused by the trematode liver flukes: Fasciola hepatica and Fasciola gigantica (F. gigantica). Infections may cause acute and chronic hepatobiliary tract diseases in herbivore animals and humans. Fascioliasis is present worldwide, particularly in regions where sheep and cattle are raised. The global burden of human fascioliasis is estimated to be 2.7 million. Human infections are rare in the United States, and most infections were acquired abroad. In the 1950s, several human cases of F. gigantica infection were reported from Hawai‘i, but no subsequent cases have been reported until the case described here.

This case report describes a man from Hawai‘i Island who ate raw wild watercress, and 22 days later, developed acute phase fascioliasis with fever, acute hepatitis, peripheral eosinophilia, and hypodense lesions seen on liver diagnostic imaging. Immunodiagnostic tests were positive for Fasciola species. Based on earlier reports from Hawai‘i, F. gigantica infection was likely. Clinical abnormalities resolved after treatment with triclabendazole. Physicians should consider fascioliasis in immigrants and travelers from endemic areas with acute hepatitis and eosinophilia after eating raw watercress or chronic hepatobiliary disease. Hepatobiliary imaging and serological testing are useful for diagnosis. Oral triclabendazole is the preferred treatment. Animal fascioliasis appears to be spreading in the United States, and the popularly perceived health benefits of eating raw wild watercress and other aquatic plants may lead to more human infections. The rarity of human infections in Hawai‘i suggests that it is safe to eat commercially grown watercress cultivated in Hawai‘i.

Keywords

Fascioliasis, Fasciola gigantica, Liver disease, Hawai‘i, United States

Abbreviations and Acronyms

CDC = Centers for Disease Control and Prevention
EIA = enzyme immunoassay
ELISA = enzyme-linked immunosorbent assay
ES = excretory-secretory
FAST = Falcon assay screening test
FDA = US Food and Drug Administration
RV = reference value

Introduction

Fascioliasis is a foodborne zoonosis of animal herbivores caused by the parasitic trematodes (liver flukes), Fasciola hepatica (F. hepatica), and less commonly, Fasciola gigantica (F. gigantica). Animal fascioliasis occurs on all continents except Antarctica. In the mid-1990s, human fascioliasis emerged in many countries worldwide, predominately in populations living in cattle and sheep-rearing regions with limited resources. The estimated global burden of human infection is 2.7 million. The parasitic life cycle is complex. Briefly, animal herbivores, typically domesticated cattle and sheep are the primary hosts, aquatic snails are the intermediate hosts, and freshwater aquatic plants are the means of transmission. Humans may become incidental hosts after eating raw aquatic plants (eg, watercress). Human fascioliasis is rarely reported in the United States, and most patients were immigrants or American travelers returning from endemic regions. Hawai‘i is considered an endemic area based on livestock studies and human cases reported in the 1950s. However, no Hawai‘i human cases have been reported subsequently. Informal inquiries of local Hawai‘i infectious diseases and gastroenterology specialists found no cases diagnosed in the past 45 years. This case report describes a patient who developed acute phase hepatic fascioliasis acquired in 2008 after eating raw wild watercress on Hawai‘i Island. Further, this report examines documented cases of human fascioliasis acquired in the United States.

Case Report

A previously healthy 58-year-old man living in a rural area of Hawai‘i developed chills and fevers approximately 101.5°F, followed by progressive fatigue, weight loss, diffuse pruritus, and moderate, intermittent pain in the right upper abdomen and right anterior lower chest. His past medical history was unremarkable. He was born and raised in the northern part of the United States and had not traveled abroad in 30 years. His initial physical examination was remarkable for a slightly tender liver felt 3 cm below the right costal margin and slight dermatographism. Laboratory studies revealed a leukocytosis of 12,600 cells/mm$^3$ with 33% eosinophils (absolute eosinophil count $4,158$ cells/mm$^3$). The combination of liver tenderness and eosinophilia in a patient on Hawai‘i Island led an infectious disease consultant to suspect acute hepatic fascioliasis. Further history revealed that 2 weeks before the onset of symptoms, the patient had eaten a half-cup of raw wild watercress collected from a pond adjacent to a cattle pasture. Blood levels of liver-associated enzymes were elevated, with an alkaline phosphatase of 324 IU/L (reference value [RV]: 33–130), alanine aminotransferase 91 IU/L (0–40), and aspartate aminotransferase 43 IU/L (0–37). The serum bilirubin level was 0.7 mg/dL (RV: 0.2–1.5). Ultrasonography revealed multiple hypoechoic areas in the liver, and a computed tomography scan showed multiple hypodense areas in the liver (Figure 1). Fasciola serologic testing by a commercial laboratory (Parasitic Disease Consultants, Georgia) using enzyme-linked immunosorbent
assay (ELISA) to crude *F. hepatica* worm extract was initially borderline positive with a titer of 1:32 (reference range: negative <1:32; positive >1:32). Six weeks later, the titer rose to 1:64, and the sample was also positive for antibodies against *Fasciola hepatica* excretory-secretory (ES) antigens by Falcon assay screening test ELISA testing done at the Centers for Disease Control and Prevention (CDC) reference Laboratory of Parasitic Immunology, San Juan, Puerto Rico. Pending availability of triclabendazole, an investigational drug, he was treated with 500 mg of nitazoxanide orally twice daily for 7 days without clinical improvement. A second nitazoxanide treatment also failed. The hospital’s Institutional Review Board approved a US Food and Drug Administration (FDA) Investigational New Drug program. With the patient’s consent, he was treated with 10 mg/kg of triclabendazole once by mouth with a meal. Following treatment, his symptoms resolved, and the blood eosinophil counts and liver-associated enzyme tests became normal 10 weeks later. *Fasciola* antibody levels gradually decreased and were normal 9 months after treatment. Eight stool specimens obtained before and after treatment were negative for ova and parasites. Although no adult parasites were recovered from this patient, the epidemiologic, clinical, and serological findings were strong evidence for acute hepatic phase fascioliasis. Based on earlier studies in Hawai‘i, *F. gigantica* was probably the culprit species in this patient.6

