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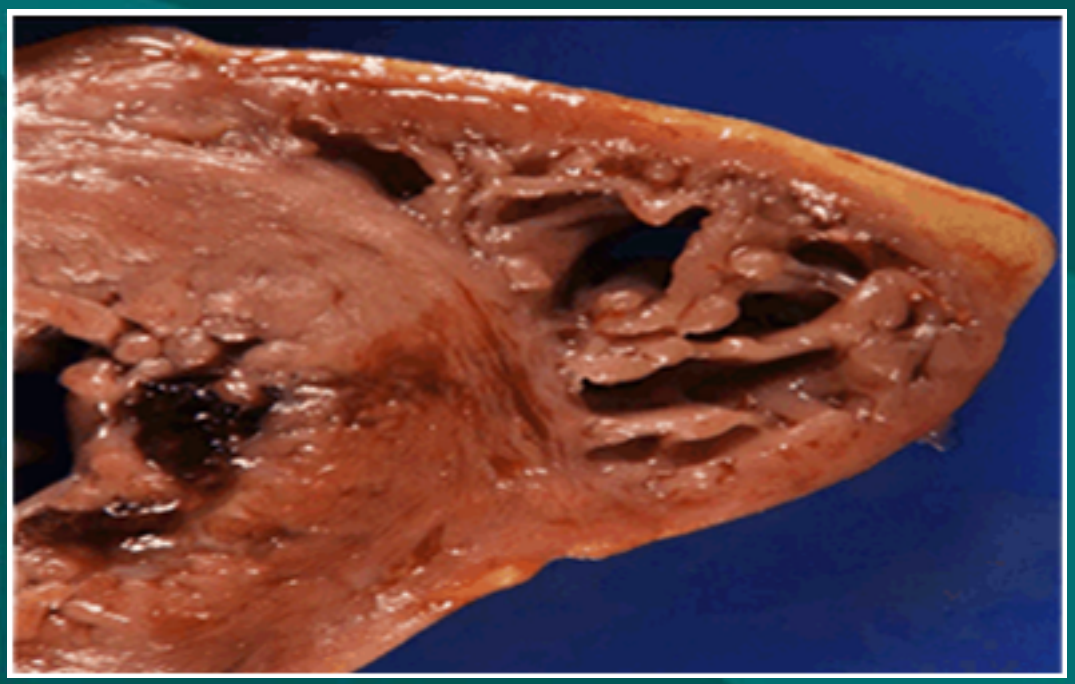

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Contents

Vol. 4, No. 2 (February 2013)

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REVIEW ARTICLE OPEN ACCESS

Camphor (Cinnamomum camphora), a traditional remedy with the history of treating several diseases

Rafie Hamidpour, Soheila Hamidpour, Mohsen Hamidpour, Mina Shahlari

ABSTRACT

Introduction: The purpose of this literature review is to gain knowledge of the long history, wide variety and extensive applications of camphor, both in traditional and modern medicine. Camphor (Cinnamomum camphora**) is obtained from the wood of camphor tree. It has been used for centuries, throughout the world as a remedy for treating variety of symptoms such as inflammation, infection, congestion, pain, irritation, etc. The studies have shown that some of the components of** Cinnamomum camphora **have suppressive and antimutagenic effect in number of human cancer cells without harming the healthy cells. In this paper our focus is on the use of camphor as a remedy for daily minor problems as well as reporting some information about the new applications of this traditional medicine to treat or prevent some serious life-threatening diseases sucha as cancer and diabetes. We hope to get the attention of researchers for conducting more studies on the effects of camphor on patients with memory and brain disorders as well.**

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Keywords: Camphor, Cinnamomum camphora**, Application, Treatment, Cancer**

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INTRODUCTION

Camphor (Cinnamomum camphora) is a white, crystalline substance with a strong odor and pungent taste, derived from the wood of camphor laurel (Cinnamomum camphora) and other related trees of laurel family. Camphor tree is native to China, India, Mongolia, Japan and Taiwan and a variety of this fragrant evergreen tree is grown in Southern United States; especially in Florida [1, 2]. Camphor is obtained through steam distillation, purification and sublimation of wood, twigs and bark of the tree [3]. There are many pharmaceutical applications for camphor such as topical analgesic, antiseptic, antispasmodic, antipruritc, anti-inflammatory, anti-infective, rubefacient, contraceptive, mild expectorant, nasal decongestant, cough suppressant, etc. [3–5]. Camphor is easily absorbed through the skin and can also be administrated by injection, inhalation and ingestion [3, 6].

Camphor has several chemical varieties, each with different essential oil compositions [1]. The leaf of Cinnamomum camphora contains camphor, as the main component along with cineol, linalool, eugenol, limonene, safrole, α-pinene, β-pinene, β-myrecene, α-humulene, p-cymene, nerolidol, borneol, camphene and some other components [1, 5, 7, 8].

APPLICATIONS

Camphor is a natural product with many applications in traditional and modern medicines. Traditionally, camphor has been used as a cold remedy for the relief of chest congestion and the treatment of inflammation related diseases such as rheumatism, sprains, bronchitis, asthma and muscle pain [9]. Camphor is usually prepared as a balm, oil or cream to relieve the pain and inflammation in joints and muscles. Camphor oil (20% camphor in cotton seed oil), when applied on the skin produces the feeling of coolness which is related to the stimulation of nerve endings sensitive to cold. Camphor activates some of TRP (transient receptor potential) channels like TRPV1, TRPV3, TRPM8 and inhibits TRPA1, causing warm sensation, excitation and desensitization of sensory nerves, relieving the pain, itch and irritation in applied area [4, 10–12].

There are many reports which prove that the use of camphor, solely or in combination with other treatments can be very effective for treating and preventing some serious diseases. A cancer study says that the use of camphor odor as a conditioning agent for the cancer cells of YC8 lymphoma in mice could have a suppressive effect on the growth of YC8 tumor, when it is combined with immunotherapy treatment [13]. Camphor also can be potential radiosensitizing agent in radiotherapy. Treatment with camphor prior to a radiation showed reduced growth of tumor volume [3].

A camphor based drug called 714-X, was developed by a Canadian researcher more than forty years ago and it is reported by some institutions, to be effective on the treatment of some patients with cancer, especially breast and prostate cancer [14]. Padma 28 is another multi compound herbal preparation, based on camphor formula which has shown to be effective against chronic inflammatory diseases. The result of a study indicates that Padma 28 has the ability to suppress the development of autoimmune diabetes in female non-obese diabetic (NOD) mice which could be an experimental model for type 1 diabetes mellitus in humans [15].

There are a number of applications for different parts of Cinnamomum camphora tree. The study of Cinnamomum camphora leaves extract (CLE) has shown the protective effects against DNA damage and biochemical changes in mice caused by atrazine (AT) which is one of the commonly used grass and weed herbicides [9]. The widespread usage of AT has caused contamination in the environment, resulting in genotoxicity and biochemical disturbances in animals and human cells. In this experiment, all the tested tissues which were treated with CLE showed a significant and time dependant decrease in chromosomal abnormalities and DNA damage [9]. Two ribosome inactivating proteins (RIPs), cinnamomin and camphorin are found in the seeds of Cinnamomum camphora; studies have shown their inhibitory effect on the cultured carcinoma cells [16]. In addition, cinnamomin has shown to have inhibitory effect on the growth of solid melanoma in the skin of the nude mouse [16]. The application of RIPs can be very significant in drug development and crop-plant technology due to their toxicity against viruses, tumor cells, insects and plant fungal pathogens [17].

One use of camphor is for carbon nanotubes (CNT). In recent years, the finding of CNT which are made of very light and strong fibers of one atom-thick sheet of carbons, rolled in tubes, have been very exciting developments with many applications in medicinal and industrial fields [18]. One of the most important uses of CNT is in the cancer treatments. Single wall CNT can be used as a drug delivery vehicle with high surface area to deliver chemotherapy drugs to the tumor cells and later, these purely carbon-made nanotubes can be excreted out of the body by biliary pathway without causing any toxicity [19]. Carbon nanotubes to this point are synthesized from purified petroleum products like methane, benzene, acetylene, etc. However, camphor can be the environment-friendly, alternative new option [18]. Camphor is a botanical hydrocarbon which is very cheap and can be easily cultivated without fear of shortages unlike petroleum products. Therefore, camphor is an excellent carbon source for the production of a high yield, high purity and high efficiency carbon nanotubes in future [20].

The essential oil of Cinnamomum camphora and some other aromatic camphor containing plants such as sage, rosemary and basil which are widely used in traditional medicines contain monoterpenes. The studies have shown that some essential oil components, especially monoterpenes have suppressive and anti-mutagenic effect in number of human cancer cells including colon cancer, gastric cancer, liver tumor, breast cancer, leukemia and others [21]. Most cancer chemotherapy treatments include highly cytotoxic drugs against proliferating cancer cells as well as healthy cells which can be harmful for the body. With a different mechanism of action, essential oils with their monoterpene components can have multiple pharmacological tumor-suppressive activities, mostly without such harm [21].

Many studies have been done about the various applications and benefits of camphor in pharmaceutical, industrial and environmental fields. Camphor has been used traditionally for many years as a remedy for the relief of pain, inflammation and irritation in the body and skin. Recent studies have focused on the role of camphor in preventing and curing serious and life- threatening diseases, when it is used purely or combined with other treatments. The study on some species in the Lauraceae family, shows that a number of extracts have significant antioxidant, anti-inflammation and anti-tumor activities [7, 21, 22]. These studies indicate that Lauraceae tree species and other camphor containing plants could have very important potential nutraceutical and pharmaceutical applications in the future [22], taking medicine just another step forward.

DOSAGE AND TOXICITY

Camphor like any other medication should be used for certain patients within the indicated dosages and contraindications [3]. The concentration of 3–11% has been approved by the FDA for topical use as a pain reliever and anesthetic [23]. Camphor and other terpenoid compounds do not accumulate in the environment since many soil bacteria like Pseudomonas putida readily degrade these compounds [24]. Although herbal medicines and essential oils have been widely used in folk and modern alternative medicine for many years and have shown to be very effective in curing many symptoms and diseases, the misuse of them can be very harmful for the body causing serious problems [25]. Camphor intoxication has been reported in humans and especially children but mostly because of accidental ingestion or exceeding the recommended amount [3].

CONCLUSION

Camphor has been used traditionally for many years, solely or in combination with other treatments for the relief of pain, inflammation and irritation in body and skin. It can also be very effective in treating and preventing some serious, life threatening diseases. Considering the growing number of cancer patients, Cinnamomum camphora and its components should be investigated further as a viable option in the treatment of different types of cancer. In addition, more studies on the application of camphor for patients with memory disorders and brain dysfunctions such as in Alzheimer's and autism are needed.

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Author Contributions

Rafie Hamidpour – Conception and design, Analysis and interpretation of data, Critical revision of the article, Final approval of the version to be published
Soheila Hamidpour – Analysis and interpretation of data, Critical revision of the article, Final approval of the version to be published
Mohsen Hamidpour – Analysis and interpretation of data, Critical revision of the article, Final approval of the version to be published
Mina Shahlari – Acquisition of data, Drafting the article, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CASE SERIES OPEN ACCESS

Pleomorphic adenoma of hard palate: A report of four cases

Sheela Chaudhari, Deepa Hatwal, Ashok, Vijay Suri

ABSTRACT

Introduction: Pleomorphic adenoma is the most common tumor of the salivary gland. The tumor most commonly arises in the parotid or submandibular salivary glands. Infrequently, it arises from minor salivary glands. Minor salivary gland tumors are mostly malignant. Case Report: We report four cases of pleomorphic adenoma of hard palate. All four cases range from 30–45 years. All were benign. Conclusion: Pleomorphic adenoma of minor salivary gland of hard palate is a rare benign tumor. Benign tumors at this site are more common then malignant ones. Complete evalutation of patients and complete removal of the tumor must be ensured so that tumor does not recur.

Keywords: Pleomorphic adenoma, Hard palate, Benign

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INTRODUCTION

Salivary gland tumors accounts for 2–6% of all head and neck neoplasms. Among all salivary gland tumors, pleomorphic adenoma is the most common tumor accounting for 60% of salivary gland tumors. Pleomorphic adenoma most commonly arises in major salivary glands which include parotid or submandibular glands. Rarely, pleomorphic adenoma arises from minor salivary glands. Among the minor salivary glands, hard palate is the most common site. Tumor in minor salivary glands are more likely to be malignant than their counterpart in major salivary glands. In contradiction to this only benign pleomorphic adenoma of minor salivary glands were found in our institute in last one year which prompted us to write this case series.

CASE SERIES

All four patients visited in the ENT outpatient department of our institute from where they were sent to department of pathology for fine needle aspiration cytology (FNAC). The clinical, radiological and histopathological features of the patients are given in Table 1.

Clinical features: Out of the four patients, three were female and one was male. Age ranged from 30–45 years. All patients presented with the complaints of

painless swelling over the hard palate for last 6–10 months. None of the cases had associated ulceration or any discharge (Figure 1). General examination of all patients did not reveal any significant findings. No history of chronic alcoholism or tobacco was given by any patient.

In all cases on examination there was a firm, smooth, non-tender, well circumscribed lesion in the middle of the hard palate. Overlying mucosa was healthy. No significant lymphadenopathy was found in the neck region.

Cytological features: Fine needle aspiration cytology of the lesion was performed and the smears were stained with Giemsa stain. Smears revealed the presence of bimodal pattern of epithelial cells and spindle cells in a myxoid stroma. The epithelial cells were of uniform size with round to oval nuclei, moderate amount of cytoplasm and well defined cell boundaries (Figure 2). A diagnosis of pleomorphic adenoma was made. All patients had surgery for removal of mass and tissues were sent for histopathological examination.

Histopathological features: Histopathological examination revealed a well encapsulated tumor outside which small amount of normal salivary gland tissue was seen (Figure 3A–B). Tumor tissue consisted of gland like structures and sheets of epithelial cells (Figures 4, 5) with myxoid (Figure 6A–B) and chondroid areas (Figures 4, 7). No mitotic figures were found. These features were consistent with diagnosis of a pleomorphic adenoma.

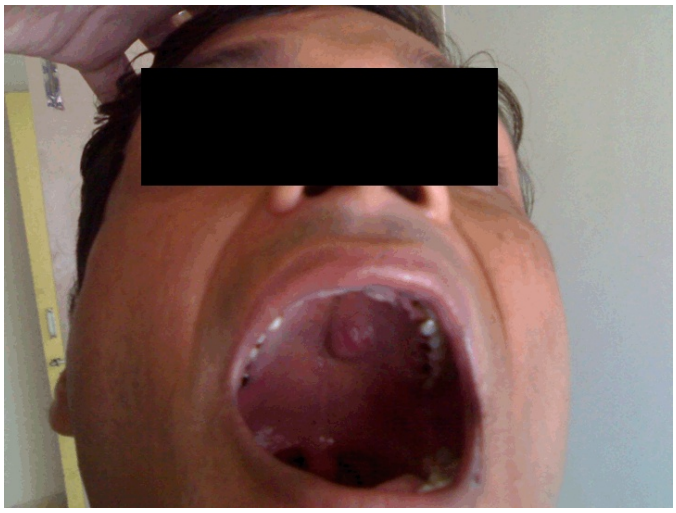

Figure 1: Hard palate showing a well-circumscribed growth.

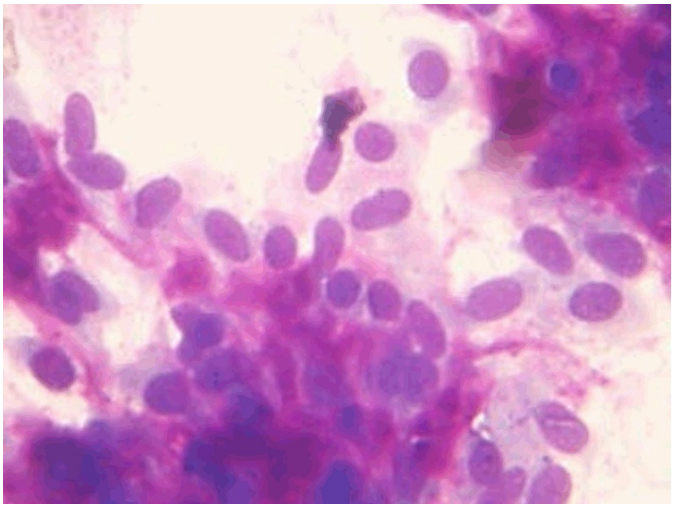

Figure 2: Photomicrograph of fine needle aspiration cytology showing clusters of epithelial cells with ovoid nuclei and blue cytoplasm overlying pink fibillary material (Giemsa, x40).

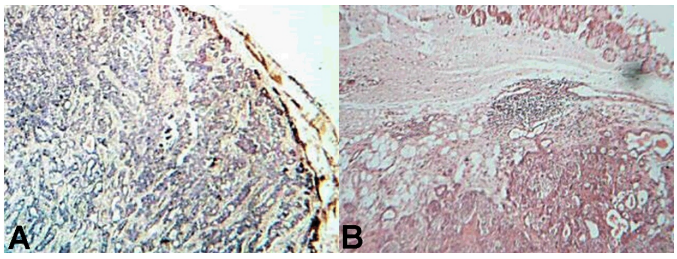

Figure 3: (A) Photomicrograph of histology section showing fibrous capsule (x50), (B) Photomicrograph of histology section showing fibrous capsule with tumor tissue and normal salivary gland tissue (H&E, x200).

Table 1: Clinical/radiological/pathological features for all patients.

	Clinical feature of swellings					Radiological feature MRI		Histopathological feature		
S.No.	Age (years)	Sex	Size (cms)	Pain	Consistency	Hard palate IND	Bone ERO/DIS	Epithelial pattern	Stroma	Cartilage
1.	32	F	1.3x1	+	Soft to firm	No	No	Gland, sheets	Fibromyxoid	+
2.	37	F	1.2x9	-	Firm	No	No	Glands sheets	Fibromyxoid	-
3.	39	M	1.4x1.0	+/-	Firm	No	No	Glands, sheets	Fibromyxoid	+
4.	42	F	1.5x1.0	-	Soft to firm	No	No	Glands, sheets	Fibromyxoid	+

Abbreviations: IND — Indentaton, ERO — Erosion, DIS — Distruction, F — Female, M — Male

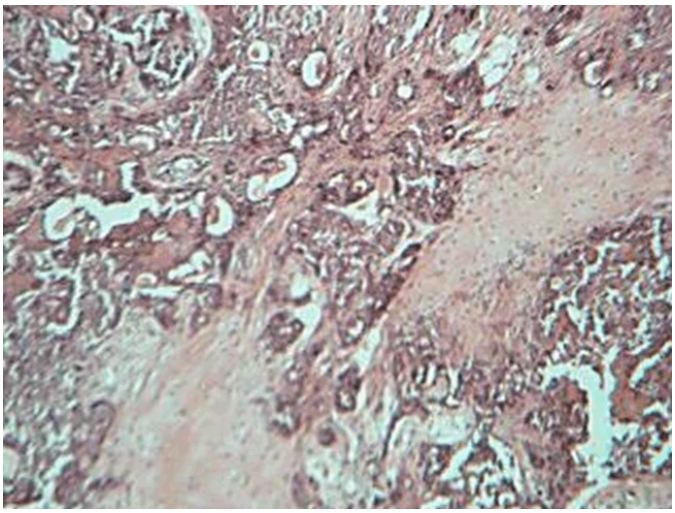

Figure 4: Photomicrograph of histology section showing tubular and glandular structures surrounded by myoepithelial cells and two chondromyxoid areas (H&E, x100).

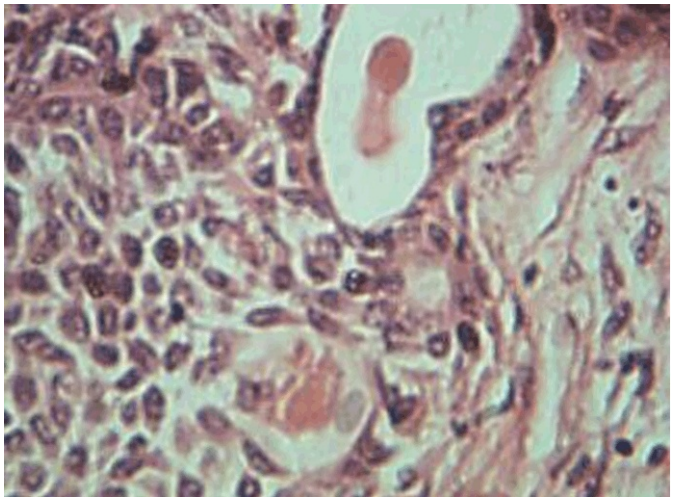

Figure 5: Photomicrograph of histology section showing epithelial cells in sheets and few gland like structures (H&E, x400).

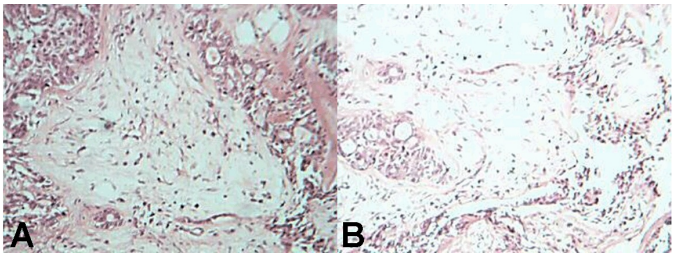

Figure 6: Photomicrograph of histology section showing epithelial glands and myxoid stroma, (H&#, A: x40, B: x100).

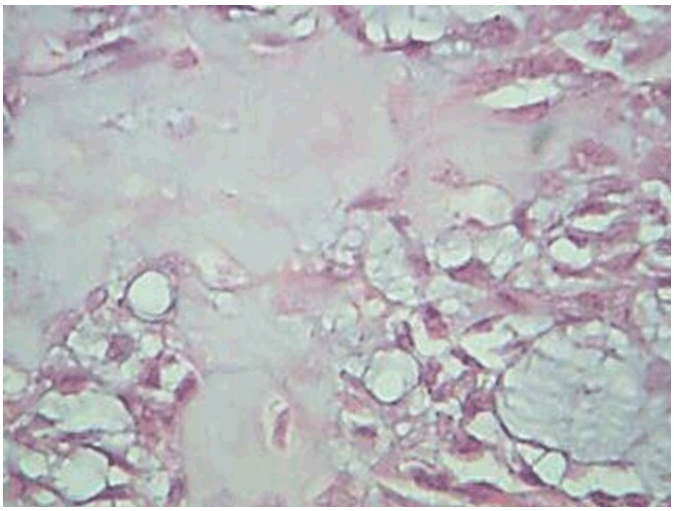

Figure 7: Photomicrograph showing chondroid change and epithelial like structures (H&E, x400).

DISCUSSION

Tumors of minor salivary glands constitute 2–4% of the head and neck tumors and about 10% of tumors of oral cavity [1]. About 22% selivary gland tumors arise in the minor salivary glands [2–4]. Most common tumor of minor salivary glands is a pleomorphic adenoma [5]. Most common site for minor salivary gland pleomorphic adenoma is the hard palate followed by lip, buccal mucosa, floor of mouth, tongue tonsils, pharynx, retro molar area and oral cavity [2, 4]. In our study of four cases we found all arising from the palate.

Intraoral pleomorphic adenoma appears as slow growing painless mass, usually in the fourth or fifth decade [6]. Study done by Moshy et al. showed slight predilection for older patients [5]. On the other hand, study done by Waldrom et al. revealed that tumor was more common in younger age group [7]. These findings suggest a possible variation in the presentation of intraoral minor salivary gland tumors in different population. In our study, the tumor was found in fourth and fifth decade.

Most studies have shown that minor salivary gland tumors are more common in females than male [8]. Male to female ratio is 1:1.8 to 1:2.4 [9, 10]. The tendency for female predominance is especially marked in benign tumors [9]. Our case series of four cases also underscores this fact.

Study done by Moshy et al. and Waldron et al. shows predominance of malignant neoplasm over benign ones [5, 7], while other studies show higher number of benign salivary gland tumors than malignant ones [8, 11]. This difference may be because these studies are from major referral centre which receives all the referred and complicated cases [8, 11]. Therefore, we can conclude that relative incidence of benign versus malignant tumors reflects the character of each institute. Likewise, in our study also we found all cases to be benign, as our institute is having many referral hospitals nearby.

Pleomorphic adenoma of the palate although being a benign tumor has a high recurrence rate. Lack of well defined fibrous capsule is a feature most commonly associated with a high recurrence rate [7].

The diagnosis of pleomorphic adenoma is suspected on the basis of history and physical examination and confirmed with cytology and histopathology. Comupted tomography scan and magnetic resonance imaging are helpful in providing information about the size and

extension of the tumor to the surrounding structures. This tumor usually does not recur after adequate surgical removal.

CONCLUSION

We presented four cases of pleomorphic adenoma from one of the rare site of minor salivary gland tumor. These tumors should be evaluated thoroughly for any extension into deeper tissues. During surgery complete removal must be ensured so that tumor does not recur.

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Author Contributions

Sheela Chaudhari – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Deepa Hatwal – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Ashok – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Vijay Suri – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CASE REPORT OPEN ACCESS

Primary oral myiasis: A clinical presentation in cerebral palsy

Abdul Ahad G Khan, Kanchan Milind Shah

ABSTRACT

Introduction: Myiasis is the infestation of live human and vertebrate animal with dipterous larvae which feed on the host's dead or living tissue. The most common anatomic sites for myiasis are the nose, eye, lung, ear, anus, vagina and more rarely, the mouth. Incidence of oral myiasis as compared to that of cutaneous myiasis is less as the oral tissues are not permanently exposed to the external environment. Case Report: A case of oral myiasis in the anterior maxillary region in a 12-year-old male with mental retardation and cerebral palsy caused by the larvae (maggots) of Musca Nebulo **(Family Diptera) (common housefly) is reported. The treatment consisted of manual removal of the larvae by topical application of turpentine oil (used for medicinal purpose) and surgical debridement of the oral wound. Conclusion: Preliminary measures taken to maintain good oral and general hygiene can prevent as well as treat myiasis.**

Keywords: Oral myiasis, Cerebral palsy, Common house fly, Maggots

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INTRODUCTION

The term myiasis is derived from Latin word 'muia' which means 'fly' and 'iasis' means 'disease'. It is a pathology caused in human and animal tissue by the larvae that evolve to a parasite. The term was coined by Hope in 1840 [1]. Myiasis was defined by Zumpt as the infestation of live human and vertebrate animals by dipterous larva, which at least for a certain period feed on host's dead or living tissue, liquid body substances or ingested food [2]. Myiasis frequently occurs in rural areas infecting livestock and pets such as dogs and cats. In humans, myiasis prevails in unhealthy individuals frequently found in undeveloped and tropical countries.

Myiasis can be classified clinically as primary (larvae feed on the living tissue) or secondary (larvae feed on dead tissue) [3]. Myiasis can also be classified depending on the condition of the involved tissue into accidental myiasis (larvae ingested along with food), semi-specific (larvae laid on necrotic tissue in wounds) and obligatory myiasis (larvae affecting the undamaged skin) [4]. Further classification can be based on the site as cutaneous, external orifice, internal organs and generalized [5].

The most common anatomic sites for myiasis are the nose, eye, lung, ear, anus, vagina and more rarely, the mouth [6]. Incidence of oral myiasis is less as compared to cutaneous myiasis, as the oral tissues are not permanently exposed to the external environment [7].

Oral myiasis was first described by Laurence in 1909 [8]. Conditions leading to persistent mouth opening

along with poor hygiene, suppurative lesions, severe halitosis and facial trauma may predispose the patient to oral myiasis [9]. It has been reported among epilepsy patients with lacerated lips following a seizure, incompetent lips and thumb sucking habits [10], advanced periodontal disease [11], at tooth extraction sites [12], fungating carcinoma of buccal mucosa [5] and patient with tetanus with mouth propped open to maintain his airway [13] .

Mouth breathing during sleep, cerebral palsy, alcoholism, mental handicap and hemiplegia may facilitate the development of myiasis. Other contributing factors are poor public and personal hygiene [14].

CASE REPORT

A 12-year-old male was referred to our department with a complaint of pain, fever and diffuse swelling over the upper lip, predominantly on the right side extending up to the lower eyelid causing inability to open the eye. He had a history of exfoliation of upper anterior teeth secondary to fall about one month back. Medical history revealed severe mental retardation with cerebral palsy. He had a habit of mouth breathing. Severe halitosis was also present.

On local examination the swelling was firm, tender and seemed to be of inflammatory origin. Lips were incompetent. Gingival inflammation was generalized but more severe in the anterior maxillary region having progressed to periodontitis. General as well as oral hygiene was very poor. Maxillary right central and lateral incisors were missing but their sockets were bare and not in the healing phase (Figure 1). White worm like creatures were moving in and around the empty sockets, the gingiva of the maxillary left central incisor and the vestibular area labial to the maxillary incisors (Figures 1, 2) that lead to a provisional diagnosis of oral myiasis.

Our line of management began with parenteral hydration, administration of antibiotics (augmentin + metronidazole), vitamin supplements and manual removal of maggots by means of tweezers and oil of turpentine that was done for three consecutive days. Approximately, 30–35 maggots were removed (Figure 3) during each of the two days from the wound, due to which the local condition of the tissue improved progressively. The use of ivermectin [3] was not needed for therapy as local debridement of tissue after removal of the maggots resulted in uneventful healing (Figure 4).

DISCUSSION

Musca Nebulo is the most common Indian housefly. They are seen in abundance in human dwellings and are very active during summer and rainy season [15]. The life cycle of a fly begins with egg stage followed by the larvae, pupa and finally the adult fly. The conditions required for egg laying and survival of the larvae are

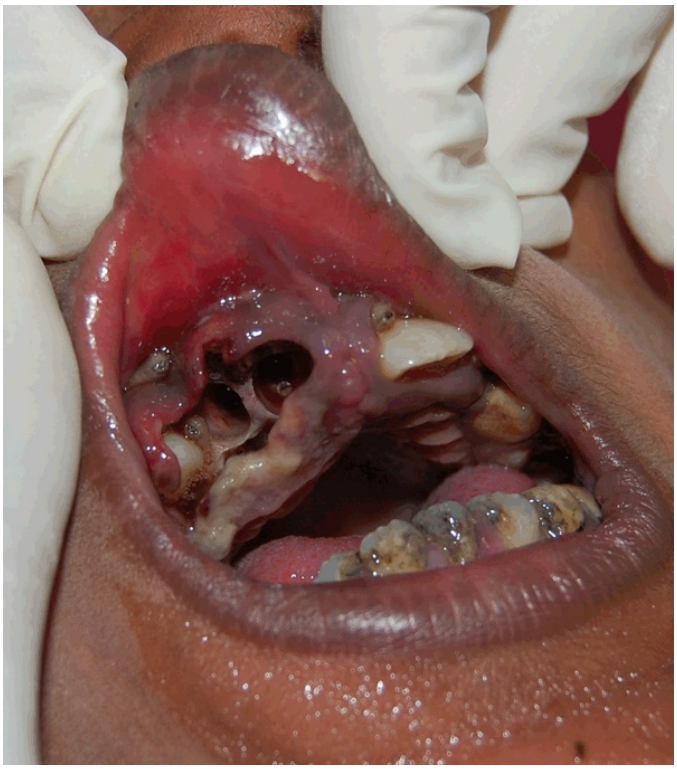

Figure 1: Pretreatment photograph depicting the maggots moving around the wound.

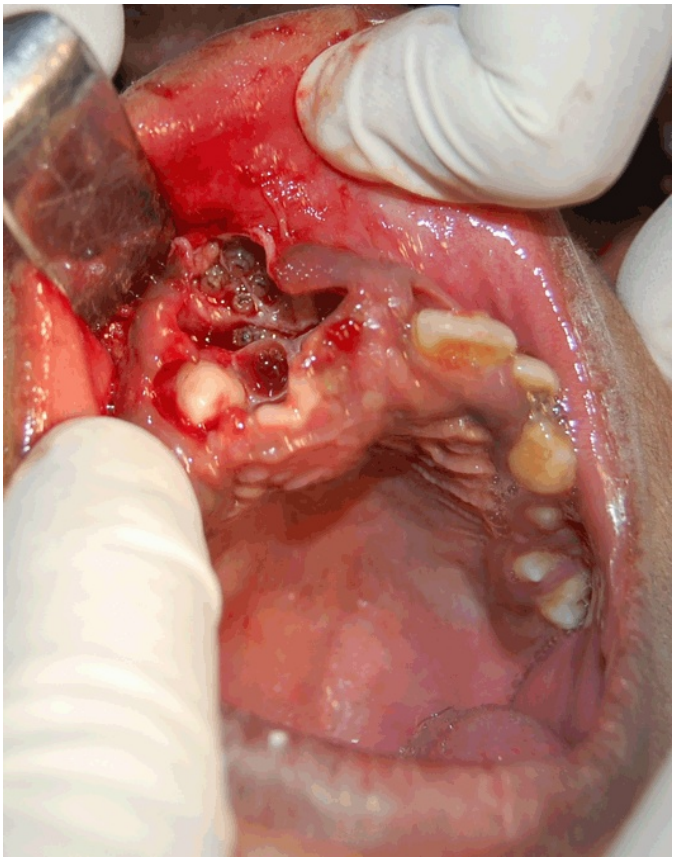

Figure 2: Pretreatment photograph depicting the maggots deeply embedded in the wound.

Figure 3: Removed maggots in a single sitting for two consecutive days.