## Discussion

### Fasciola Life Cycle and Epidemiology

The complex *Fasciola* life cycle is similar in both animal and human hosts. Adult *Fasciola* flukes dwell in the principal host’s hepatobiliary system and produce eggs passed in the feces into freshwater environments (Figure 2). The eggs hatch into miracidia (immature larvae) that infect specific aquatic snail intermediate hosts, releasing cercariae that attach to aquatic plants as infectious metacercariae. After the host ingests the plants, the metacercariae excysts become motile juveniles, which penetrate the host’s intestines, enter the peritoneum, and invade the liver. In this acute phase of infection, the migrating juveniles move through the liver for several weeks, causing acute hepatitis and leaving visible tracks of destruction. Some juvenile flukes may appear in various ectopic sites, such as the skin and respiratory tract. In the chronic phase of infection, the larvae enter the host’s hepatobiliary system, where they mature into adult egg-producing flukes to complete the life cycle. *F. hepatica* adult flukes are approximately 30 mm long, and *F. gigantica* adults are approximately 75 mm long (Figure 3). Adult flukes live in the hepatobiliary system for about 10 years. They might cause chronic or relapsing hepatobiliary tract-related diseases. In low-resource countries with poor sanitation, humans are infected by eating various freshwater aquatic plants grown

![Figure 1. Contrast-Enhanced Computed Tomography of the Abdomen Showing Multiple Hypodense Areas in the Liver.](https://www.cdc.gov/parasites/fasciola/biology.html)
in wetlands adjacent to infected animal grazing areas or areas irrigated with contaminated water. Infection can also occur after drinking contaminated water or ingesting food washed with the water. The means of disease transmission varies with local geographic agricultural and nutrition practices. In poor, hyperendemic regions, human feces may contribute to the spread of infection. F. hepatica is found mainly in temperate regions and is responsible for most human infections. Human infections occur predominantly in the Andean Highlands of Latin America (Bolivia, Ecuador, Peru), North Africa, the Middle East (Egypt, Iran), East Asia (China), Iran, and to a lesser extent, Western Europe (Portugal, France, and Spain), and several Caribbean islands including Puerto Rico. F. gigantica is closely related to F. hepatica and is found in tropical and subtropical regions of Africa, Asia, the Western Pacific, and Hawai‘i. However, in Asia, the distribution of F. hepatica and F. gigantica overlap, making it difficult to identify the infective species unless adult flukes are recovered.

Clinical Features of Human Fascioliasis

The manifestations of human fascioliasis depend on the phase of the infection. Many infections are asymptomatic, but infections may cause acute hepatitis or chronic relapsing hepatobiliary disease. The clinical features of F. hepatica and F. gigantica infections are similar. The acute liver phase is due to immature parasites migrating through the liver. Frequent complaints are right upper quadrant abdominal and epigastric pain, fatigue, and fever; other symptoms might include cough, pruritus, urticaria, and dermatographia. Less commonly, larval flukes may migrate to ectopic sites in the skin, respiratory system, pancreas, genitourinary tract, eyes, or brain. Laboratory studies during the acute phase usually show elevated liver-associated enzymes and intense blood eosinophilia. The chronic phase of human fascioliasis begins several months later and may persist for years. Chronic fascioliasis is mainly asymptomatic, but ongoing hepatobiliary inflammation may manifest as liver fibrosis, cirrhosis, intermittent abdominal pain, biliary obstruction, cholangitis, and liver abscess, or pancreatitis. Eosinophilia is less common than during the acute stage, and eggs appear only intermittently in the feces. Chronic fascioliasis is usually unsuspected until adult flukes are discovered accidentally during surgery or endoscopy. Human infection with 2 other liver flukes, Clonorchis sinensis (Chinese or oriental liver fluke) and Opisthorchis viverrini (Southeast Asian liver fluke), can induce liver cancer and cholangiocarcinoma; however, there is no evidence that Fasciola infections are associated with hepatobiliary tract cancer. Hepatic imaging studies reveal multiple liver lesions in the acute hepatic phase of infection and various hepatobiliary tract abnormalities in the chronic phase.

Diagnostic Testing

Diagnosis by fecal examination for Fasciola eggs is problematic. Eggs are not present in the stool during the acute phase of infection, and egg production is irregular in chronic infections (Figure 4). Egg morphology cannot reliably distinguish F. hepatica from F. gigantica. Fasciola spp. eggs can be difficult to distinguish from eggs of other trematodes, for example Fasciolopsis buski (the giant intestinal fluke). Recovering adult worms in endoscopic or surgical specimens is definitive and allows species identification based on the adult size of F. hepatica.
hepatica and F. gigantica. Serologic testing is beneficial for diagnosing both acute and chronic phase infections. Specific antibodies to Fasciola become detectable 2 to 4 weeks after infection. Immunodiagnostic tests for fascioliasis include an enzyme immunoassay (EIA) with ES or recombinant antigens with confirmatory testing of EIA-positive specimens with an immunoblot assay. Serological testing is available from commercial reference laboratories and the CDC. Current serological testing is reliable for both F. hepatica and F. gigantica infections but does not discriminate between the 2 species. Immunodiagnostic testing methods continue to evolve, and sensitivities and specificities can be 94%. However, both infections have similar clinical features and responses to treatment.

Treatment

Triclabendazole is the drug of choice for fascioliasis. The drug is active against immature and adult parasites and is an effective treatment for acute and chronic infections. In February 2019, the FDA approved triclabendazole treatment for fascioliasis in patients at least 6 years of age. Triclabendazole is given orally, with food, to improve absorption. The current recommended dosage is 2 oral doses of 10 mg/kg given 12 hours apart. After treatment, ELISA-based testing results become negative in 91% of the cured cases after 1 year and become negative 6 months after retreatment in the remaining cases. However, triclabendazole resistance has been documented, particularly in infected animals and some infected humans. An alternative drug, nitazoxanide, is available in the United States and may be an effective therapy in some patients. The adult dosage is 500 mg orally with meals twice a day for 7 days. However, nitazoxanide was ineffective in the case-patient presented here. Praziquantel is active against most trematodes but is not active against Fasciola parasites and should not be used for fascioliasis.

Review of Human Fascioliasis Acquired in the United States

Few cases of domestically acquired human fascioliasis have been reported in the United States. In 2010, Fried found 54 human cases reported in the United States during approximately 120 years. Twenty-six cases were infected while outside the United States. Two cases were in Puerto Rico, 21 cases were in Hawai‘i; only 2 proven and 3 probable patients were in the continental United States. Norton reported the first case of human fascioliasis acquired in the United States in 1961. In this report, Norton describes a 50-year-old woman in California who developed upper abdominal pain and tenderness. Laboratory test results were normal, and there was no eosinophilia. She underwent a cholecystectomy, and a single adult F. hepatica fluke was discovered incidentally in the common bile duct. Her symptoms resolved after the fluke was extracted.

Further history revealed that 10 years earlier, while living on a livestock ranch in California, she and 3 neighbors had gathered and eaten raw wild watercress. Some weeks later, she developed right upper abdominal pain, erythema nodosum, and eosinophilia; her 3 neighbors had similar symptoms. This patient probably had unrecognized acute fascioliasis followed by chronic fascioliasis. Neff reported a Florida man who developed right upper abdominal pain, eosinophilia, and abnormal liver imaging studies. At laparoscopy, serpiginous lesions were seen on the liver’s surface; serological studies were positive for F. hepatica. The patient then admitted to having eaten wild watercress shortly before his illness. His symptoms resolved after treatment with triclabendazole. Perlada and Weisenberg reported 2 patients in northern California who developed acute hepatic fascioliasis after eating raw wild watercress, which they had shared. Both patients had eosinophilia, liver lesions on imaging, and positive immunodiagnostic testing for fascioliasis. The patients recovered after triclabendazole treatment. One case report described a woman with a long history of liver and bowel disease diagnosed with fascioliasis after parasite fragments were recovered from a liver cyst and positive Fasciola serology tests. Her infection was described as acquired in the United States; however, she had a history of traveling to France, where fascioliasis is more prevalent. In 1982, a limited survey of human fascioliasis in the US territory of Puerto Rico found 12 of 110 fecal samples positive for F. hepatica eggs; 11 of the 12 had a history of eating watercress grown on local farms.

Fascioliasis in Hawai‘i

In 1938, Joseph E. Alicata, PhD, a noted parasitologist in Hawai‘i, described the life history of F. gigantica, the common liver fluke of cattle in Hawai‘i. The parasite was first reported in Hawai‘i cattle in 1892. The snail intermediate host was a freshwater snail, Fossaria ollula, found in wet lowlands where cattle fluke infections were common. F. gigantica infections were believed to have been introduced into Hawai‘i by water buffalo from Asia, which spread F. gigantica infections to Hawai‘i cattle, sheep, goats, and swine. Alicata reviewed a 46-year history of human fascioliasis in Hawai‘i. At that time, cattle in the Hawaiian Islands were commonly infected with F. gigantica; only 1 infection was due to F. hepatica. Alicata’s review included 19 cases of human F. gigantica infection in the Hawaiian Islands. The first case was described in 1906, and in all cases, adult F. gigantica flukes were discovered by chance during surgical operations. Mature flukes were recovered from the liver and ectopic sites in the peritoneal cavity, upper respiratory tract, skin, and in 1 case, in the external ear canal. Stemmermann reviewed Alicata’s cases and added 3 more cases, including 1 autopsy case. He described cattle grazing in unfenced wetlands, which were also used for growing watercress for local markets. At the time, hog hunters reported finding liver flukes in wild hogs, known to frequent these wetlands. F. gigantica accounted for all the infections in which flukes were found. Many patients had been symptomatic for 3 months to 10 years before the diagnosis was made. All patients resided in Hawai‘i, but their travel histories and countries of origin were not described.
Animal fascioliasis is not a reportable disease in Hawai‘i or the United States, and current information regarding animal disease in Hawai‘i is unavailable. Before 1995, the Hawai‘i State Department of Agriculture was responsible for animal slaughterhouse food inspections. Animal liver fluke infections, detected by gross inspection of the liver, were described as widespread. In 1995, this responsibility was assumed by the US Department of Agriculture (personal communication from Jason Moniz, DVM; Hawai‘i Animal Disease Control Branch). Animal fascioliasis is enzootic in regions of the continental United States, primarily in the Gulf coast regions with high annual rainfall, and in California and other western states with areas of poorly drained or irrigated pastures.  

Most human fascioliasis cases are due to eating raw watercress or other aquatic plants harvested from areas contaminated by infected livestock.  

Nearly all human fascioliasis cases acquired in the United States are caused by eating raw wild watercress. In France, a 2002 outbreak of acute human fascioliasis was attributed to ingesting watercress grown in a commercial aquaculture bed adjacent to a cattle pasture. The popular perception of the health benefits of watercress and the practice of foraging for wild foods may increase human fascioliasis infections in the United States.

**Conclusion**

Few physicians in industrialized countries are familiar with fascioliasis. Most of the reported cases described above were unsuspected until an adult fluke was discovered incidentally during an investigation of hepatobiliary diseases. A patient history of eating raw, wild watercress is an important clue to the diagnosis, but this history was usually obtained after fascioliasis was diagnosed by other means. Likely, human fascioliasis occurs more often in the United States than is reported in the medical literature. Physicians should consider fascioliasis in travelers and immigrants from endemic areas who present with acute hepatitis with eosinophilia or chronic or intermittent hepatobiliary disease. A history of eating raw wild watercress or other raw freshwater aquatic plants is an important diagnostic clue. The diagnosis is best made with immunodiagnostic testing performed at reference laboratories. Treatment with triclabendazole is usually curative. The case-patient with acute hepatic fascioliasis described here indicates that fascioliasis persists in Hawai‘i livestock, and eating raw wild watercress is a risk for infection. However, the rarity of human infections in Hawai‘i suggests that it is safe to eat commercially grown watercress cultivated in Hawai‘i.

**Conflict of Interest**

The author has no disclosures or conflicts of interest.

**Disclosure Statement**

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Podoconiosis: A Possible Cause of Lymphedema in Micronesia

Paul M. Gahlinger MD, PhD, MPH

Abstract

Podoconiosis is a type of tropical lymphedema sharing some clinical characteristics with lymphatic filariasis. Also referred to as endemic non-filarial elephantiasis, podoconiosis is a non-infectious disease from barefoot exposure to irritant red clay soil of volcanic origins. Podoconiosis is most common in Ethiopia and has also been reported in many other countries, but not in the Pacific Islands. Lymphatic filariasis is endemic in the Pacific Islands and was historically reported as elephantiasis in Micronesia. It was considered to have been eradicated in Guam and the Northern Mariana Islands following World War II. A small number of patients in Saipan exhibited characteristics of lymphatic filariasis but were seronegative for filariasis. Clinical examination of these patients matched podoconiosis much more closely than filariasis. Moreover, these patients reported a history of chronic barefoot exposure to irritant red clay soil and a prodrome characteristic of podoconiosis. While this study is limited to several cases, the results suggest that podoconiosis could be considered a cause of non-filarial lymphedema in Saipan and perhaps other islands in Micronesia. Preventive patient education is focused on discouraging barefoot exposure to red clay soils, particularly in those with a family history of lymphedema. Early recognition of the possibility of podoconiosis would allow appropriate treatment and prevent progression to later debilitating stages of the disease.