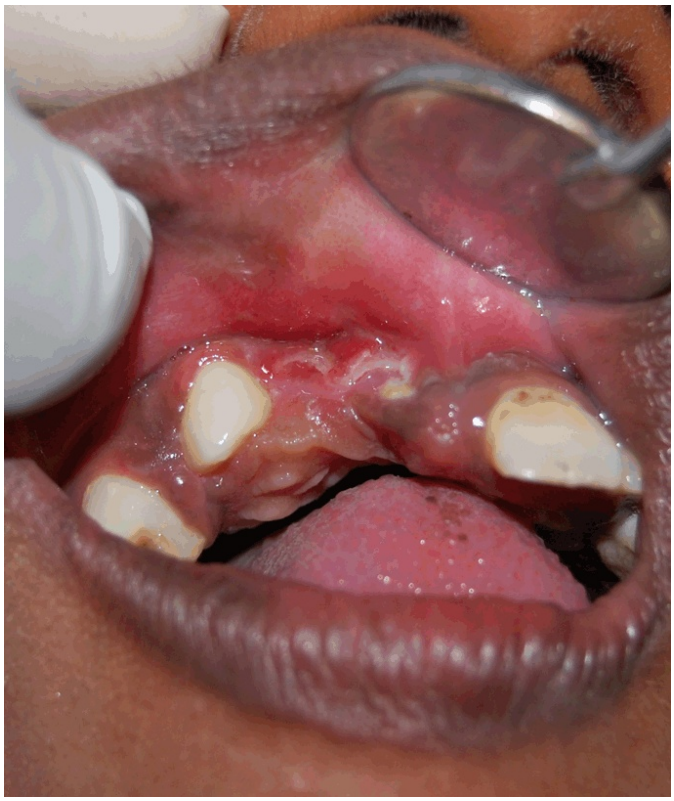

Figure 4: Healed wound after complete removal of maggots.

moisture, necrotic tissue and suitable temperature. Thus, wounds, open sores, scabs, ulcers contaminated with discharges facilitate the same. The developmental transition via the larval stage requires an intermediate host. In our case an empty socket and existing periodontitis contributed to the mechanical support and suitable substrate and temperature for the survival of the larvae. The stage of larvae lasts for six to eight days during which they are parasitic to human beings. The larvae have backward directed segmental hooks with which they anchor themselves to the surrounding tissue. They are photophobic and tend to hide deep into the tissues for a suitable niche to develop into pupa [15]. The present case also showed the larvae burrowed deep inside the socket (Figure 2). Manual removal of larvae from the host is difficult due to presence of these hooks. So, when multiple maggots are detected, as observed in our case, elimination can be achieved with agents like turpentine oil or topical irritants such as ether, chloroform, olive oil, calomel, iodoform and phenol mixture. These larvae release toxins to destroy the host tissue [16, 17]. Proteolytic enzymes released by the surrounding bacteria decompose the tissue and the larvae feed on this rotten tissue [18]. The infected tissue frequently releases a foul smelling discharge [16]. The necrotic ulcer around the teeth sockets and intense halitosis seen in the present case is suggestive of the destruction caused by toxins released by the larvae.

The role of antibiotics in oral myiasis is only supportive so as to prevent the development of infection secondary to the presence of necrotic area providing a nidus to the growth and multiplication of pathogenic microorganisms and should be discontinued when the wound has healed completely.

CONCLUSION

Myiasis could be prevented by maintaining good oral and personal hygiene, cleaning and covering the wounds and by educating the susceptible population about basic sanitation measures to be taken. Special care needs to be taken by the guardian/care-taker in medically compromised, dependent patients as the patients are unable to maintain their basic hygiene.

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Author Contributions

Abdul Ahad G Khan – Conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Critical revision of the article, Final approval of the version to be published
Kanchan Milind Shah – Conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CASE REPORT OPEN ACCESS

Acute presentation of deep vein thrombosis in Achilles tendon rupture: A case report

Rafik Yassa, Kiran Singh Kandola, A Sharma

ABSTRACT

Introduction: Rupture of the Achilles tendon is relatively common. High incidence of thromboembolism during the first eight weeks after Achilles tendon rupture has been reported in literature. The use of venous thromboembolism prophylaxis after an Achilles tendon rupture is controversial. Delayed deep vein thrombosis (DVT) is a well-known complication after operative and non-operative treatment of Achilles tendon rupture but it has not been reported as an acute presentation following achillies tendon rupture. Case Report: We report a case of DVT presenting acutely after Achilles tendon rupture. It presented with soft clinical signs. High index of suspicion was required to diagnose DVT. The authors could not find similar association between the two clinical problems reported in literature. Conclusion: We recommend that risk assessment should be undertaken in any patients following lower limb injury even if it is not a major injury and adequate thromboprophylaxis should be started according to the risk rating when deciding on immobilization of the lower limb.

Keywords: Achilles tendon, Rupture, Venous thrombosis

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INTRODUCTION

Venous thromboembolism is a recognized complication after lower limb injury [1]. Rupture of the Achilles tendon is relatively common and deep vein thrombosis (DVT) is a well-known complication after operative or non-operative treatment of Achilles tendon rupture.

High incidence of thromboembolism during the first eight weeks after Achilles tendon rupture has been reported [2]. To the best of the authors' knowledge acute presentation of DVT after Achillis tendon rupture has not been reported in literature. The use of venous thromboembolism prophylaxis after an Achilles rupture is controversial. The rates of reported deep vein thrombosis (DVT) range from 6.3–34%. There is no agreement regarding prophylactic therapy after an Achilles tendon rupture [3].

We present a case of acute presentation of Achilles tendon rupture with concomitant DVT. Such a diagnosis should be considered when assessing for a ruptured achilles tendon and planning the method of treatment.

CASE REPORT

A 44-year-old female suffered forced dorsiflexion of the ankle, when she lost her footing on a step. Medical history revealed discectomy three years back. She was on regular use of paracetamol and non-steroidal anti-inflammatory medication (ibuprofen) for residual low back pain. She was a smoker of 1 pack/day. On clinical examination in the emergency department, Achilles tendon was found to be tender on palpation. She was unable to stand on tip-toes. Simmonds-Thompson test was positive. The test involves having the patient lie prone on an examination bed or kneeling on a chair with both feet extending past the end of examining table or chair, on squeezing the calf muscles on the affected side by the examiner's hand, the foot will demonstrate plantar flexion if the tendon is intact. On the other hand, if tendon is ruptured, the foot will not demonstrate plantar flexion. This test may lose its accuracy one week after injury. A below knee back slab was applied with foot in equinus as a initial temporary measure.

Three days later, the patient was seen in the orthopedic clinic to decide on the definitive treatment, when it was noticed that the swelling had became diffuse involving the whole leg from the knee downwards which was more than that expected for Achilles tendon rupture alone. She also had slight calf tenderness. As this raised the additional possibility of deep venous thrombosis (DVT), a venous duplex scan as well as an ultrasound scan of the tendon was performed in the same day. The ultrasound scan revealed Achilles tendon rupture 5 cm above its insertion into the calcaneus with a gap of 1 cm. The venous duplex scan also surprisingly revealed DVT of the popliteal and posterior tibial veins (Figure 1).

Because of the concomitant ipsilateral DVT, the decision was made to treat her non-operatively with a dorsal plaster of paris slab, first in equinus for three weeks, then followed by a further three weeks with the ankle in a neutral position. On hemotological advice, low molecular weight heparin in the therapeutic dose (1.5 mg/kg body weight) was administered for six weeks, followed by warfarin for six weeks at therapeutic levels (INR 2–3). The patient had a follow up in the orthopedic clinic till she had a complete recovery and healing of the Achilles tendon rupture but she continued her regular visits to the hematology clinic for about six months thereafter. There was no reported evidence of pulmonary embolism or any complication from either the DVT or its treatment.

DISCUSSION

Deep vein thrombosis is a recognized complication after an injury to the lower limb [1]. A prospective study reported high incidence of thromboembolism during the first eight weeks after an acute achilles tendon rupture [2], but no study has reported acute presentation of DVT after an achillis tendon rupture. Clinical diagnosis

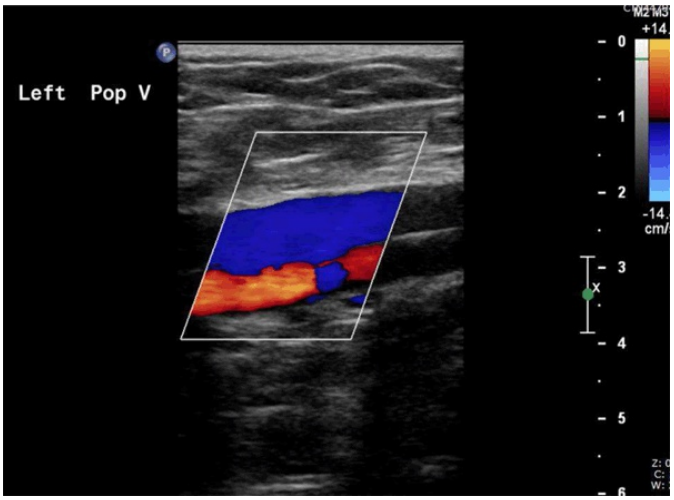

Figure 1: Duplex Scan Showing DVT of the popliteal and posterior tibial veins.

in the acute setting is difficult and unreliable due to pain, ecchymosis, tenderness and swelling from the trauma itself. In addition, ultrasonographic diagnosis in the acute setting involves some technical problems due to pain, hematoma and soft tissue oedema.

The DVT, pulmonary emboli (PE) and cardiovascular events are serious complications after orthopedic surgery. The risk of thromboembolism after acute achillies tendon rupture has been demonstrated in several studies [4–6]. Venous thromboembolism was demonstrated in 34% patients, two months after an acute total Achilles tendon rupture [2]. In a large retrospective study by Patel et al., the overall rates for DVT and PE after Achilles tendon ruptures were 0.43% and 0.34%, respectively. Age older than 40 years, congestive heart failure, history of DVT or PE, obesity, and whether a patient had surgery; did not predict occurrence of DVT or PE (Table 1) [3].

However, after injury or surgery to the lower limbs the clinical diagnosis of DVT is fairly challenging. This is because of the combined factors of pain, tenderness, swelling and immobilization. Objective methods are required for DVT diagnosis. Moreover, the majority of DVTs are asymptomatic and the true incidence is unknown. A proportion of the clots in subtle DVTs can be complicated by PE [2].

The use of colored duplex sonography (CDS) to detect DVT, especially after trauma and surgery has been challenged [7, 8]. In a recent study CDS was performed eight weeks after the injury to avoid the technical problems and ambiguity [2].

Thromboprophylaxis for lower limb injury was evaluated in several previous studies. In patients immobilized with plaster casts due to lower leg injuries, Kujath et al. demonstrated a reduction in the incidence of DVT when prophylactic treatment was used [9]. On the other hand, Patel et al. believe that routine use of anticoagulation might be unwarranted (Table 2) [3].

In United Kingdom, NICE provide clear guidelines for the thromboprophylaxis after minor surgery, lower limb injury and lower limb plaster casts (Figure 2) [10].

Table 1: Venous thromboembolism (VTE) risk assessment

Patients who are at risk of VTE

Medical patients

- If mobility significantly reduced for ≥3 days, or
- If expected to have ongoing reduced mobility relative to normal state + any VTE risk factor

Surgical patients and patients with trauma

- If total anaesthetic + surgical time >90 minutes, or
- If surgery involves pelvis or lower limb and total anaesthetic + surgical time >60 minutes, or
- If acute surgical admission with inflammatory or intra-abdominal condition, or
- If expected to have significant reduction in mobility, or
- If any VTE risk factor present

VTE risk factors

- Active cancer or cancer treatment
- Age >60 years
- Critical care admission
- Dehydration
- Known thrombophilias
- Obesity (BMI >30 kg/m^2)
- One or more significant medical comorbidities (for example, heart disease, metabolic, endocrine or respiratory pathologies, acute infectious diseases, inflammatory conditions etc.)
- Personal history or first-degree relative with a history of VTE
- Use of hormone replacement therapy
- Use of oestrogen-containing contraceptive therapy
- Varicose veins with phlebitis

Table 2: Risk of bleeding assessment

All patients who have any of the following are at risk of bleeding:

- Active bleeding
- Acquired bleeding disorders (such as acute liver failure)
- Concurrent use of anticoagulants known to increase the risk of bleeding (such as warfarin with INR >2)
- Lumbar puncture/epidural/spinal anaesthesia within the previous four hours or expected within the next 12 hours
- Acute stroke
- Thrombocytopenia (platelets <0. 75x10^5/mm^3)
- Uncontrolled systolic hypertension (≥230/120 mmHg)
- Untreated inherited bleeding disorders (such as hemophilia or von Willebrand's disease)

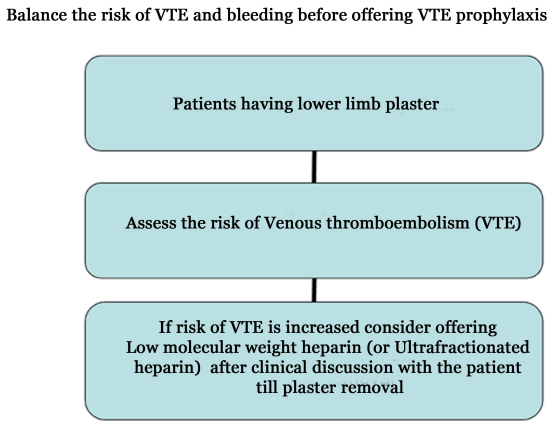

Figure 2: NICE algorithm for DVT prophylaxis in lower limb plaster. Venous thromboembolism, Low molecular weight heparin, Ultrfractionated heparin for patients with renal failure.

CONCLUSION

Deep vein thrombosis may presents acutely with lower limb injury. Risk assessment should be undertaken in any patient following lower limb injury even if it is not a major injury. Adequate thromboprophylaxis should be started, if it is indicated, when deciding on immobilization of the lower limb, either above or below knee immobilization, especially in plaster.

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Author Contributions

Rafik Yassa – Conception and design, Acquisition of data, Drafting the article, revising it critically for important intellectual content and final approval of the version to be published
Kiran Singh Kandola – Conception and design, Acquisition of data, Drafting the article, revising it critically for important intellectual content and final approval of the version to be published
A Sharma – Conception and design, Acquisition of data, Drafting the article, revising it critically for important intellectual content and final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CASE REPORT OPEN ACCESS

Intestinal hydatidosis in unusual location: A case report

Nagarekha Kulkarni

ABSTRACT

Introduction: Hydatid cyst in the small intestine is extremely rare and only a few cases have been reported in literature. Case Report: A 60-year-old male presented with abdominal pain in the periumbilical region, nausea and fever. Hematological investigations revealed neutrophilic leukocytosis with eosinophilia. Ultrasound scan revealed multiple hypoechoic cysts in the small intestine. Serological tests were positive for hydatid disease. Surgical laparotomy was done and patient was treated with antihelminthic drugs. Histological diagnosis of hydatidosis was made. Conclusion: Intestinal hydatidosis is a rare entity and can be considered as one of the differential diagnosis of acute abdomen or abdominal mass.

Keywords: Hydatid cyst, Echinococcus, **Hydatidosis**

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INTRODUCTION

Hydatid disease (Echinococcosis) is a parasitic disease caused by larval stage of Echinococcus granulosus, in which humans are accidental intermediate host and animals are both definitive and intermediate host [1]. It is endemic in the cattle grazing areas particularly Australia, New Zealand, Middle East, India, Africa, South America and Turkey. Humans get infected by consuming raw vegetables, undercooked animal products and water contaminated with the hydatid ova. Humans can also become infested after close contact with animals such as cats and dogs [2]. This disease is a major cause of morbidity and mortality in many parts of the world. In humans, liver is the most frequently affected organ followed by the lungs. The traumatic or spontaneous rupture of a hydatid cyst may put the patient's life at risk due to anaphylaxis by cyst contents. If the patient survives, the development of new hydatid cysts may lead to a difficult to treat situation [3]. Reports on cases of hydatid cysts involving the intestine are exceedingly rare. In Lyon's report of a series of 241 cases in North America the intestine was involved in only one case. There have been several cases reported as abdominal hydatids, but there was no mention of the intestinal involvement [4].

CASE REPORT

A 60-year-old male living in a rural area presented to surgical out patient department with abdominal pain, nausea and fever since 15 days. The abdominal pain was localized in the periumbilical region. The pain was intermittent and colicky with intense pain of 10–15 minutes followed by 2–3 hours of remission. The pain aggravated after intake of heavy meals and during sleep. He had nausea 2–3 hours after meals. The fever was

mild and continous without any chills and rigors. Patient gave a history of similar dull, aching abdominal pain since one year. He had taken treatment from local practitioner who treated symptomatically and the diagnosis was not made. There were no past history of trauma, jaundice, alteration in bowel and bladder habits or surgery. The family history was unremarkable. Patient was nonalcoholic and nonsmoker. He had frequent contact with farm animals. The patients gave a past history of hypertension which was controlled by antihypertensive drugs. Physical examination revealed a mass in the periumbilical region measuring 8x6 cm. The mass was firm with restricted mobility. Clinically, a tumor in periumblical region was suspected. There was no hepatosplenomegaly. Bowel sounds were normal. The vital signs, laboratory results and radiographic findings of the patient are given in Table 1. Hematological investigations showed leukocytosis with neutrophilia and eosinophilia. Serological tests indirect hemagglutination assay (IHA) and enzyme linked immunosorbant assay (ELISA) were positive for hydatidosis. Abdominal ultrasound scan revealed multiple hypoechoic cysts in the small intestine. Surgical laparotomy was done on the same day of admission. Intraoperative findings confirmed the diagnosis of intestinal hydatidosis. During surgery the exact location of the cyst was identified and the surrounding tissues were protected by covering them with cetrimide soaked pads. The cyst along with 15 cm of small intestine (jejunum) was removed and cetrimide was injected into the abdominal cavity. After 10 minutes cetrimide was evacuated and the abdominal cavity was irrigated with isotonic sodium chloride solution. Care was taken to ensure no spillage occurred to prevent seeding and secondary infestation. The abdominal cavity was then filled with isotonic sodium chloride solution and closed. Peroperatively liver and spleen were normal. There were no peritoneal deposits. Antibiotics were used prophylactically for surgery. Postoperative period was uneventful and the patient was discharged after 15 days.