Keywords

podoconiosis, lymphedema, filariasis, elephantiasis, Saipan, Micronesia

Abbreviations and Acronyms

LF = lymphatic filariasis
KCHC = Kagman Community Health Center

Introduction

The Kagman Community Health Center (KCHC) is a rural federally qualified health center in Saipan, Northern Mariana Islands, established in 2013. While serving as Medical Director of KCHC intermittently from 2013 to 2018, and as founder of the Sefin clinic in Chuuk, Federated States of Micronesia from 2016 to 2017, the author treated a number of patients previously diagnosed with presumptive lymphatic filariasis (LF), more commonly known as elephantiasis. These cases all presented in an advanced stage of lymphedema, with bilateral fibrotic swelling in the feet and legs. The patients had been given the standard antihelminthics without benefit. Moreover, laboratory tests were unremarkable and they were seronegative for filariasis. Intrigued with the possibility of a non-filarial cause of lymphedema in these patients, the author collected extensive histories and a more detailed physical examination of 8 cases. The results revealed a history and clinical presentation much more suggestive of podoconiosis than LF. However, podoconiosis has not been previously reported in the Pacific islands. Lymphedema is the failure of lymph drainage, typically presenting with swelling in the extremities. Primary lymphedema is a genetic condition with incompetent or missing lymph valves, known as Milroy disease. Secondary lymphedema can result from many types of injury to the lymphatic system, including radiation, surgery, trauma, inflammation, and infection. The most common cause in the United States is radiation treatment for cancer.

In tropical countries, lymphedema usually results from infection by a parasitic worm (filaria, usually *Wuchereria bancrofti*) transmitted by the bites of infected mosquitoes. LF is endemic in 49 countries, with an estimated 120 million infections and 40 million disfigured by the disease. The Pacific region was endemic in American Samoa, the Cook Islands, Fiji, French Polynesia, Micronesia, Samoa, Tonga, Tuvalu, Niue, Papua New Guinea, and Vanuatu. Many of these have reported elimination. During World War II, the elephantiasis presentation of lymphedema was commonly observed in the Northern Mariana Islands. Recalling his experience during the invasion, the United States Marine Corps Captain John C. Chapin wrote: “Physical conditions of many were pitiful. Every illness that we had been briefed on was observed [in the area]: leprosy, dengue fever, yaws and many cases of elephantiasis.”

Following the war and transfer of the Northern Mariana Islands to a US Trust Territory, public health programs were considered to have successfully eradicated filariasis in Guam and the Northern Mariana Islands. Podoconiosis (“dust in the feet”) presents as bilateral asymmetrical swelling of the feet and legs. It is a type of tropical lymphedema sharing some clinical characteristics with LF. Also referred to as endemic non-filarial elephantiasis, or “mossy foot,” podoconiosis is a non-infectious disease arising from chronic barefoot exposure to irritant red clay soil of volcanic origins. The precise trigger is unknown, but this disabling condition is considered to be caused by an abnormal inflammatory reaction to the passage of microparticles of silica and aluminum silicates through the skin. These particles are taken up by macrophages into the lymphatic system and result in an inflammatory process leading to fibrosis and obstruction of the vessels.

The disease process has 3 phases: prodromal, early, and advanced. Prodromal symptoms include itching of the skin of the forefoot and a burning sensation in the foot and lower leg, with variable chills or generalized joint pains. These symptoms may progress to the early phase of swelling and splaying of the forefoot and leakage of clear lymph fluid. The skin develops hyperkeratotic papillomata that resemble moss or velvet, giving rise to the African term “mossy foot.” Acute episodes may cause...
fever, rigors, and a rapid increase in pain and swelling of the leg. In the advanced phase, hard, fibrotic swelling develops in both legs and feet, though 1 is usually more severely affected than the other.5

Podoconiosis is most common in Ethiopia but is also widespread in tropical Africa, Central, and South America, Indonesia, India, and has also been reported in many other countries. It has not, however, been previously reported in the Pacific Islands.6

The purpose of this study is to consider whether podoconiosis may be a cause of non-filarial lymphedema in the Northern Mariana Islands.

Case Report

Detailed histories, physical examinations, and laboratory tests were collected on 8 patients in Saipan who presented with fibrotic bilateral lymphedema in the lower extremities. The age range was from 41 to 66 years, with 5 males and 3 females. All were Pacific Islanders: 5 of Chamorro (Guam and the Northern Mariana Islands) and 3 of Carolian (2 Chuukese, 1 Yapese) ancestries. All had a lengthy residence in Saipan of over 10 years and reported their symptoms began after residing in Saipan. There were likely other cases that did not come to medical attention. In Africa, “mossy foot” can cause great social stigma and is a “hidden disease” since the afflicted hide from sight, not wanting to suffer the social embarrassment of their disfigurement. Several of the Saipan cases reported relatives with the condition who were reluctant to seek medical attention because they felt it was futile or feared surgery (amputations for gangrenous diabetic feet were very common).

Physical examination and laboratory results were unremarkable except for obesity, mild hypertension, or mild diabetes in several, but not all, cases. All were seronegative for microfilariae, Wuchereria bancrofti antigen, or specific IgG4 antibodies. All cases met the podoconiosis diagnostic standard: bilateral, asymmetrical lymphedema of the lower limb present for more than 1 year, with negative serology for LF, and a history of any of the associated clinical signs and symptoms.5 These cases also reported the characteristic exposure and prodromal symptoms of podoconiosis, which differ from LF and other causes of lymphedema.