Table 1: The patient's vital signs, laboratory and radiographic findings

Vital Signs	
Pulse Rate	92/minute
Blood Pressure (mmHg)	130/80
Respiratory Rate	24/minute
Temperature (°C)	38.5
Laboratory Findings	
Hemoglobin (g/dL)	10 (Normal: 13.6–17.2 g/dL)
RBC Count ($\times 10^5$/mm^3)	3.2 (Normal: 4.5–5.5)
Haematocrit	31% (Normal: 35–50%)
WBC count ($\times 10^3$/mm^3)	19x10^3 (Normal: 4–11)
Differential Count:	
Neutrophils	90%
Eosinophils	08%
Lymphocytes	02%
Total Bilirubin (mg/dL)	0.9 (Normal: 0.1–1.2)
Alanine Transferase (IU/L)	22 (Normal: up to 40)
Aspartate Transferase (IU/L)	34 (Normal: up to 40)
Alkaline Phosphatase (IU/L)	67 (Normal: 60–170)
Serological (Test): IHA	Positive
ELISA	Positive
Radiographic Findings	
Chest X-ray	Normal
Abdominal X-ray	Normal

Abbriviations: IHA — Indirect hemagglutination assay, ELISA — Enzyme linked immunosorbant essay

Albendazole 400 mg orally, twice daily was started after the surgery and continued for three months post-operatively. Postoperative follow-up was for one year. The complete blood counts and liver enzyme evaluation were performed at biweekly intervals for three months and then every four weeks to monitor for albendazole toxicity. Serology assay (ELISA and IHA) and ultrasonography was performed at 3rd, 6th, 12th month intervals as screening to monitor for recurrence of disease. No recurrence was noted after one year.

Fifteen centimeters of small intestine (jejunum) was sent for histopathological examination. Macroscopic examination of resected portion of jejunum showed multiple cysts in the wall of the intestine. The largest cyst was 8 cm in diameter. Cut section along the antimesentric border revealed multiple translucent cysts containing nonviscous fluid (Figure 1). Histological examination revealed features of hydatid cyst involving mucosa, submucosa and muscularis layer surrounded by chronic inflammatory infiltrate (Figure 2). The diagnosis of intestinal hydatidosis was made.

Figure 1: Gross appearance of hydatid cyst involving small intestine (jejunum).

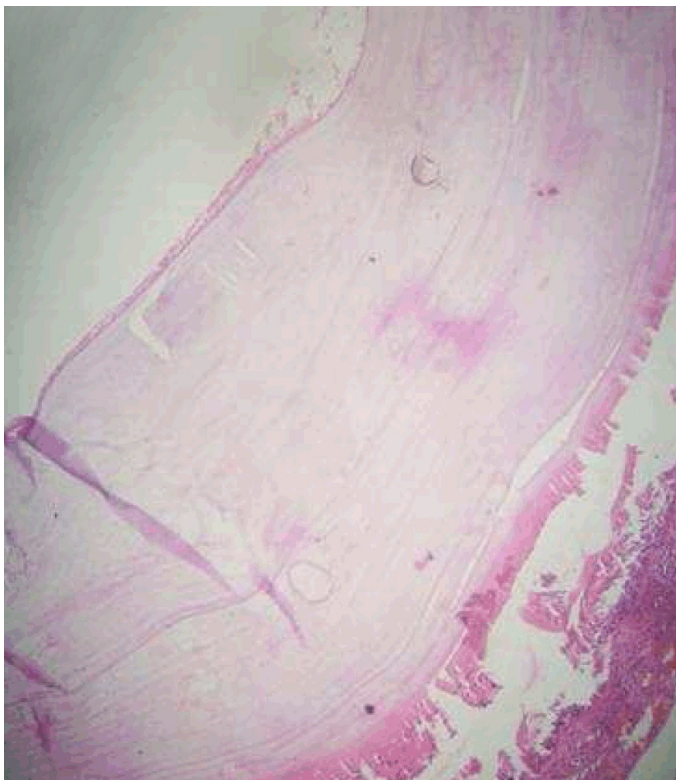

Figure 2: Histopathology of cyst showed features of hydatid cyst involving mucosa, submucosa and muscularis layer surrounded by chronic inflammatory cells (H&E stain, 200x).

DISCUSSION

Hydatid disease is caused by Echinococcus granulosus, Echinococcus oligarthus and Echinococcus multilocularis. Echinococcus granulosus is most common and represents an important medical problem in many countries. Humans are accidental intermediate hosts infected by ingestion of food contaminated with eggs shed by dogs or foxes and are common in rural areas. Eggs hatch in the duodenum and enter the mesenteric venules and become lodged in the capillary filter bed in various organs except for hair, teeth or finger nails [5]. The most important site is liver (70%), lung (15%), kidney (3%), spleen (4%), cerebrum (2%) and heart (0.02–2%). The other sites that have been reported include bone, pancreas, breast, ovary, scrotum, thyroid gland, inguinal canal and soft tissue [6, 7]. Hydatid cyst in the small intestine is extremely rare and only a few cases have been reported [8]. Majority of cases of hydatid disease come from rural areas or people who have settled in urban centers after spending life in villages. Most of the people acquire the disease during their childhood but they are asymptomatic until late adulthood because of the slow growing nature of the cysts [9].

Hydatidosis affects human beings without predilection for age or sex. In a study of 2,013 patients by the Tunisian Surgical Association, the mean age was 32 years [9]. Ayadi–Kaddour et al. reported a case in which the patient was 66 years old [5]. Clinical manifestations of hydatidosis in humans are variable. Most patient seem to tolerate the infection for extended periods with out any symptomatology or they may suddenly show dramatic and acute symptoms [10]. In a case reported by Najih et al., the patient presented with intermittent attacks of abdominal pain, abdominal distention, recurrent vomiting and nausea [11]. Kusaslan et. al. reported a case in which the abdomen was tense and tender, especially in the lower quadrant with guarding and rebound tenderness. Sometimes the clinical findings may mimic other abdominal disorders [9]. For an unusual location the diagnosis can be difficult; all abdominal cystic lesions including mesenteric, pancreatic, gastrointestinal duplication, ovarian cysts and lymphangioma must be considered in the differential diagnosis. Pain is the most common symptom of hydatid disease. Pain may be of acute onset if the cyst ruptures or it may be continous, dull, aching in nature. Fever may occur if there is secondary infection [12]. Although the physical findings are varied,

the diagnosis is best made by a combination of hematological, biochemical and serologic laboratory investigations and by radiographic examination. Ultrasonography is the first line of screening test for abdominal hydatidosis. Computed tomography (CT) scan has become an extremely useful and valuable diagnostic tool in the management of patients with hydatidosis [7]. In the present study CT scan was not done as the diagnosis was made on ultrasound examination. The treatment of choice is surgical excision of the cyst alone or en bloc with a part or the whole involved organ with adjuvant therapy to prevent the recurrence. In this case the cyst was removed along with 15 cm of small intestine (jejunum) because of adhesions. Anaphylactic shock due to spontaneous or traumatic rupture or during surgery is a rare phenomena but with severe complications [13]. In the present case we did not come across anaphylactic reactions as cetrimide was injected into the abdominal cavity. Medical treatment with antihelmenthic drugs such as albendazole is used preoperatively and post-operatively. Some authors report better results when medical treatment is used along with surgical treatment [1]. Our patient was prescribed albendazole (400 mg orally, twice daily) after surgery and it was continued for three months postoperatively. Morbidity and mortality in patients with perforated hydatid cyst are higher than in those with nonperforated cysts. Mortality rate is variable, ranging from 0–20% in published reports of perforated cyst [5].

CONCLUSION

To conclude, intestinal hydatid cyst is a rare entity and can be considered as one of the differential diagnosis of acute abdomen or mass per abdomen. Serological, ultrasonography and CT should be performed before any invasive procedures. The complications of the disease can be potentially devastating. The disease still continous to challenge the public and health professionals with its rare presentations.

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Author Contributions
Nagarekha Kulkarni – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor
The corresponding author is the guarantor of submission.

Conflict of Interest
Authors declare no conflict of interest.

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CASE REPORT OPEN ACCESS

Pulmonary alveolar microlithiasis

Anita Flynn, Anuradha D Agastyaraju

ABSTRACT

Introduction: Pulmonary alveolar microlithiasis is an important cause of diffuse parenchymal lung disease and is characterised by calculospherites in the alveolar spaces. Usually occurring in the 4th to 6th decade. Sex distribution is roughly equal. Clinical course is progressive and fatal. About 300 cases of pulmonary alveolar microlithiasis have been reported in literature. Case Report: Here we report a case of a 56-year-old male who presented with progressively increasing breathlessness. Imaging revealed characteristic "sandstorm" appearance on X-ray. A transbronchial lung biopsy confirmed the diagnosis of pulmonary alveolar microlithiasis. Conclusion: The etiology of pulmonary alveolar microlithiasis is unknown and there is no definite medical treatment. Hence, therapy is mostly empirical.

Keywords: Breathlessness, Alveoli, Calculospherites

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INTRODUCTION

Pulmonary alveolar microlithiasis is a rare disease of unknown etiology. It is characterized by intra-alveolar deposition of calcific concretions in the absence of abnormal calcium metabolism [1]. Pulmonary alveolar microlithiasis is a slowly progressive disease leading to respiratory insufficiency associated with cyanosis, clubbing and pulmonary hypertension [1]. There is no satisfactory therapy for this condition [1]. This disease was first described by Malpighi et al. in 1686 [2]. Puhr et al. gave this disease an appropriate name in 1932 [2]. In 1947, Mariani et al. described the clinical, functional and radiological features of this entity [2].

The diagnosis of pulmonary alveolar microlithiasis depends on the characteristic "sandstorm" picture in chest X-ray and the finding of alveolar calculospherites on histology.

CASE REPORT

A 56-year-old male presented in the pulmonology out patient department with the chief complaint of progressively increasing breathlessness for the past one year accompanied by dry cough which was not relieved by medication. There was no history of fever, hemoptysis or chest pain. Patient was a non-smoker. There was no history of tuberculosis within the family members or any known history of pulmonary disease. On physical examination, the patient had no clubbing, cyanosis, lymphadenopathy or peripheral edema.

Cardiac auscultation was normal. Auscultation of lungs revealed coarse crepitations in all the lung fields. Pulmonary function test (PFT) revealed mild restrictive ventilatory defect with reduced total lung capacity. HIV was non-reactive. Routine lab investigations were non-contributory. Chest X-ray showed bilateral finely granular calcified densities predominantly in the middle and lower lobes (Figure 1).

Fiberoptic bronchoscopy, was performed bronchoalveolar lavage and transbronchial biopsy. The bronchoalveolar lavage fluid was negative for acid fast bacilli and fungi. Calculospherites were not found. The transbronchial biopsy was sent for histopathology. Microscopy showed alveolar spaces filled with deeply eosinophilic lamellated calcospherites (Figure 2). The alveolar walls showed a scattered chronic inflammatory infiltrate and mild fibrosis. A histologic diagnosis of pulmonary alveolar microlithiasis was made. The patient was started on steroids and came for subsequent follow up visits for a period of six months. Minimal improvement in the repeat X-ray was documented. The patient was subsequently lost to follow-up.

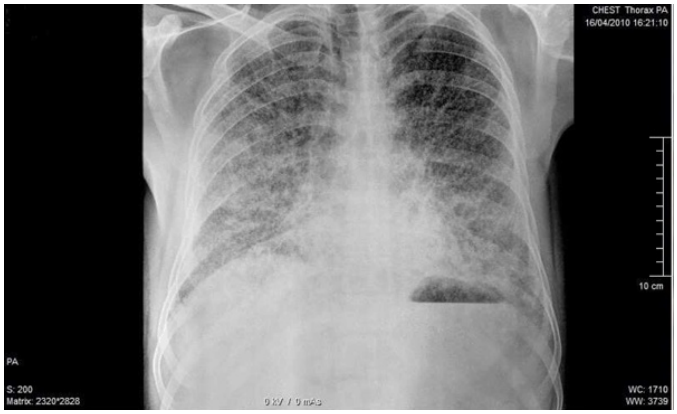

Figure 1: Chest X-Ray showing typical “sandstorm” appearance.

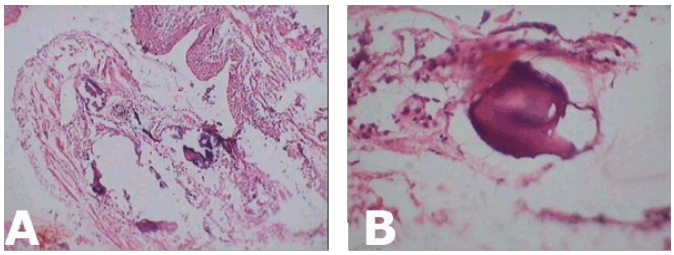

Figure 2: (A) Lamellated calculospherites scattered in the lung parenchyma, (H&E, x100), (B) Intra-alveolar calculospherites (H&E, x400).

DISCUSSION

Pulmonary alveolar microlithiasis is an autosomal recessive lung disease characterized by filling of the lung alveoli by calcospherites [3]. It is caused by mutations in the SLC34A2 sodium dependant phosphate transporter that is normally expressed in the type II alveolar macrophages [4]. Half of the cases, however, are familial [5]. In the remaining, the cause is obscure [5]. The average age of presentation is between the 4th and 6th decade. The M:F sex ratio is roughly equal [3]. Patients are usually asymptomatic or present late, usually with cough or breathlessness [5]. In advanced cases, hemoptysis, pneumonia, end-stage interstial pneumonia and cor pulmonale may develop [3]. Pulmonary function tests mirror the stage of the disease [6]. The progress of these patients is variable. Most patients suffering from a protracted disease which is usually progressive and fatal [2, 5]. There is no effective treatment except lung transplant [5].

Imaging findings are described as a typical “sandstorm” appearance with fine granular opacities throughout the lungs, worse at the bases [2]. The airways appear unremarkable on bronchoscopy; however, concretions may be aspirated in the BAL fluid [2].

At autopsy, the lungs are extremely difficult to cut. Cut surfaces are hard to gritty with numerous 0.2–0.3 mm, yellowish brown, sand-like particles [5]. Histologically, the alveoli are filled with concentric lamellated basophilic calcospherites measuring 50–200 microns in size [3]. The calcospherites are composed of calcium, phosphate or calcium hydroxyapatite with traces of magnesium, silicon, iron and aluminium [3]. The central cores of the microliths are PAS-positive while the surrounding laminations are PAS-negative and von Kossa positive [3, 6]. In our case, no attempt was made to do any chemical analysis of the calcareous material. The calculospherites are sometimes birefringent, giving a Maltese cross pattern with polarization [6]. The differential diagnosis of calcific bodies include pulmonary blue bodies, corpora amylacea, metastatic calcificaltion and heterotopic ossification [3].

CONCLUSION

The etiology and pathogenesis of pulmonary alveolar microlithiasis is still largely unknown. Hence, there is no effective and definitive medical therapy and treatment is mostly empirical. Radiology may be confusing, keeping in mind the numerous differential diagnoses. It will only be possible to develop effective treatment of the disease after its etiology has been fully established.

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Author Contributions

Anita Flynn – Conception and design, Acquisition of data, Drafting the article, Critical revision of the article, Final approval of the version to be published
Agastyaraju D Anuradha – Acquisition of data, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest
Authors declare no conflict of interest.

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CASE REPORT OPEN ACCESS

Delayed presentation of psoas abscess previously misdiagnosed as deep venous thrombosis: A potentially devastating error

Victor Kong, George Oosthuizen, Musa Mthethwa, Kriban Reddy, Damian Clarke

ABSTRACT

Introduction: Psoas abscess is an uncommon condition seen only occasionally in daily surgical practice. Often, its clinical presentation is non-specific, frequently causing diagnostic uncertainty. Late presentation, delayed recognition and inappropriate management of this condition can often precede eventual diagnosis, especially in resource poor settings. Case Report: We describe the case of a 22-year-old man who presented with a large psoas abscess which unfortunately had been misdiagnosed and treated as a deep venous thrombosis. Conclusion: This unusual case highlights the difficulties often encountered by clinicians when assessing and managing a patient with a psoas abscess. The diagnosis of a psoas abscess remains challenging. Clinicians must always remain vigilant to this diagnosis. Early recognition, confirmation with imaging and swift drainage remains the key to optimising patient's outcome.

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INTRODUCTION

Psoas abscesses are rare. Clinical diagnosis is extremely difficult because presentation is often subtle and non-specific. This case report focuses on a patient who presented to our surgical unit with a large psoas abscess after the condition had been wrongly diagnosed and mistreated as deep venous thrombosis at another rural hospital.