The most detailed history was obtained from the 41-year-old female, the youngest of the cases. In adolescence and early adulthood, she lived with her aunt in a simple dwelling with a dirt floor of hard red clay that she swept daily. She was barefoot almost all of the time. She states that she was healthy until her early 20s when she began to experience itching in her feet and burning pain and swelling in her left foot and leg. When the pain and swelling became acute, she sought medical attention and was given a short course of antibiotics with no benefit. The swelling was gradually progressive over several years, with the right foot and leg also affected. She developed severe fungal infections on her legs and weeping lesions of clear fluid that became malodorous and attracted flies. The pain was worse at night and partly relieved by uncovering her legs. She again sought medical attention and was treated with doxycycline and albendazole without improvement. She became increasingly homebound because of her difficulty ambulating and because she was embarrassed about her unsightly condition. She gained weight leading to obesity.

On examination, she was normotensive and did not have diabetes. Both feet and legs were grossly enlarged, hyperkeratotic, with non-pitting edema and evident areas of fungal infection (Figure 1). There appeared to be mild lymphatic or serous oozing. Her lower extremities had normal sensation and capillary refill, but it was impossible to detect peripheral pulses because of the edema and induration.

She was followed for 3 years and given manual decompressive therapy and trials of compression stockings and Unna boot, achieving only very modest improvement. Because of the advanced state of fibrosis, her only other therapeutic option would have been radical surgical debulking, which she declined.

Diagnosis

These cases show lymphedema that does not appear to have characteristic features of LF. However, it is difficult to rule out LF simply by the absence of microfilariae or seronegative antigen results. Microfilariae of Wuchereria and Brugia exhibit a nocturnal periodicity, and an accurate diagnosis is best achieved on smears collected at night (10:00 PM to 2:00 AM). In advanced stages, microfilariae may be absent. Because sensitivity for detecting microfilariae can be low and variable, immunoassay for circulating filarial antigens is a useful diagnostic approach. However, late stages of the disease are often seronegative. The distinction is, therefore, based mainly on clinical signs, symptoms, and characteristic exposure.

Podoconiosis typically has a prodrome of itching. Symptoms start in the foot and ascend to the knee, with swelling rarely above the knees, although there may be femoral node tenderness. The swelling is bilateral but asymmetric. Conversely, LF lacks the prodrome of itching or burning feet. Symptoms tend to start in the groin and are descending, with unilateral leg swelling. Podoconiosis requires chronic barefoot exposure to irritant red clay soil of volcanic origins, containing silica and aluminum silicates. Saipan has regions of this type of soil, notably in the area of the previous home of the described case.6 In Africa, podoconiosis is found in highland areas (typically over 1500 m) where rainfall is higher (mean annual rainfall of over 1500 mm), which causes weathering of volcanic rock into silicate clays with particle size less than 2 μm. It is not found in the lower, more arid regions. In Africa, elevation is, therefore, a
A differential diagnosis of tropical lymphedema should also include leprosy, mycetoma pedis, and Milroy disease. Podoconiosis can be distinguished from leprosy by preserving sensation in the affected limb and the isolation of disease to the lower extremities. Mycetoma pedis (mycetoma of the foot), often referred to as Madura foot, is endemic in Africa, India, and Central and South America. It is caused by a bacterial or fungal infection and presents purulent exudate. Milroy disease typically has its onset of swelling and edema at birth or early infancy, although rare variants have a later onset. Each of these conditions differs in clinical presentation from podoconiosis.

Discussion

In 905 CE, the Persian physician Rhazes first recorded that elephantiasis “of the Greeks” was different from that “of the Arabs.” The first likely referred to lepromatous leprosy, while the second is now considered to be podoconiosis. In the 1920s, persistently negative tests for filaria in Guatemala led investigators to suggest that the endemic form of elephantiasis was associated with walking barefoot rather than an insect vector. With the discovery in the 1970s of lymph nodes and macrophages laden with silicon, aluminum, and other soil metals, the term podoconiosis was proposed for this form of non-filarial elephantiasis. Podoconiosis is now thought to have been prevalent in Northern Africa (Algeria, Tunisia, Morocco, and the Canary Islands) and Europe (France, Ireland, and Scotland) but no longer found in these countries due to the widespread use of shoes. The global burden of podoconiosis is estimated at 4 million afflicted people in 32 countries, mainly in tropical countries of Africa, Central and South America, and Southeast Asia. Underreporting of podoconiosis is likely because of diagnostic challenges and a low index of suspicion.

Podoconiosis is a non-communicable disease, easily prevented by avoiding exposure to irritant volcanic soils by good foot hygiene and the use of socks and shoes. Late-stage disease can be treated with lymphatic decompression therapy, either with manual or pneumatic devices, and compression stockings, or Unna boot. For very severe hyperplastic and verrucous elephantiasis, debulking surgery is an option. Early stages are reversible given good foot hygiene and protective footwear, whereas late stages result in considerable economic and social difficulties and, despite treatment, may never fully resolve.
At the beginning of the author’s work in Saipan, barefoot walking was commonly observed, even in the main villages and modern buildings. However, the island was rapidly developing with a burgeoning tourism industry. The increasing prosperity led to upgrades in dwellings. In 2015, typhoon Soudelor devastated a large part of the island. Extensive reconstruction created new housing and further hastened the transition to modern footwear. By the end of the author’s tenure in Saipan in 2018, barefoot walking was seldom observed. It is interesting to note that all of the observed cases were middle-aged or older and in an advanced stage of the disease. None showed early signs. Economic development in Saipan may be responsible for the decrease in cases.

**Conclusion**

The purpose of this case series and review is to show that podoconiosis may be the cause of non-filarial lymphedema in Saipan and perhaps other regions of Micronesia and therefore encourage healthcare providers to be alert to this entirely preventable disease.

**Conflict of Interest**

The author has no relationship with any person or entity that could be viewed as presenting a potential conflict of interest.

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**References**

Ask the Keiki: Perceived Factors that Affect Asthma Among Adolescents from the Wai‘anae Coast Using Photovoice

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Abstract

Hawai‘i has among the highest prevalence of childhood asthma compared to all other states in the United States. Native Hawaiian children have a higher prevalence of asthma compared to other racial/ethnic groups in the state. Photovoice is a method in community-based participatory research that enables participants to use photos to express themselves and advocate on behalf of their community. In this study, students from the Wai‘anae Coast used Photovoice to identify perceived factors that affect asthma management. Seven students, ages 14 and 18, with self-reported asthma met virtually, with facilitators, after school once a week for four weeks. Students identified eight factors as positively or negatively impacting the students’ asthma and explained how these factors influence their health. The Photovoice results provided an in-depth understanding on the role a student’s culture and environment plays in asthma management. Continued efforts to develop asthma education programs tailored to address the specific factors that youth identify as impacting their asthma may be more effective in reducing asthma disparities. Future research should expand on the key themes identified in this study and include continued advocacy efforts among students to improve asthma-related outcomes in this community.