CASE REPORT

The patient was a 22-year-old male with no previous known medical history. He presented to another rural hospital with complaints of gradual onset of a vague discomfort in his right hip. He also noted a slight swelling of his right foot and reduced mobility of the right hip, which remained unchanged after weight bearing. He had no previous history of a similar presentation and no recent history of trauma. The patient's HIV status was unknown at the time and he reported no previous tuberculosis contact. He had been treated by a non-specialist doctor at another rural hospital who diagnosed a deep venous thrombosis (however, an ultrasound Doppler facility was not

available at this hospital) and the patient was commenced on therapeutic dose of subcutaneous enoxaparin, 40 mg daily for one week. The patient did not experience any improvement and continued to complain of an increasing pain in the affected side. Subsequently, he did not attend his follow up appointment at his local hospital due to the lack of available transportation. However, after two weeks, he managed to return to the hospital and was eventually referred to our surgical unit for a second opinion.

On examination, the patient appeared emaciated and was in obvious discomfort. He was apyrexial, with a pulse of 100/min and a blood pressure of 135/65 mmHg. An ill defined tenderness in the right iliac fossa was noted, extending along the inguinal ligament, with no palpable mass. A slightly swollen right foot with the swelling extending to the calf was also noted (Figure 1). The range of motion in his right hip was slightly reduced. His leukocyte count was $1.8x10^4/mm^3$. His remaining laboratory tests and a plain radiographs of the chest, abdoman, pelvis and hips were unremarkable.

A psoas abscess was suspected and an urgent ultrasound scan was performed, as a computed tomography (CT) scan was not available due to other concurrent emergencies. Ultrasound scan revealed a large psoas abscess that had spread beyond the inguinal ligament, with marked compression of the femoral vein. A doppler imaging study of the lower limb was performed and did not reveal a deep venous thrombosis. The patient was commenced on intravenous antibiotics (co-amoxiclav, 1.2g, q.i.d.) and was taken to the operating theatre immediately for open drainage. An oblique incision was made in the right iliac fossa and a muscle splitting manoeuvre was performed in order to access the retroperitoneal space (Figure 2). A large pus collection was found (approximately two litres) and this was drained immediately (Figure 3). The wound was thoroughly irrigated and a drain was left in situ (Figures 4, 5). The patient made an uneventful recovery and was discharged on day 5. Subsequently, he was tested positive for HIV and was commenced antiretroviral treatment.

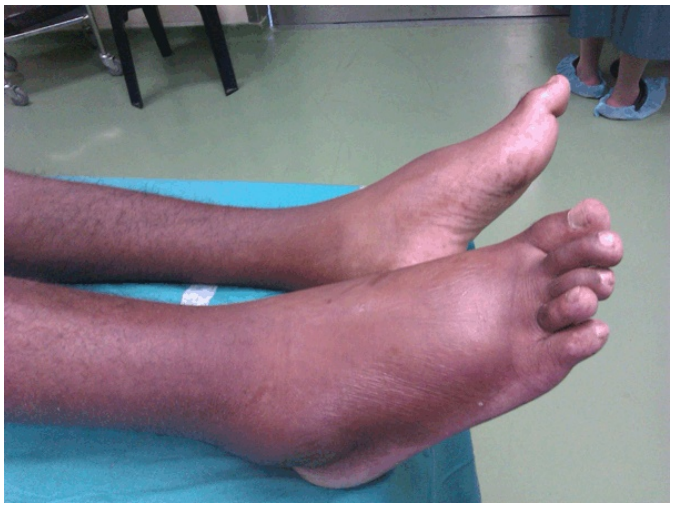

Figure 1: Patient presented with swelling of right foot extending to the calf.

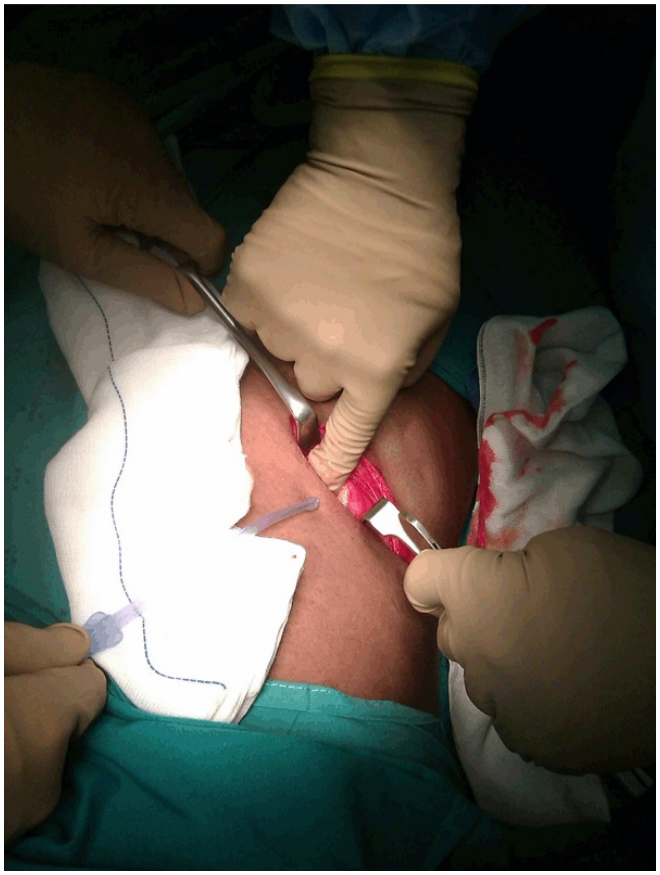

Figure 2: Dissection to access the retroperitoneal space.

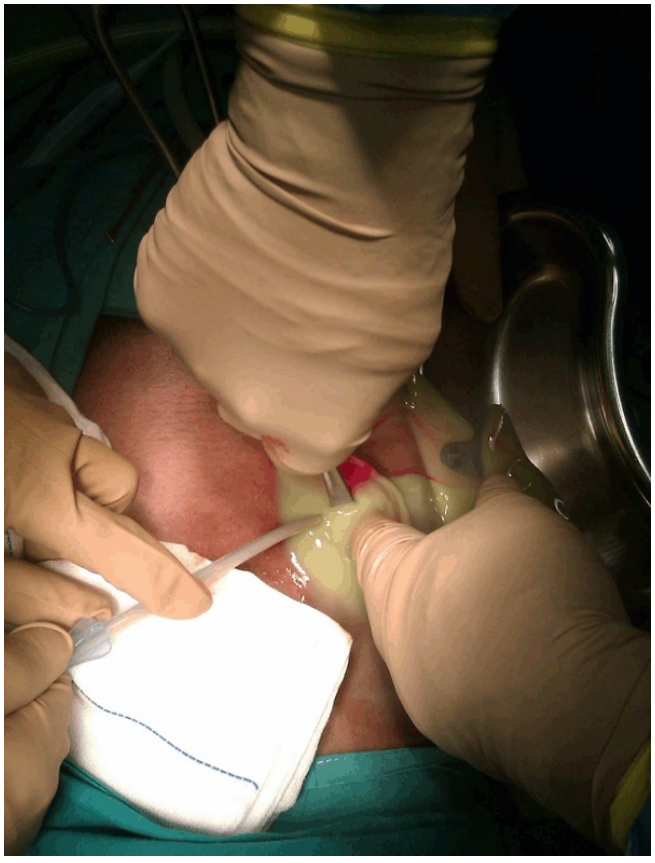

Figure 3: Retroperitoneal space showed, large gush of thick pus collection.

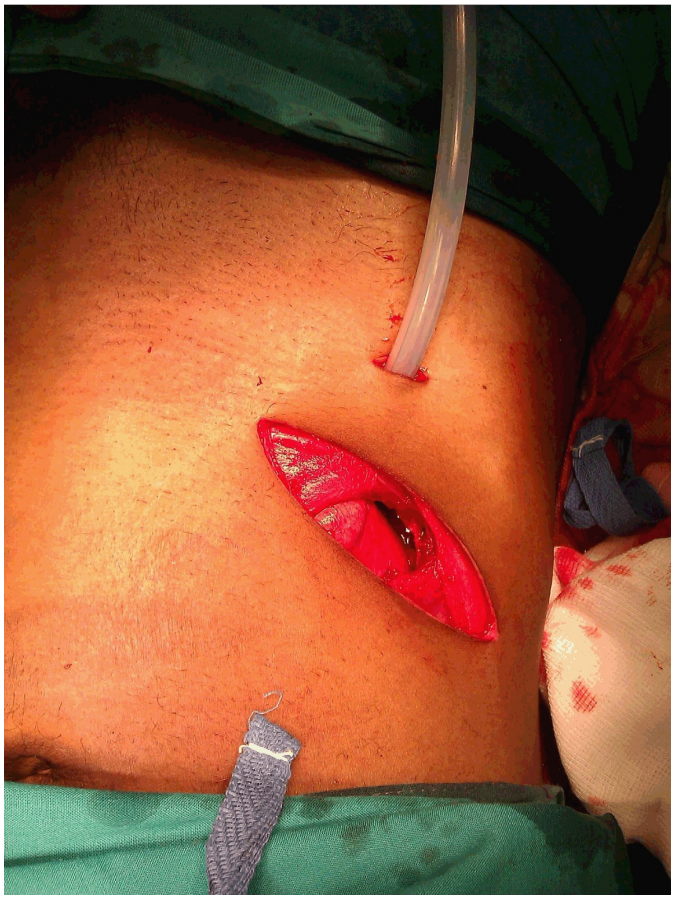

Figure 4: Drain left in situ.

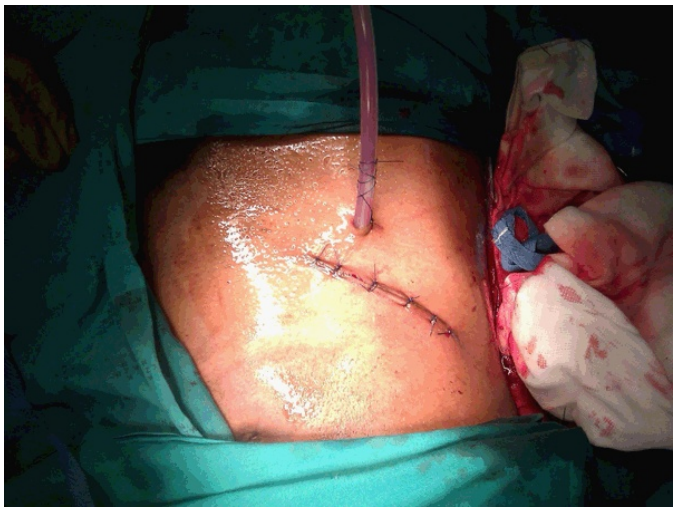

Figure 5: Wound apposed with drain in situ.

DISCUSSION

Psoas abscess was first described by Mynter et. al. in 1881 and was originally referred to as 'psoitis' [1]. A psoas abscess is a relatively rare condition and has a worldwide incidence of approximately 12 cases per 100,000 per year [2]. It carries a morality rate of up to 20% [3]. A psoas abscess is usually classified as either primary, if there is no obvious source of infection, or as secondary, if an obvious source of infection is present (such as Crohn's disease, perforated appendix etc). There is a worldwide variation in its etiology. For example, in Africa, 99.5% of all psoas abscesses are primary, compared with only 61% in the United States and 18.7% in Europe [4]. In over 88% of primary psoas abscess cases, Staphylococcus aureus is the causative organism. However, in the developing world, Mycobacterium tuberculosis is a common cause of psoas abscess, especially in cases of to tuberculosis of the spine [4].

This case highlighted several important issues. First, the patient obviously presented with extremely vague symptoms that were not suggestive of any particular condition. It is well known that clinical presentation of psoas abscesses is often vague and non-specific (especially in children), and this can lead to a delay in diagnosis and treatment [5–7]. The classic triad of fever, back pain and limp is only present in less than 30% cases [8], and there is no single clinical sign that is specifically diagnostic of this condition. In this particular case, it could be argued that the initial diagnosis of a deep venous thrombosis was reasonable, but the diagnosis could not be realistically supported because of a lack of adequate imaging equipment at the rural hospital. In the absence of a diagnostic imaging, there was a failure to consider other possible different diagnoses, and this resulted in inappropriate management and subsequent delay in referral to our surgical unit, which was potentially disastrous.

Patients in developing countries, especially in rural areas, often have difficulty in accessing even the most basic healthcare facilities. In this case, a lack of transportation alone meant that the patient was not able to attend the follow up appointment, and this delayed the referral for another two weeks. The patient somehow managed to return at a later date and at this point a swift referral was made to our unit. It is important that clinicians working in rural settings in the developing world remain vigilant and open to consideration of this important diagnosis. A correct diagnosis continues to hinge on a high index of suspicion, with judicious use of imagining (where available). Early referral for surgical opinion and early drainage remains the cornerstone of good management that will optimise the outcome for these patients.

CONCLUSION

Psoas abscess is uncommon and its diagnosis remains challenging. Clinicians must always remain open to consideration of this diagnosis. Early referral, judicious use of appropriate imaging and early drainage remains the key to optimise patients' outcome.

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Author Contributions

Victor Kong – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
George Oosthuizen – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Musa Mthethwa – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Kriban Reddy – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Damian Clarke – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

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CASE REPORT OPEN ACCESS

Psychosocial interventions in a case of fibrodysplasia ossificans progressiva: A case report

Manisha Jha, Ruchi Varma Shanker, Anand Kumar, Tej Bahadur Singh

ABSTRACT

Introduction: The developmental needs and challenges of children with chronic illness are different from those of a normal childhood. Moreover, rare disease with no effective treatment option and poor prognosis is devastating for parents also. Case Report: This case report aims to present guided sustained adaptation of a child from India who was diagnosed as a case of fibrodysplasia ossificans progressiva. The child and her parents were referred for psychological intervention in view of rare genetic disease to facilitate positive health. A detailed assessment and psychological intervention was done by trained clinical psychologist. Conclusion: The case report highlighted that caring relationship, formulation of positive meanings about the illness and an opportunity to seek help along with promoting the strengths of the child while she was undergoing a negative experience facilitated child's capacity for innovative survival and ability to resist adversities.

Keywords: Fibrodysplasia ossificans progressiva (FOP), Well being, Psychological intervention

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INTRODUCTION

Fibrodysplasia ossificans progressiva (FOP) is a rare and disabling genetic condition characterized by congenital malformations of the great toes and progressive heterotopic ossification in specific anatomic patterns. The condition was first reported in the 17th century by Patin, a French physician, who described a woman who "turned into wood". The prevalence of FOP has been estimated at 1/1.64 million persons. Fewer than 200 cases have been described worldwide. FOP is more common in females than in males. FOP is an autosomal dominant condition, but most cases are sporadic. Recently, genetic analysis revealed that the FOP gene is located on chromosome 4 and mutation in this gene causes an over expression of a bone morphogenetic protein (BMP4) in almost all sporadic and familial cases of classic FOP. Minor trauma can trigger proliferation of connective tissue (muscles, ligaments and tendons) resulting clinically as painful swelling of the muscles and connective tissue. This swelling subsides, then after approximately six months or more, heterotrophic ossification starts at some sites in which extra bone formation occurs outside the skeleton. The disease generally presents itself in childhood between 2–6 years and progressively disables the body as the new bones forms bridges and ribbon like structures across the joints and creates immobility. Since no effective medical treatment still exist, most of the patients become bedridden by the time they are in

fourth decade. This case report aims to present guided sustained adaptation of a child from India who was diagnosed as a case of fibrodysplasia ossificans progressiva. The child and her parents were referred for psychological intervention in view of rare genetic disease to facilitate positive health. Parents offered consent for scientific publication, expecting to receive informal support regarding the disease through this medium. They received psychological intervention for about two years along with continued occupational therapy and physiotherapy. The patient was quite compliant for the therapeutic sessions considering the preventive nature of the therapies.

CASE REPORT

Patient R was an eight-year-old child studying in 3rd standard. She was third in birth order among her four siblings and belonged to a family with middle socio-economic status. She presented with a history of spontaneous flare ups (painful and highly inflammatory soft tissue swellings over her body, specifically in right elbow and dorsal surface of medial border and inferior angle of scapula). Flare ups marked the generation of new bone, although not every flare up ends in completion of process. Progressive restrictions in movements of cervical spine, right knee and both elbows were present since two years of age. Joint contractures were present due to bony ankylosis. She had progressive limitation of movement following pain and tumour like growth at the multiple sites (Figure 1). At the time of presentation she had multiple contractures and used compensatory techniques to perform her daily activities. Radiological investigations (X-rays, bone scans) showed microdactyly of big toe (Figures 2 and 3), hallux valgus and ossifications at various sites in body (scapular region and both elbows, more in right elbow). Investigations revealed a normal EMG and normal MRI brain study, however, computed tomography (CT) scan of head showed possible basal ganglia calcification. There were no other neurological deficits. She was diagnosed as a case of fibrodysplasia ossificans progressiva and was advised non- steroidal anti-inflammatory drugs (NSAID) only. A detailed assessment by trained professional was done to assess the current level of cognitive and emotional functioning of the child and family functioning in terms of stress and coping pattern of parents in relation to a chronic illness. This included assessment and intervention by a clinical psychologist and occupational therapist respectively. Her assessment of current functioning revealed inability to use upper and lower limbs properly due to limited movement, inability to sit and she needed physical help to perform self care activities. The patient used log like pattern for movement as there was severe limitation of range of motion (ROM) in all the axial joints. While walking she had fair balance but had a tendency to fall more on the front. Her fine motor prehension was still relatively spared, thus she could use a pen, pencil and computers for academic purposes. She

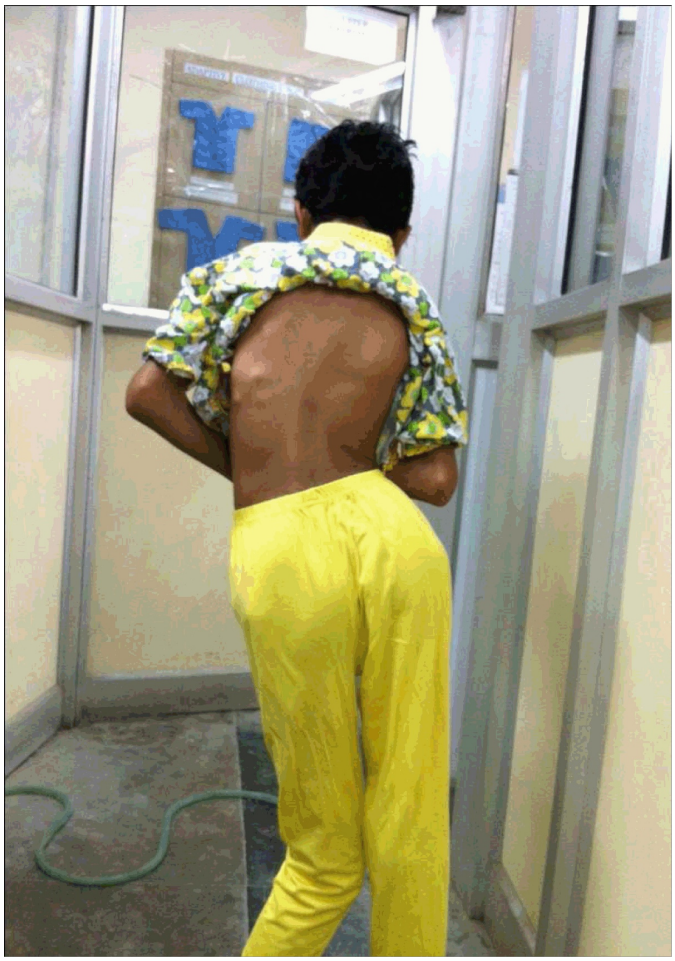

Figure 1: The patient showed spinal deformity in the form of scoliosis. Multiple small ossifications are seen along the inferior angle of left scapula.

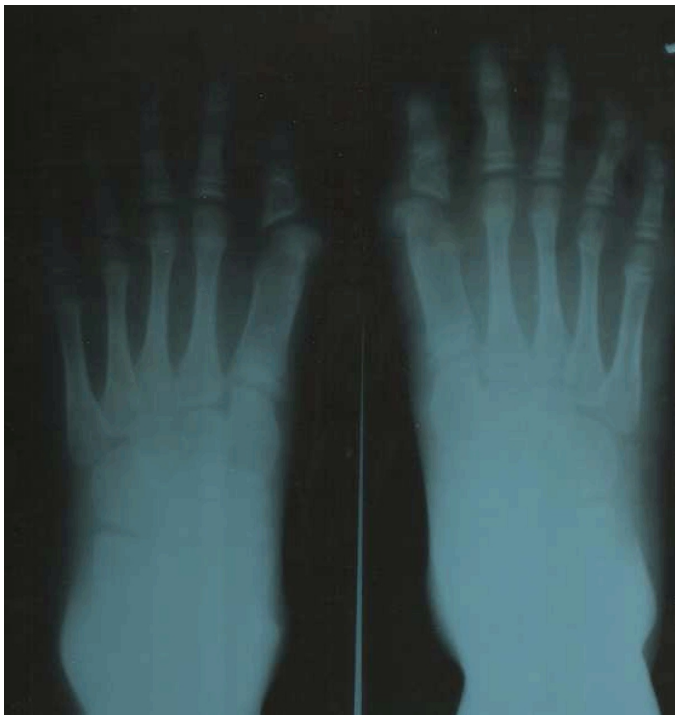

Figure 2: X-ray showing malformation (microdactyly) of big toe.

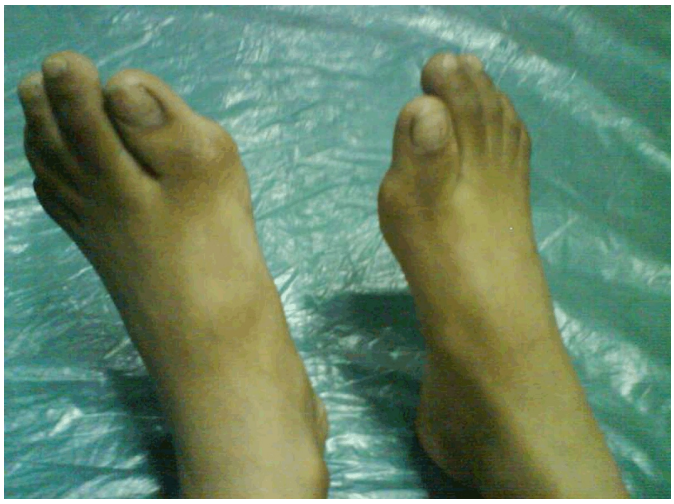

Figure 3: Characteristic feature of fibrodysplasia ossificans progresiva: malformation (microdactyly) of big toe.

had a fair handwriting and reported little difficulties in studies, at school. She attended school regularly. Some modification in her sitting arrangement had to be done at school considering her difficulty to sit on chair. The child did not feel herself to be different from other children in any way and could maintain her friendships.

The psychological tests administered included WISC IIIR [1], Luria Nebraska Neuropsychological Battery for Children [2], House Tree Person Test [3], Rosenzwig Self Esteem scale [4], Children Depression Index [5], Parental Stress Index [6] and Ways of Coping Questionnaire [7]. Since the intervention targeted a range of developmental outcomes, which more closely represent resilience, the General Well Being 20 test [8] was used which measures the impact on a variety of psycho-social constructs. One factor measures the level of occurrence of positive psychological well-being during the past month (or other time frame), and the other factor measures the level of occurrence of psychological distress during the same time frame. Findings of cognitive functions suggested average intelligence with an I.Q. of 101. On assessment, deficits in neuropsychological functioning were noticed in the tactile functions only. Qualitative scoring indicated deficits in the area of motor perseveration, right-left disorientation, stiff motor movements, motor awkwardness and unilateral neglect. Her self-esteem appeared to be moderately high irrespective of her illness status. Assessment on projective test revealed strong need for affiliation and dependence on the peer group. She was quite open to social interaction yet showed control and social tact for her inabilities, with intention to grab opportunity from environment. As far as family unit was concerned; she appeared more assertive, decisive and depicted controlled behaviour which was reflective of a normal secure person. Findings were not suggestive of depressive features. On measure of well-being and distress she scored 32 and 43 respectively, indicating low on well-being and high on distress dimensions. Findings suggested that it could be the parent's perceived or real inability to observe and understand the child's feelings or needs accurately, which somehow restricted them in terms of skill or knowledge regarding child rearing. They often found themselves distancing away from relationships and poor in seeking support. They manifested guilt and unhappy feelings emerging from difficulty to mobilize the psychic and physical energy needed to fulfil parenting responsibility in view of progressively deteriorating health condition. It appeared that their commitment to being ideal parents magnified the source of stress. On the child domain high on demandingness and adaptability may be due to child's physical constraints that made her unable to adjust to changes in her physical or social environment. Coping strategies seen in father was a tendency of wishful thinking to escape or avoid the problems and in accepting responsibility along with poor support seeking. Both the parents were making attempts for planned problem solving and making efforts to create an environment for personal growth.

In psychotherapeutic sessions, information was provided to the child and her parents together, to address the underlying apprehensions, anxieties and concerns considering the nature of illness and process of treatment. At the initial stage of diagnosis, availability of formal sources of social support adequately and on a consistent basis such as advantage of talking with experts, education about the illness and prognosis and information about resources available in meeting with the daily needs of families, improved their understanding and decision making related to disease. The central idea was to reduce parental anxiety which often gets communicated to the child by verbal or non-verbal channels. A few sessions were also devoted to reflect back and restore the belief in self with regard to parental responsibility. This stemmed from the fact that no significant observed change in illness status could be seen in spite of extensive efforts. After the initial impact of the diagnosis of chronic illness was settled, the child and her parents were encouraged to normalize the situation. Supportive therapy with this child was mainly focused on assimilation of her personal asset and basic needs by modifying ways of achieving the set goals. Promoting the strengths of the child while she was undergoing a negative experience was relevant for child's well-being (such as, to seek peer group affiliation, her computer proficiency and interpersonal skills were emphasised to compensate for physical limitation). Thus, improving her social competence, increased comfort with emotional states and creating opportunities together with normally developing peers. For parents problem solving strategies facilitated minimizing the disturbances in day to day activities and conveying the impression of normalcy to others thus, bolstering their coping skills and self-esteem. It seemed intervention was needed to focus on assisting the parents in mobilizing their pride in their child and improving support seeking.

It was focused that parents must maintain a sense of balance in meeting the needs of their ill and healthy children. The central concept was the sense of

coherence that allowed them to mobilize resources, promote effective coping, and resolve tension in a health promoting manner.

After intervention, score on well-being and distress was 58 and 18, respectively indicating valuable appreciation in her sense of well-being and reduction in distress. Extensive psychological sessions were terminated three years back and presently the child could negotiate well with the significant developmental challenges inspite of some progression in the disease. Family continued therapeutic contact as an when required.

DISCUSSION

Parents of children and adolescents with newly diagnosed chronic illnesses experience a range of emotions such as guilt, anxiety, shame, or anger [9]. It is more pertinent for rare diseases with lesser known prognosis which could influence child care practices. From the outset of a chronic illness a child also faces potential stressors in the form of threats to bodily integrity and aversive treatment regimens. A change also occurs in the child's interaction with peer group, disruptions in normal family affairs, social, and educational activities; all of which could be a significant threat to child's survival. Eventually, the past decade has seen a paradigm shift in the study of disease origins (pathogenesis) and psychopathology towards positive psychology [10]. It has been increasingly recognised that many children and parents successfully negotiate salient developmental tasks inspite of major stressors such as acute or chronic illness and possible underlying emotional distress [11]. As noted in recent literature on other childhood chronic illnesses and disabilities [12], some positive implications of illness seen in this patient was normalization and the attribution of meaning. A central theme of psychological intervention was empowerment, which referred to providing the chronically ill child and her caregivers with the maximum amount of control over their own lives.

CONCLUSION

We may state that in order to achieve and sustain resilient adaptation, the child must receive support from adults in the environment along with his/her own personal attributes. Caring relationships, promoting effective coping and opportunities for meaningful participation proved quite important in child's capacity for innovative survival and ability to resist adversities.

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Author Contributions

Manisha Jha – Substantial contributions to conception and design, Acquisition of data, analysis and interpretation of data, Drafting and revising article critically for important intellectual content; and Final approval of the version to be published

Ruchi Varma Shanker – Substantial contributions to conception, design and acquisition of data, Drafting the article for important intellectual content; and Final approval of the version to be published

Anand Kumar – Substantial contributions to conception and design, acquisition of data and interpretation of data, Drafting the article and revising it critically for important intellectual content; and Final approval of the version to be published

Tej Bahadur Singh – Substantial contributions to conception and design, Drafting the article for important intellectual content; and Final approval of the version to be published

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Conflict of Interest

Authors declare no conflict of interest.

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CASE REPORT OPEN ACCESS

Quetiapine associated dyslipidemia and hyperglycemia in a pediatric overweight patient

Elizabeth Carreño, Vivekananda Dasari, Aditya Badheka, Shanti Yogananda, Swati Dave-Sharma

ABSTRACT

Introduction: Atypical antipsychotics like quetiapine, can cause undesirable alterations in lipid profile and hyperglycemia. They are associated with metabolic and cardiovascular-related adverse events in pediatric population, especially when multiple antipsychotics or classes of psychotropic medications are co-prescribed. Case Report: A 15-year-old, overweight, Hispanic male child was referred to the endocrinology clinic by his primary care provider due to high serum cholesterol, triglyceride and glucose levels. Past history was significant for autism, anxiety with depression and attention deficit disorder for which he was getting psychiatric care and was receiving quetiapine 50 mg daily for more than two years. Physical examination was significant for increased adiposity and overweight. Fasting laboratory studies showed elevated cholesterol and triglycerides. In addition to lifestyle modification including diet and exercise, quetiapine was discontinued and replaced by aripiprazole. Repeat testing after four months showed normalized cholesterol and triglyceride levels. Conclusion: The use of atypical antipsychotics prescribed to children and adolescents with neuropsychiatric disorders has been associated with various adverse effects, including significant weight gain. These adverse effects are of particular concern in children and adolescents due to the immediate and long term health risks associated with weight gain, including obesity, diabetes mellitus and hyperlipidemia.

Keywords: Quetiapine, Dyslipidemia, Hyperglycemia, Overweight, Pediatric

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INTRODUCTION

Medications such as a typical antipsychotics, like quetiapine can cause undesirable alterations in lipid profile and hyperglycemia. Adverse metabolic effects such as diabetes mellitus, lipid abnormalities and weight gain have increasingly been recognized with the use of the newer, so-called atypical antipsychotic drugs. There are several possible means by which an agent may induce hyperlipidemia. For most agents associated with lipid dysregulation these mechanisms are not clearly elucidated and that remains the case for antipsychotic-induced hyperlipidemia. Nonetheless, several biologically plausible hypotheses have been advanced focusing on weight gain, dietary changes and the development of glucose intolerance, to explain the high incidence of hyperlipidemia with certain antipsychotic medications [1–5]. One mechanism which is operative in some patients is the development of glucose intolerance, with a direct correlation found in a number

of cases between the development of new onset Type-2 diabetes mellitus and elevated serum triglyceride (TG) levels, often with reversal of these problems upon discontinuation of the offending agent [6–8]. There is, however, ample evidence that the mechanism underlying hypertriglyceridemia related to atypical antipsychotic therapy is generally not dependent on glucose intolerance in light of the finding that increased serum TG or hypertriglyceridemia is prevalent in studies involving dibenzodiazepine-derived atypical antipsychotics. Hyperlipidemia is not mentioned in the side effects by manufacturers of quetiapine. The following is a case report of possible quetiapine-induced severe hyperlipidemia.

CASE REPORT

A 15-year-old overweight, Hispanic, asymptomatic male child was referred to the endocrine clinic by his primary care provider due to high serum cholesterol, triglyceride and glucose levels. No personal or family history of hypercholesterolemia, early cardiovascular event or diabetes was present. Past history was significant for autism, anxiety with depression and attention deficit disorder for which he was getting psychiatric care and was talking quetiapine 50 mg daily for more than two years. Physical examination was significant for increased adiposity and BMI in the overweight range (85–95th percentile) with pubertal tanner stage 5. Levels at the time of presentation showed elevated fasting total cholesterol level of 388 mg/dL, triglyceride of 1420 mg/dL and HDL cholesterol of 46 mg/dL, (LDL could not be calculated due to abnormally high TG).

Repeat levels of lipid panel and glucose showed similar results with elevated TGs and cholesterol two days later. LFTs showed elevated AST 47 IU/dL and ALT 105 IU/dL. Serum amylase, lipase, thyroid stimulating hormone, fasting insulin and urine micro-albumin levels were normal. Electrocardiogram, echocardiogram and cardiac examination was normal. Quetiapine was discontinued immediately and replaced by aripiprazole by the psychiatrist. Lifestyle changes including dietary and exercise program was started. The laboratory tests repeated four months later showed normalizing serum cholesterol of 209 mg/dL, triglycerides 261 mg/dL, HDL 44 mg/dL, LDL 113 mg/dL, AST 46 IU/dL and ALT 68 IU/dL with normal amylase, lipase, thyroid stimulating hormone and fasting insulin levels.