Keywords

Photovoice, participatory action research, community-based participatory research, adolescents, school-age children, asthma, asthma triggers, Native Hawaiian

Abbreviations and Acronyms

ACT = Asthma Control Test
COVID-19 = Coronavirus disease 2019
IRB = Institutional Review Board
SBHCs = School-based health centers
SBHEs = School-based health educators
WCCHC = Waianae Coast Comprehensive Health Center

Introduction

Hawai‘i has among the highest prevalence of childhood asthma compared to all other states in the United States. In 2016, asthma affected a quarter (25.3%) of Hawai‘i’s children under the age of 18. However, Native Hawaiian children have the highest asthma prevalence compared to other major racial/ethnic groups in the state. In 2012, more than half of Hawai‘i’s children diagnosed with asthma were Native Hawaiian, while the state population prevalence of Native Hawaiian children less than 18 years old is 34.2%. Therefore, compared to other racial/ethnic groups, Native Hawaiian children and their families will likely experience the greatest burden of asthma related outcomes.

Asthma prevalence among children also vary based on area of residence in Hawai‘i. In O‘ahu, children residing in the Nānākuli/Wai‘anae area are more likely to suffer from asthma than children living in other areas throughout the island. For example, the prevalence of childhood asthma was 17.5% in Nānākuli/Wai‘anae compared to 6.9% in the Kaimuki/Pālolo/Waikīkī area of O‘ahu. There is no cure for asthma, but it is a treatable disease. However, limited studies have examined asthma management from the child’s perspective. This knowledge is invaluable since children have autonomy in health decisions and are often responsible for managing their asthma. Therefore, a better understanding of managing and preventing asthma triggers and exacerbations are needed from the child and adolescent’s perspective, especially from populations with asthma-related disparities. Additionally, there is lack of research that explores how cultural values could have an impact on asthma management among children of Native Hawaiian ancestry. Traditionally, Native Hawaiian views of health include a balance between spirituality, environment (ie, land), and people and poor health was a result of an imbalance in the domains. Such information is important to address health disparities in a sustainable way from a strength-based approach.

The objective of this study was to identify supports and barriers to managing childhood asthma from the perspective of
children who reside on the Wai‘anae Coast of O‘ahu in the state of Hawai‘i. The Wai‘anae Coast communities of Wai‘anae and Nānākuli was selected because of the disproportionate rate of asthma among children. A large percentage of residents in these communities are also Native Hawaiian.

Methods

Study Design

Photovoice is a form of community-based participatory research that is helpful for promoting transformative change.\textsuperscript{11,12} Photovoice enables participants to use photos to express their concerns and empowers socially marginalized groups to advocate for themselves and their community.\textsuperscript{11,12} The study followed Wang’s suggested Photovoice procedures for researchers to: (1) select and recruit a target audience of community leaders; (2) recruit Photovoice participants; (3) introduce the Photovoice methodology and facilitate group discussion; (4) obtain informed consent; (5) determine a theme for photos; (6) distribute cameras and review instruction; (7) provide time to take photos; (8) discuss and analyze photos; and (9) develop a plan for disseminating photos and findings with community leaders and members.\textsuperscript{11} The students were also administered the Asthma Control Test (ACT) to assess the student’s asthma severity and control.\textsuperscript{13}

Recruitment

This study obtained approval by the Wai‘anae Coast Comprehensive Health Center (WCCHC) Institutional Review Board (IRB). Students in grades 6\textsuperscript{th} to 12\textsuperscript{th} who were a part of the WCCHC school-based health centers (SBHCs) student internship program(s) that are led by school-based health educators (SBHEs). Students who self-reported as having asthma were encouraged to participate. Informational packets were provided to all participating interns. Students submitted a signed parental consent and student assent to the SBHEs prior to the first session.

Enrollment

The Photovoice group met once a week for 4 weeks between February and March 2021. Due to the coronavirus disease 2019 (COVID-19) pandemic, the Photovoice group met virtually via Zoom for 1.5 hours during their internship time. Each student received a digital camera and an e-gift card for their commitment and participation.\textsuperscript{14}

Participants

The Photovoice group included 3 group facilitators (2 SBHEs and the OU) and 7 students. All students attended at least 3 sessions, with 3 students missing 1 session. The SBHEs assisted the first author by facilitating the group discussions by mitigating power dynamics and creating a safe place for each student’s voice to be heard and valued.

The 7 students identified as Native Hawaiian/Pacific Islander and were majority female (71%) between the ages of 14 to 18 years. The ACT indicated 71% had well-controlled asthma. All students reported they had missed at least 1 day of school per month pre-COVID due to their asthma.

Orientation and Data Collection

Table 1 outlines the details of each session. The first week covered background information, study relevance and future implications, Photovoice methodology, basic photography instruction, ethics, power, and risks. The research team provided all students with a digital camera but also allowed personal cell phones to capture photos. The students agreed on the following research questions: (1) What makes it easy to breathe? and (2) What makes it hard/difficult to breathe? Over the following 3 weeks, the students collected data by taking photographs that responded to the prompts. Data also included the student’s written and verbal reflections on photographs and experiences.

Analysis

Wang’s “Participatory Analysis” method was used for group analysis.\textsuperscript{13} The method is comprised of 3 steps: (1) selecting; (2) contextualizing; and (3) codifying. At the beginning of sessions 2 and 3, students selected 2 to 4 photographs that they had taken the previous week. Each student contextualized their photographs, explained why the photograph was taken, and what the photograph represented. The Photovoice group collectively identified major themes (ie, codifying) in the photographs and used the technique known as SHOWeD to discuss the photos’ significance. The mnemonic SHOWeD consists of 5 questions: S: What do you See here?; H: What is really Happening here?; O: How does this relate to Our lives?; W: Why does this situation, concern, or strength exist?; D: What can we Do about it?”.\textsuperscript{11} The group facilitators assured the analysis was a collective process and that the students agreed to the final themes.