DISCUSSION

Atypical antipsychotics are increasingly prescribed to children and adolescents with neuropsychiatric disorders. Although their profile of potent antagonism at specific serotonin and dopamine receptors offers certain advantages compared with typical antipsychotics, their use has been associated with various adverse effects, including significant weight gain. No large controlled trials have been published quantifying the prevalence of adverse effects on glucose-insulin homeostasis and lipid metabolism in patients receiving atypical antipsychotics [6]. Quetiapine binds to multiple receptors, including D1, D2, 5-HT2A, 5-HT1A, and H1 receptors. Less information is known about the properties of quetiapine in children and adolescents. Martin et al. published a 16 week open-label trial of quetiapine (mean dosage 225 mg/day range 10–350 mg/day) in six patients (mean weight gain 2.9 kg, range of wight gain 0.9–8.2 kg). Mean weight gain was not statistically significant in the six patients as a whole [9]. Shaw et al. investigated the effectiveness and safety of quetiapine (mean dosage 47 mg/day; range 300–800 mg/day) in 15 children (mean age 15 years; range 13-17 years) with psychotic disorders. Mean weight gain over the 8 week open-label trial was 3.4 kg [6]. This adverse effect is of particular concern in children and adolescents, secondary to the immediate and long term health risks associated with weight gain, including obesity, diabetes mellitus, and hyperlipidemia [11, 7, 8]. We recommend that when evaluating the overall benefit-risk ratio of antipsychotics in children and adolescents, the practitioner needs to give careful consideration to possible metabolic disruptions or cardiovascular toxic effects, especially in individuals with comorbid metabolic conditions and those receiving concomitant psychotropic medications.

CONCLUSION

In summary, in past decades, the use of second generation and atypical anti-psychotics is increasing in young populations. Antipsychotics are associated with metabolic and cardiovascular-related adverse events in pediatric populations, especially when multiple antipsychotics or classes of psychotropic medications are co-prescribed. The cardio-metabolic adverse effects and tolerability is well known in adult populations but in pediatric population, cardio-metabolic adverse effects can be pronounced specially with long term use, obesity, use of multiple psychotropic medications, family history of coronary artery disease and diabetes. Pediatricians should closely follow such patients for their high risk of developing dyslipidemia and hyperglycemia. Routine laboratory studies including fasting lipid profile and glucose should be performed frequently.

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Elizabeth Carreño – Substantial contributions to conception and design, Acquisition of data, Drafting the article, revising it critically for important intellectual content, Final approval of the version to be published

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Vivekananda Dasari – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Shanti Yogananda – Substantial contributions to conception and design, Acquisition of data, Drafting the article, revising it critically for important intellectual content, Final approval of the version to be published
Swati Dave-Sharma – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

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The corresponding author is the guarantor of submission.

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Authors declare no conflict of interest.

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CASE REPORT OPEN ACCESS

Sliding hernia with appendix, cecum, ascending colon forming sliding component and transverse colon and greater omentum forming contents of the sac: A case report

Manash Ranjan Sahoo, T Anil Kumar

ABSTRACT

Introduction: Inguinal hernia is quite common in surgical practice. However, sliding hernia is rare with an incidence of 2–5%. The exact diagnosis of sliding hernia is made on the operating table. We report the case of sliding hernia containing transverse colon and its greater omentum as contents and appendix, caecum, ascending colon forming the sliding component. Case Report: A 70-year-old man presented with huge right sided groin hernia. Under general anesthesia reduction of the hernia was tried but it could not be reduced fully. Hence, very carefully the sac was opened and was found to contain transverse colon and greater omentum with appendix, caecum, ascending colon forming it's posterior wall. The contents were reduced and the sac was closed carefully. Posterior wall was reinforced with prolene mesh. Patient was discharged after seven days. Conclusion: Sliding hernia is a rare entity and those containing transverse colon, ascending colon, caecum and appendix are even rarer. Care must be taken to identify the contents of the hernia to avoid inadvertent injury to the structures.

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Keywords: Sliding hernia, Transverse colon, Ascending colon, Cecum

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INTRODUCTION

Sliding hernia is rare with an incidence of 2–5%. A sliding hernia is a protrusion of a retroperitoneal organ(s) through an abdominal wall opening, with or without its mesentery and with or without an adjacent peritoneal sac. These retroperitoneal organs may be the cecum, ascending colon or appendix on the right side and the sigmoid colon on the left side, or the uterus, fallopian tubes, ovaries, ureters and bladder on either side. The presence of these organs has been reported exceptionally in the literature and very infrequently it might even contain ascending colon and stomach. We report the case of sliding hernia containing transverse colon and its greater omentum as the contents and appendix, cecum and ascending colon forming the sliding component.

CASE REPORT

A 70-year-old male presented with huge right sided groin hernia. Clinically it was diagnosed as indirect inguinal hernia as it was extending till the level of the testes. However, deep ring occlusion test could not be

carried out as hernia was difficult to reduce. Routine blood investigations were within normal limits. Ultrasound of abdomen and pelvis was normal. Chest X-ray had changes of chronic obstructive pulmonary disease but the patient did not have any respiratory symptoms like cough, wheeze or breathlessness. The patient was posted for surgery. Under general anesthesia through an incision above and parallel to inguinal ligament extending to root of scrotum, cremasteric box was reached. It was incised longitudinally for better access to spermatic cord and internal ring. The cord was then separated from the sac and dissection was kept close to spermatic cord. The sac was palpated between two fingers and was found to be thick in most of the areas except anteriorly and at its tip. It was now tried to reduce the contents of the sac by holding the tip of the sac with artery forceps at its thinnest point but it could not be reduce fully, hence, the sac was opened carefully at its thinnest point and found to contain transverse colon and greater omentum within the sac. At the thick area posteriorly, to our surprise cecum, appendix and ascending colon was found forming part of the sac, identified by taenia coli and haustrations (Figure 1). The contents within the sac were reduced (Figure 2). Only part of the sac which was thin was removed leaving the posterior part formed by cecum, appendix and ascending colon and thin part of sac (Figure 3). Adhesions at the internal ring were released carefully. Sac was closed carefully, starting a little away from the posterior wall where a small part of the thin sac was left behind (Figure 4). Then whole of the hernial sac including the posterior wall after closure, was reduced into the peritoneal cavity. Big patulous internal ring was closed with 1-0 prolene after orchidectomy. Posterior wall was reinforced with prolene mesh. Post-operative period was uneventful except that the patient developed cough on second post-operative day. He was given antitussives and bronchodilators for four days after which the symptoms subsided. Patient was discharged after seven days.

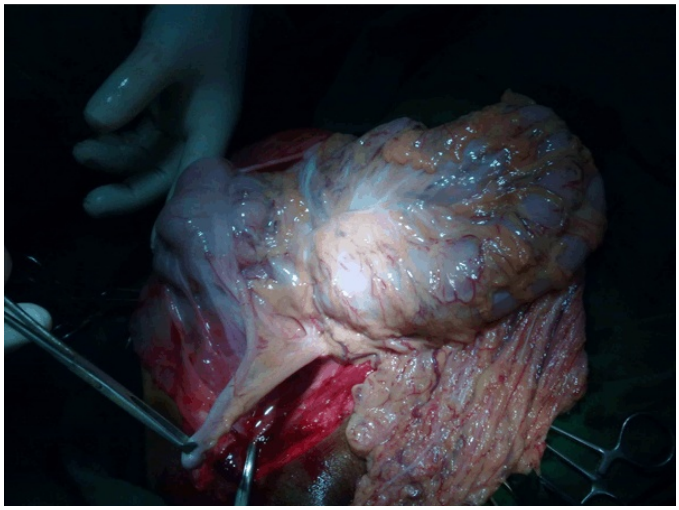

Figure 1: Hernial sac containing transverse colon with greater omentum; with appendix, cecum, ascending colon forming it's posterior wall.

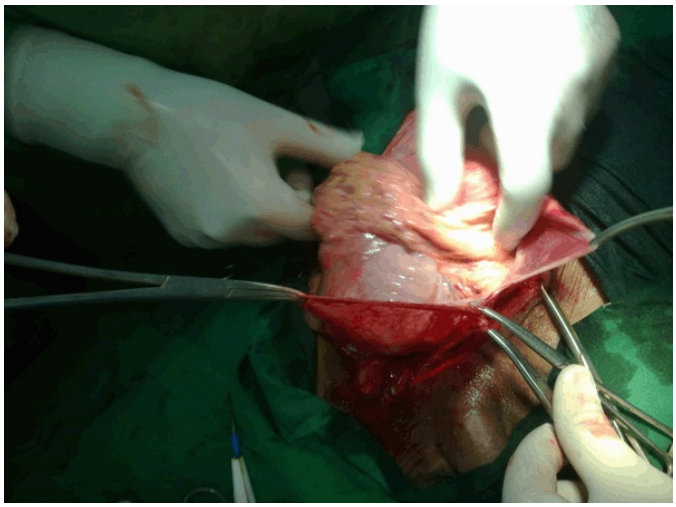

Figure 2: Contents were reduced after opening the hernia sac.

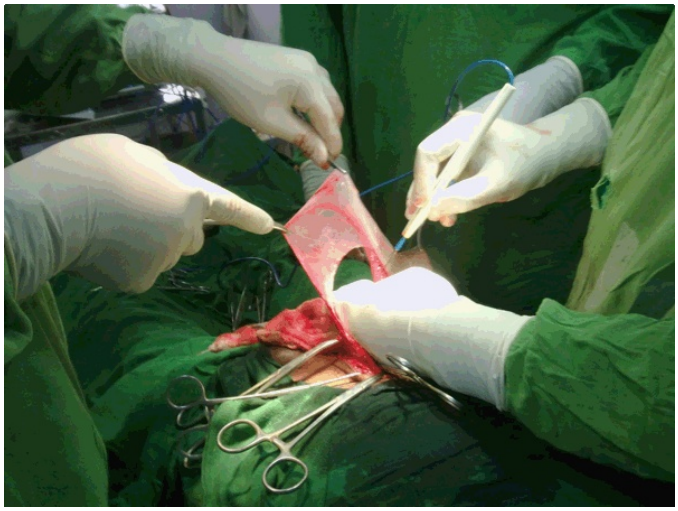

Figure 3: Removal of part of the hernial sac.

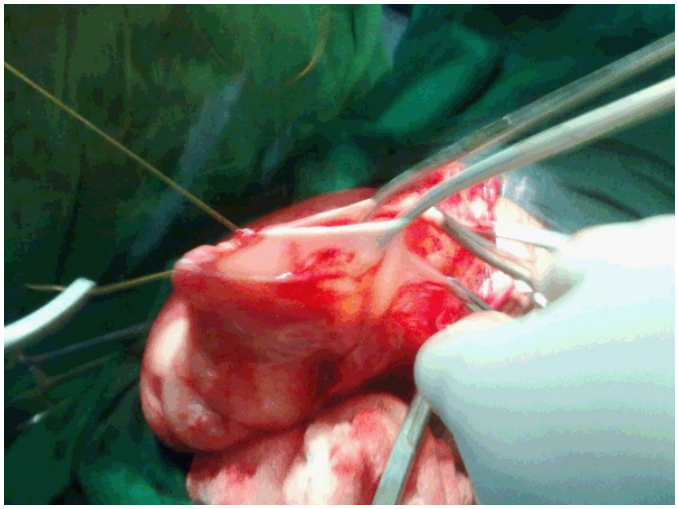

Figure 4: Closure of the hernial sac.

DISCUSSION

Galen (130–200 AD) gave us the first description of a sliding hernia involving the cecum. If Condon's [1] dictum "the anatomy of the inguinal region is misunderstood by some surgeons at all levels of seniority" is correct, it is safe to say that sliding hernias are understood by few surgeons at any level of seniority. As observed by Ryan et al. [2, 3], Glassow et al. [4] and Welsh et al. [5], sliding inguinal hernias accounts for 8% groin hernias, with a left to right ratio of 4.5:1. Maingot et al. [6], however, found a 1.5:1 preponderance of right sided sliders. In the series of Ryan et al. [2, 3] 8% were bilateral and women made up only 1% of the 3,000 patients analyzed. After the age of 50 years the incidence of sliding hernias is 3.5 times more frequent. The incidence of sliding inguinal hernia increases with the age of the patient. It is nearly zero before the age of 30 years and increases to as much as 20% after the age of 70 years. Our patient was seventy years old, one of the factors that might have contributed to sliding hernia. In the pediatric population boys are not subject to sliding hernias, whereas in "female pediatric patients, inguinal hernias are usually sliding hernias" with the mesosalpinx adherent to one side of the sac (type II) [7]. The exact diagnosis of sliding hernia is made on the operating table. In our case too, diagnosis of sliding hernia was made intra-operatively. Complications like incarceration, strangulation and obstruction of the hernia can occur. The presence of vermiform appendix, acute appendicitis, ovary, fallopian tube and urinary bladder has been reported in sliding hernia, exceptionally, in literature. Very infrequently it might even contain transverse colon and stomach. The mechanism whereby the viscus or viscera "slide" has not been fully explained. Before the slide can take place, however, there must be a widening of the internal inguinal ring; this is the precondition of an indirect inguinal hernia. In our case the internal inguinal ring was wide and patulous.

Ryan et al. after studying a series of 313 cases reported that in 47% patients no sac was removed and in 43% only a part of the sac was removed. In the remaining 10% cases, the sliding hernia was small and most of the sac was removed [2]. In our case also since hernial sac was large, only a part of hernial sac was removed. Ryan et al. also emphasized that the important step in the operation is to reconstruct the posterior inguinal wall in order to confine the sliding elements of the hernia to the preperitoneal space. This was achieved with a recurrence of less than 1%, at a time when one report from Philadelphia admitted total recurrence rate of 55% [2]. We reinforced the posterior wall by suturing it with prolene mesh. High ligation of the sac should not be attempted, as it is not necessary.

CONCLUSION

Hernia per se is not a difficult surgical condition but assumes importance when there is delay in seeking medical care, ultimately leading to increased morbidity. Sliding hernia is rare and those containing transverse colon as content of hernia sac and ascending colon along with cecum and appendix as sliding component is very rare. A large hernia of long duration in an elderly patient should cause suspicion of a sliding hernia. Care must be taken to identify the contents of the hernia to avoid inadvertent injury to the structures.

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Author Contributions

Manash Ranjan Sahoo – Conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published

T Anil Kumar – Conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Critical revision of the article, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CASE REPORT OPEN ACCESS

Adenocarcinoma at previous gastrojejunostomy site

Saroj Kant Sahoo, Manash Ranjan Sahoo, Ved Bhaskar

ABSTRACT

Introduction: There has been a growing interest in the development of gastric cancer following gastric surgery for benign disease. However, the risk is only observed after a latency of about 15 years and is increased in patients operated for gastric but not duodenal ulcers. The incidence of malignancy ranges from 2–6% in gastric remnants and a variety of causative factors have been proposed such as alkaline duodenal gastric reflux and increased N-nitroso compounds secondary to bacterial overgrowth. Case Report: We report an unusual case of a 75-year-old female who presented with recurrent vomiting. She had a gastrojejunostomy without gastric resection 25 years back. On investigation a gastric cancer was found at the anastomotic site in the efferent loop. Conclusion: Patients who have undergone surgery for ulcers should be screened using endoscopy at least once a year to detect the cancer in early stages as symptoms of cancer appear quite late and mortality is very high in advanced cases of stomach carcinoma. A complete margin negative (Ro) resection offers chances of cure in these patients.

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Keywords: Gastrojejunostomy, Peptic ulcer, Gastric adenocarcinoma, Stump carcinoma

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INTRODUCTION

There has been a growing interest in the development of gastric cancer following gastric surgery for benign disease. However, the risk is only observed after a latency of about 15 years and is increased in patients operated for gastric ulcers but not duodenal ulcers. The incidence of malignancy ranges from 2–6% in gastric remnants and a variety of causative factors have been proposed including alkaline duodenal gastric reflux and increased N-nitroso compounds secondary to bacterial overgrowth [1].

We report an unusual case of gastric cancer occurring at the gastrojejunostomy anastomotic site, 25 years after gastrojejunostomy without gastric resection, in a 75-year-old female presenting with gastric outlet obstruction.

CASE REPORT

A 75-year-old female presented to us with complaints of recurrent vomiting, anorexia and loss of weight since two months. Vomiting was projectile in nature, non bilious, contained food particles, and usually occurred immediately after taking food. Gradually, patient developed aversion to food and was on liquid diet since last one month. She gave a history of

gastrojejunostomy without gastrectomy for peptic ulcer disease which way done 25 years back. At presentation the patient was conscious and cooperative. Vitals signs were stable. She was thinly built with mild pallor present. On physical examination, adomen was scaphoid and umbilicus was centrally placed and inverted. A midline scar was seen extending from 5 cm below the xiphisternum to the umbilicus. Visible gastric peristalsis were seen. A firm, irregular, non-tender mass about 2×1 cms was felt in midline above the umbilicus which moved with respiration. Liver was not enlarged. Spleen was not palpable. No other organomegaly or free fluid was present. Per rectal examination revealed no abnormality. Routine laboratory investigations showed hemoglobin 8 g/dL, serum protein 5.0 g/dL and serum albumin 2.8 g/dL. All other investigations were within normal limits. Ultrasonography of abdomen did not reveal any significant findings. Computed tomography (CT) scan of abdomen revealed irregular posterior gastric wall thickening with in-situ infiltration involving the gastrojejunostomy site. No evidence of any secondary spread to solid viscera or any enlarged node was seen. Upper gastrointestinal endoscopy showed gastrointestinal stoma with stomal ulcerated growth and pyloric stenosis. Endoscopic biopsy was taken which on histopathological examination, showed invasive adenocarcinoma of tubular type grade II, (moderately differentiated).