Advocacy/Dissemination

Following the data collection and analysis, the Photovoice group decided to share their results by disseminating their photographs to family members, community advisory board members, the WCCHC IRB, and WCCHC Pediatricians. Additionally, upon completion of the study, the Photovoice group held a videoconference presentation with other community stakeholders, including researchers and Hawai‘i Department of Health staff and administrators, who are involved in pulmonary research, healthcare, and chronic disease management.
Table 1. Timeline and Description of the 4 Photovoice Sessions

<table>
<thead>
<tr>
<th>Session</th>
<th>Activities</th>
<th>Outcomes</th>
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| 1: Orientation meeting | • Answer demographic questionnaire and all survey tools  
• Introduce research questions for study  
• Discuss Photovoice methods  
• Distribute cameras, journals, timelines, and guidelines for photo-taking  
• Provide instructions regarding ethical camera use  
• Introduce SHOWeD mnemonic for writing narratives/captions | • Increase understanding among students about the prompts for taking photos  
• Increase knowledge and skills related to using a camera and tracking photos taken based on the prompts |
| 2: Review of photographs that represent supports and barriers | • Upload photographs to Jamboard  
• Instruct students in selecting 1-2 photos taken  
• Instruct students to share selected photos using the SHOWeD mnemonic for creating narratives/captions  
• Collectively identify the most significant supports and barriers among photos shared  
• Discuss potential audience for final session | • Identification of themes  
• Identification of potential audience members for final session |
| 3: Dissemination product | • Encourage and assist students in completing their dissemination product  
• Further discuss audience for final session | • Complete dissemination product  
• Finalize audience for final session |
| 4: Dissemination of findings to stakeholders | • Facilitate presentation of dissemination product by students | • Present findings to stakeholders |

Results

Of the 19 photographs reviewed, the Photovoice group agreed upon 8 dominant themes. The 4 support themes were ʻĀina (land), safe haven, alternative remedies, and community support. The 4 barrier themes for asthma triggers were physical activity, foods, environmental, and emotional.

Supports

ʻĀina. Most students identified ʻāina as a factor that helps their asthma. Five of the students discussed that being outside has a calming effect, which helps their breathing. Students identified several activities that helped relieve stress including going to the beach, looking at the sky, hiking, or sitting in their backyard to get fresh air. One student highlighted the connection between spirituality and ʻāina with their asthma: “Through spiritual connection to ʻāina, I find that I breathe easier. Though it seems in some ways counterintuitive to go outside when you are having issues with asthma, in my case it makes perfect sense… and through this process and our group discussions, it seems that this is a fairly common sentiment. I find peace in being outside: at the beach, hiking…there is something about being in ʻāina, being joined together with ʻāina that helps me become more at peace with the world around me, making it easier for me to deal with challenges that I face, such as anxiety and asthma” (Figure 1).

Safe haven. Three students discussed the importance of a sanctuary to help clear their minds and calm their body. Students described the various safe havens as specific locations such their bedroom but included pets and spiritual practices like meditation as additional supports. One student shared a photograph of the bedroom: “I took a picture of my bed, not because my bed is what helps but more of what my bed represents which is comfort. When I have my asthma attacks, I always cry, I never knew why but I think it’s honestly because of fear, just the sudden impact of something is wrong…I’ve learned however from these moments that what helps me is having someone there while also giving me space because I am freaking out. Having someone there gives me the security I need that if I get worse and just having that space to breathe helps me calm down and that I’m okay” (Figure 2). Another student shared: “Meditation has been a huge thing for me lately because I know I need to take breaks and focus on myself more. When I’m meditating my mind is clear and my body is calm, so my asthma isn’t bothering me and I’m at peace in every aspect of myself. My asthma bothers me a lot, but once I meditate, I really don’t even think about it” (Figure 3).
Alternative remedies. The use of asthma medication and natural products were important factors that improve their breathing. A student shared a photograph of a tea tree oil product saying: “My aunt has introduced Melaleuca to my family, and it has really help with my asthma…I use it to clean my room and bathroom because it makes it easier to breath” (Figure 4). Another student photographed the use of a nebulizer and shared there are other medical instruments for asthma “that help combat it”, but the most effective is their nebulizer.

Community support. The importance of community support was referenced by discussing how a strong supportive community could help asthma management when members are “motivated to fight to protect their children” (Figure 5).

Barriers

Physical activity triggers. Two students discussed the impact of physical activity on triggering their asthma. Both students described their experience as a “weight” and being “out of breath” when exercising, but wanting to participate in physical activities. For example, 1 student said: “I’m not sure if this sounds invalid but I’ve always struggled to keep up with normal exercising because of my asthma. I was always the last person to finish a lap or a simple exercise because I
Food triggers. One student shared that certain foods negatively impacts their asthma. This student loves chocolate, but the dairy causes a “really tight feeling in my lungs, I’m not exactly sure of the reason. I enjoy eating it and honestly, I wish that it didn’t have as much of a negative effect on my health as it does.”

Environmental triggers. Majority of students discussed various environmental factors that caused them to have asthma attacks. Students expressed that hot, dusty places (indoor or outdoor) make them feel like they are “suffocating” and “struggle to breathe.” However, 1 student mentioned that it is always hot in Nānākuli and the weather changes often. The student further stated: “It’s super hot outside of my house so I hate going outside because I know my asthma will flare up really bad. The day after it rains is always really bad because it’s so humid and the air is really thick and it gets really hard to breathe. That’s why I’d rather just stay in my room most of the time” (Figure 8). Additionally, other students expressed asthma exacerbations with changes in the weather and various plants that grow on the Wai’anae Coast. For example, 1 student said: “It is no secret that Hawai’i has a great deal of invasive
species in the ocean, on the land, and in our minds...when these plants flourish and seed, especially the California and Guinea grass, my family notices an unpleasant increase in unpleasant asthma” (Figure 9).

**Emotional.** Two students shared how emotions like feeling anxious or being overwhelmed can make it more difficult to breathe. One student shared that “meetings and situations” where there are “put in a position of responsibility and am in public” makes their asthma worse. This student shared they love people, but “figuring out how to deal with them is a little challenging,” therefore adding to the emotional stress. Another student discussed the importance of family stating: “When I think about my family and how far we’ve come- how far we’ve drifted, my breathing picks up, and I can feel myself spiraling. I love them with everything in me. It troubles me to know that perhaps they won’t always be there in the future” (Figure 10).

**Discussion**

This study used Photovoice methodology to empower and engage students in research on positive and negative factors that affect their asthma. To authors’ knowledge, this is the first Photovoice study that resulted in student-identified supports and barriers for asthma management. The study also provided a better understanding of the students’ experiences when managing their asthma symptoms, especially within the context of Hawai‘i, the specific community where they reside,
and their Native Hawaiian culture. Findings support childhood asthma management is multifactorial and involves individual, environmental, and community aspects that may be important to the control of their symptoms.15,16

There were 8 significant factors that were identified from the students’ photographs and group discussions. Students reported ‘āina, safe havens, alternative remedies, and a supportive community as significant supports that helped control their asthma symptoms. The concept of ‘āina is at the center of Native Hawaiian culture. There is a reciprocal relationship between ‘āina and people which contributes to their health.17 Therefore, Native Hawaiians value mālama ‘āina (land stewardship, caring for the land) and practices that foster connections to ‘āina as a critical indicator of health.17 The role that ‘āina plays for improving asthma control among Native Hawaiian children highlights prior findings that ‘āina is health (ie, maintaining healthy connections to ‘āina leads to healthy individuals).17

Students felt finding a safe haven, which could be a place, pet, or practice, where they can be alone and find comfort improved their breathing. For these students, a safe haven provides comfort and helps them deal with their asthma and aligns with research that shows that relaxation strategies reduce stress and may be associated with improvements in pulmonary function.18,19 It is well-known that medication adherence reduces asthma exacerbations.20 However, the use of natural products referenced by the students as an alternative or supplemental remedy for asthma treatment is less clear. A recently published literature review found all-natural products (eg, plant-based) improved asthma symptoms and could be used as a complementary treatment option.21 Furthermore, students reported the importance of community involvement and support in addressing childhood asthma. Engaging community members in the planning and implementation process of public health efforts is invaluable when addressing community needs.22

Students identified 4 well-known triggers that perceived to negatively impact asthma symptoms. These asthma triggers included strenuous exercise, foods, environmental triggers (eg, dust and hotter temperatures), and emotional stressors. While exercise has been found to improve asthma control in a prior study, 2 students reported participation in sports and recreational activities as a barrier.7 Students also recognized specific foods, like chocolate, can trigger asthma symptoms. This finding is consistent with studies that found diets high in dairy and sweets resulted in greater asthma symptoms.23,24 Dust and plant allergens are well-established environmental triggers, but the negative impacts on asthma symptoms with increases in the ambient temperature is less reported. Two recent studies have shown similar findings that hotter ambient temperatures are more likely to cause asthma exacerbations among children.25,26

Limitations
This study was not without limitations. The study’s processes may be transferable to other settings and populations based on the Photovoice methodology, but the findings may not be generalizable and might be specific to factors found within this community (eg, cultural beliefs, types of invasive species, and hotter climate temperatures). The intent of this Photovoice study was to gather data from a small number of students from the Wai‘anae Coast on their perspectives on factors that affect their asthma. The Wai‘anae Coast is unique in its geographic location, racial/ethnic makeup, and the social, political, and economic challenges this community experiences.27 This qualitative study is important in identifying culturally grounded interventions that address the asthma needs of children in this community. The number of Photovoice sessions was limited to 4 sessions via video teleconferencing. Findings may have resulted in different themes and subthemes if students were allotted more time to answer the research questions or if the sessions had been done in-person.

Implications for Schools and School Health Services
This study provided a better understanding of the individual factors that help or trigger asthma management among Native Hawaiian students from the Wai‘anae Coast. The experiences shared by this Photovoice group establishes a foundation for pediatric providers, SBHEs, and public health researchers. Asthma education needs to be provided to students with asthma as well as faculty and personnel at the schools. This research supports the need for teachers and staff to collaborate with students to identify and implement strategies that support asthma control. Identifying when a student needs a break to be alone and encourage the use of supports that improve asthma symptoms (eg, ‘āina, safe haven, alternative remedies) may be effective in improving asthma-related outcomes, including school attendance. Additionally, educating students that the SBHCs and SBHEs are possible safe havens on campus.

Most students emphasized the important role ‘āina plays in their asthma management. Further studies to explore how to best integrate the concept of ‘aina, and other indigenous practices, in interventions for Native Hawaiian students with asthma should be considered. For example, aquaponics systems and community gardens are ways students can connect with ‘āina and have shown positive health outcomes.28–31

This Photovoice study represents a small step toward transformative change in reducing asthma-related outcomes among students with asthma. Continued efforts are needed to create policy-level changes. A community-academic asthma group that includes the WCCHC, students with asthma, community board
members, and faculty at the schools may be effective in meeting the needs of the community. Partnerships with state-level representatives, public health organizations, and community-based organizations could help in identifying resources to implement asthma education programs for the Wai‘anae Coast community.

Conclusion

Youth from the Wai‘anae Coast continue to have disproportionate high proportion of asthma and poorer asthma-related outcomes. Photovoice was effective in helping students identify unique factors that affect their asthma management. It is also imperative that efforts are made to identify and address these unique factors that may be involved in improving asthma control among Native Hawaiian children. Future research is needed to further expand on the key themes identified in this study as well as continued advocacy efforts among students to improve asthma related outcomes in this community. Research should include more sessions, be conducted in-person, and include students who reside from the Wai‘anae Coast but are not a part of the internship program.

Conflict of Interest

None of the authors identify any conflict of interest.

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Style Guide for the Use of Native Hawaiian Words and Diacritical Markings

The HJH&SW encourages authors to use the appropriate diacritical markings (the ‘okina and the kahakō) for all Hawaiian words. We recommend verifying words with the Hawaiian Language Dictionary (http://www.wehewehe.org/) or with the University of Hawai‘i Hawaiian Language Online (http://www.hawaii.edu/site/info/diacritics.php).

Authors should also note that Hawaiian refers to people of Native Hawaiian descent. People who live in Hawai‘i are referred to as Hawai‘i residents.

Hawaiian words that are not proper nouns (such as *keiki* and *kūpuna*) should be written in italics throughout the manuscript, and a definition should be provided in parentheses the first time the word is used in the manuscript.

Examples of Hawaiian words that may appear in the HJH&SW:

| ‘āina | Mānoa |
| ali‘i | Māori |
| Hawai‘i | Moloka‘i |
| kūpuna | O‘ahu |
| Kaua‘i | ‘ohana |
| Lāna‘i | Wai‘anae |
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