The patient was taken up for surgery after correction of anemia and hypoproteinemia by blood transfusion, fresh frozen plasma and albumin infusion. A distal subtotal gastrectomy and D1 lymph node dissection and resection of previously anastomosed jejunal stoma (Figure 1) with Roux-en-Y anastomosis was done. The growth was found after opening the specimen at the gastrojejunostomy stoma (Figure 2). Postoperative recovery was uneventful. She was started on liquid diet on postoperative day 4 and semisolid diet on day 7. The patient was discharged on 12 postoperative day.

Histopathology report gave a diagnosis of gastric carcinoma stage II (T2b N1 Mx).

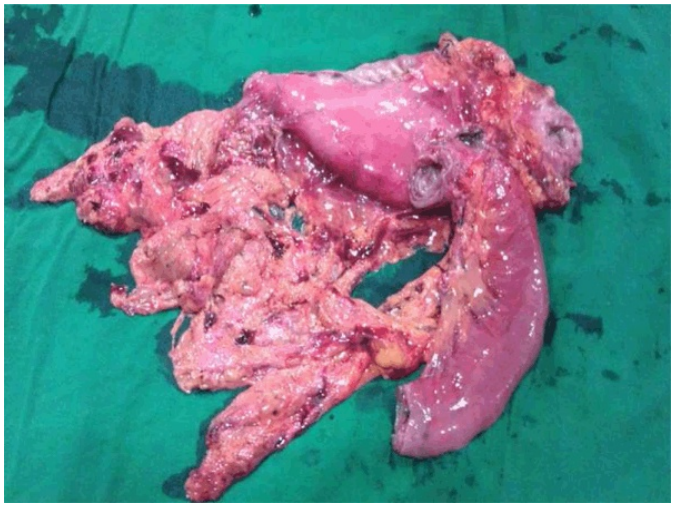

Figure 1: The subtotal gastrectomy specimen with D, lymph node dissection and resection of anastomosed jejunal stoma.

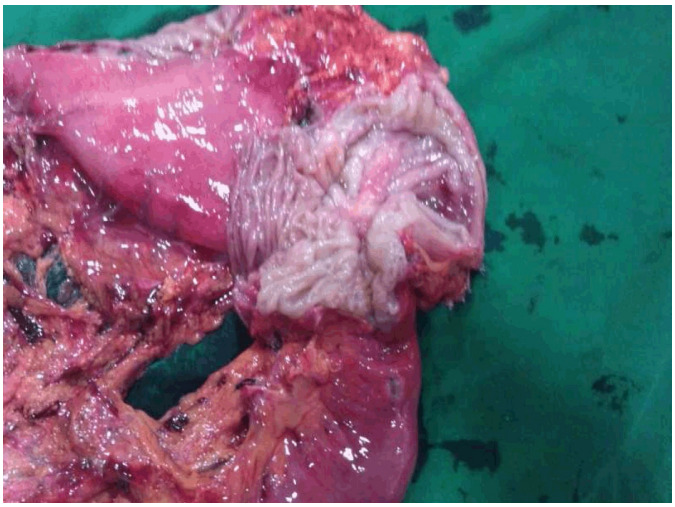

Figure 2: Gross appearance of malignancy seen at the gastrojejunostomy stoma.

DISCUSSION

It has been observed that patients who undergo gastrojejunostomy for peptic ulcer disease are at increased risk for gastric carcinoma [2]. Gastric cracinoma usually arise in an area of chronic gastritis, metaplasia and dysplasia which often occurs near the stoma. Most cases of stump carcinoma have been reported following Billroth II gastroenterostomy, but cases have also been reported following Billroth I gastroduodenostomy [1].

Gastric stump carcinoma is defined as an adenocarcinoma of the stomach occurring 10 years or more after gastrectomy for cancer or benign disease. It has been observed that patients with gastrectomy for cancer developed stump carcinoma in a significantly shorter time span than those with gastrectomy for benign diseases. The reason for these differences is that patients with gastric carcinoma already have carcinoma-related gastric mucosal changes at the time of primary surgery. Thus, gastric stump carcinoma after benign disease develops as a new lesion, while that after malignant disease may be a metachronous multiple lesion [1, 3].

Patients with Billroth II gastrectomy also develop stump carcinoma most frequently in the anastomotic area, regardless of the character of the original disease, whereas carcinoma for those with Billroth I gastrectomy is more frequently located in the non-stump area. The increased risk with the Billroth II procedure has been attributed to continuous bathing of the gastric stump anastomosis with bile acids, resulting in mucosal inflammation and regeneration [1]. The location of stump carcinoma close to the Billroth II stoma suggests involvement of duodenogastric reflux in gastric carcinogenesis [4].

There are differences in the tumor histology of gastric stump carcinomas between the early stage in which carcinomas are mostly intestinal type and the

locally advanced stage in which the carcinomas are mostly diffuse type. This suggest that stump carcinoma may develop as intestinal type and later change to diffuse type during the evolution to advanced stage cancers [1, 4].

In 1972, Littler et al. reported that polypoid tumor developed in human gastric mucosa adjacent to Billroth II stoma. Based on the characteristic changes in hyperplastic foveolar epithelium and the multiple cystically dilated glands, they termed these tumors “gastritis cystica polyposa" (GCP) [3]. The etiology and pathogenesis of GCP are considered to be chronic inflammation; a consequence of the suture technique itself; or a consequence of duodenogastric reflux into the gastric remnant. In 1975, for the first time, Qizilbash reported the association of GCP with stump carcinoma [5]. In Japan, several reports have documented GCP associated with early gastric stump carcinoma after Billroth II procedure [6, 7]. Characteristically, the foci of adenocarcinoma are found in the superficial layers of GCP. The degree of intestinal metaplasia in the surrounding mucosa was low. These findings suggest a relationship between GCP and gastric type adenocarcinoma [3].

As far as role of H. Pylori and Estein Barr Virus in the pathogenesis of stomach carcinoma is concerned, two important differences were observed between gastric stump carcinomas and conventional primary stomach cancers. H. pylori infection, which is associated with the occurrence of cancer in the intact stomach, appears rare near the anastomosis of the gastric remnant, the site prone to carcinogenesis [8, 9]. Bile reflux may provide an explanation for the observed low prevalence of H. pylori in the stomach epithelium bordering stump carcinomas. Bile salts seem to have a bactericidal effect on H. pylori and after a Billroth resection, H. pylori disappears rapidly from the gastroenterostoma.

In contrast, EBV infection appears more common in gastric stump cancer, not only in Asian countries, but in the western world as well. One possible explanation may be bile reflux into the stomach which could act as a cofactor mediating EBV infection of the epithelial cells, for instance by inducing fusion of EBV carrying B cells and epithelial cells. The EBV positive carcinomas were predominantly of the early type, which is consistent with previous observations that EBV infection perhaps occurs early in carcinogenesis [10].

A complete margin-negative (R0) resection remains the only potentially curative treatment for gastric adenocarcinoma [11, 12]. Gastric stump carcinomas often have low resectability rates and a poor prognosis. Gastric stump carcinoma has a wide range of lymph node (LN) metastases, including LN within jejunal mesentery in Billroth II reconstruction cases. Therefore, it is suggested that radical operation for Billroth I patients needs removal of gastroduodenectomy anastomosis and the above LNs, and that Billroth II patients need removal of 10 cm of jejunum besides gastrojejunostomy anastomosis, and clearance of LN within its mesentery, in addition to Billroth I stump carcinoma [13, 14].

A recent study by Newman et al. supported the opinion that the outcome after resection in gastric stump carcinoma does not differ from that of patients with other primary proximal gastric cancers of the same stage. There are no differences in prognostic factors between primary gastric cancer and gastric stump cancer patients in terms of resectability rate, death rate, and survival. Therefore, the consequences of surgical therapy in these patients are identical [15].

CONCLUSION

Recommendations has been made to screen patients with gastro-jejunostomy by endoscopy at least annually to detect the cancer in early stages. Only a complete margin-negative resection offers chances of cure and mortality is very high in advanced cases of carcinoma stomach.

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Author Contributions

Saroj Kant Sahoo – Conception and design, Analysis and interpretation of data, Critical revision of the article, Final approval of the version to be published
Manash Ranjan Sahoo – Acquisition of data, Analysis and interpretation of data, Critical revision of the article, Final approval of the version to be published
Ved Bhaskar – Acquisition of data, Drafting the article, Critical revision of the article, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CASE REPORT OPEN ACCESS

Unconventional etiology for heart failure and stroke: Non-compaction cardiomyopathy

Srikanth Seethala, Srinivasa V Jampana, Gur C Adhar, Venkataraman Krishnaswami, Thomas Generalovich

ABSTRACT

Introduction: Non-compaction cardiomyopathy is a rare, primary, genetic cardiomyopathy that results from intra-uterine arrest of the compaction process. It commonly involves the apical area of the left ventricle, and uncommonly involves the right ventricle. Case Report: A 45-year-old male presented to the hospital for heart failure. He was started on anticoagulation for the apical thrombus and later he suffered cerebrovascular accident. On careful evaluation he was found to have ventricular non-compaction both in right and left ventricles. Conclusion: Non-compaction cardiomyopathy should be considered in all refractory cases of heart failure. Anticoagulation is crucial in patients with decreased systolic function and atrial fibrillation.

Keywords: Biventricular non compaction, Ventricular non compaction, LVNC

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INTRODUCTION

Non-compaction cardiomyopathy is a rare, primary, genetic cardiomyopathy. Human cardiac morphogenesis commences with heart tube formation. Heart tube contains outer myoepicardial cells, middle acellular cardiac jelly (matrix of proteoglycans and glycosaminoglycans), and outer endocardial cells. During this time myocardial cells form deep trabeculae and intertrabecular recess so that blood percolates into intertrabecular recess in order to supply oxygen to myocardium. Compaction process, epicardial vessel formation and myocardial vascularization start around 5th–8th week of gestation. Compaction proceeds from epicardial to endocardial surface and from the base to apex. Arrest of the cardiac compaction around this time will lead to non-compaction cardiomyopathy [1]. It commonly involves left ventricle. Right ventricular involvement is rare and here we are reporting a case of biventricular involvement.

CASE REPORT

A 45-year-old male presented to our institution for progressively worsening bilateral lower extremity edema, productive cough and generalized weakness that had been present for three weeks. His medical history was significant for recurrent bronchitis, smoking and a remote history of cocaine abuse. His family history was unremarkable for any cardiac disease. On physical

examination, blood pressure was 142/90 mmHg, and heart rate was 109/min.

The patient had marked jugular vein distention, S3 gallop, bilateral lower extremity edema and normal pulmonary examination. The electrocardiogram revealed sinus tachycardia with voltage criteria consistant with left ventricular hypertrophy and repolarization abnormalities (Figure 1). Blood chemistry revealed a serum creatinine of 1.8 mg/dl, blood urea nitrogen of 24 mg/dl, and mild proteinuria. Chest X-ray revealed cardiomegaly and probable left upper lobe infiltrate. A 2Dechocardiogram was performed which revealed moderate left ventricular dilatation, severely reduced left ventricular systolic function (estimated ejection fraction of 10-20%) and a large mobile apical thrombus. He was treated with heparin, warfarin, lisnopril, carvedilol, diuretics and digoxin. After achieving therapeutic anticoagulation and a good control of symptoms of heart failure he was discharged with a diagnosis of non-ischemic cardiomyopathy with a referral to a warfarin clinic for anticoagulation monitoring.

He presented to the emergency department 10 days after discharge for worsening of bilateral lower extremity edema. On repeat laboratory work up, his serum creatinine had decreased to 1.19 mg/dL, PT was 14.2 and INR was 1.2. He was admitted to the hospital for worsening heart failure. Next morning, he was found to be unresponsive. A computed tomography (CT) scan of the head failed to reveal any hemorrhage and magnetic resonance imaging (MRI) of brain revealed a hyper acute embolic cerebrovascular accident (CVA) in the left middle and anterior cerebral artery territory. An interventional neurologist performed thrombectomy and vasodilator therapy. We were consulted for apical thrombus management. We reviewed previous 2Dechocardiogram and suspected non-compaction cardiomyopathy because of hypertrabeculations and a spongy appearance of the myocardium.

A transesophageal echocardiogram (TEE) was performed to confirm the diagnosis and for detailed evaluation of the apical thrombus. TEE revealed biventricular non-compaction with marked hypertrabeculations in left and right ventricles and a ratio of the non-compacted to compacted myocardium greater than 2 (Figure 2) [1]. A left ventricular mural thrombus could not be either confirmed or excluded due to large mobile trabeculations. Unfortunately, his post-procedural course was complicated by the development of cerebral edema and a midline shift. He underwent decompressive hemicraniotomy and later suffered multi-system organ failure and was withdrawn from life support at his family's request.

An autopsy confirmed hypertrabeculations of both right and left ventricular myocardium, and a mural thrombus (Figure 3).

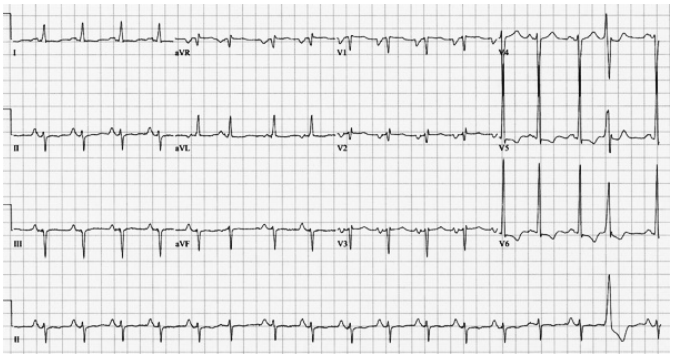

Figure 1: Sinus tachycardia with voltage criteria consistant with left ventricular hypertrophy and repolarization abnormalities.

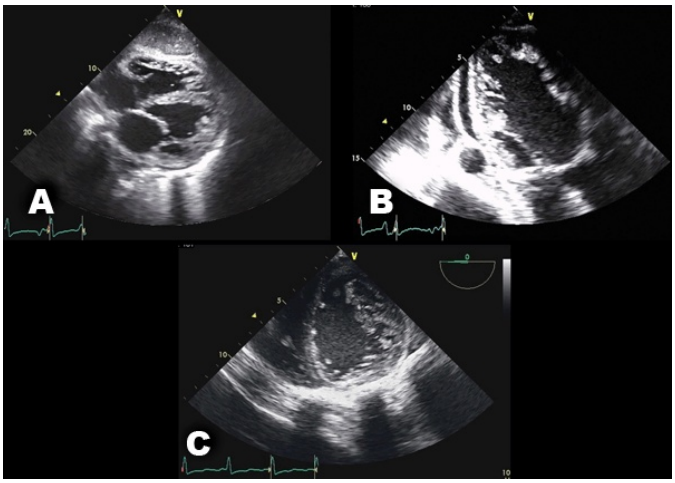

Figure 2: (A) Four chamber sub costal view showing hypertrabeculations, (B) Apical three chamber view showing hypertrabeculations, (C) Gastric short axis view of TEE showing hypertrabeculations.

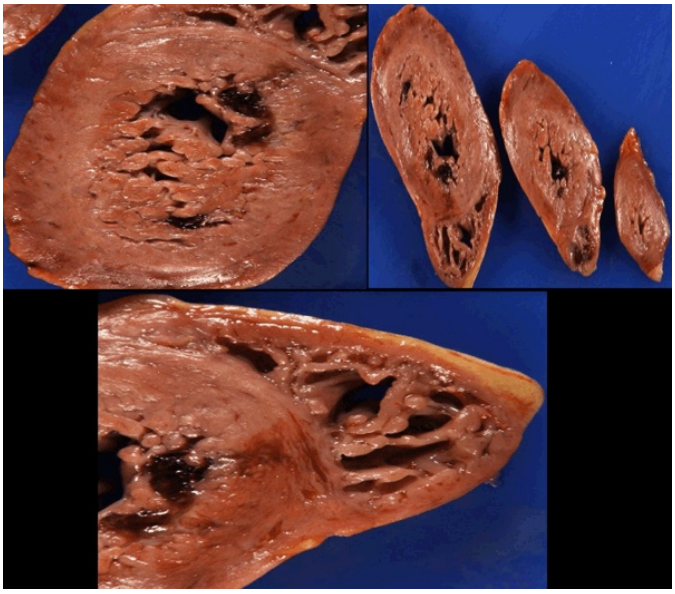

Figure 3: Autopsy findings of right and left ventricular hypertrabeculations, and a mural thrombus.

DISCUSSION

Ventricular non-compaction cardiomyopathy (VNC) is a rare, primary, genetic cardiomyopathy characterized

by hypertrabeculations and deep intertrabecular recesses. VNC is secondary to the intra-uterine arrest of the compaction process which occurs during the 5th–8th week of gestation. The prevalence of VNC is reported to be between 0.06–0.24% [2]. VNC commonly involves the apical, mid-lateral, inferior, anterior and septal areas of left ventricle, leading to the name left ventricular non compaction (LVNC). Right ventricular involvement is uncommon [1, 2].

The clinical presentation of LVNC is widely variable and ranges from incidental diagnosis to sudden cardiac death. The most common presentations of VNC are heart failure, arrhythmias and thromboembolic phenomenon respectively [1, 2].

Systolic dysfunction is the primary contributor of heart failure. Even though the exact etiology for systolic dysfunction is not clear, it is postulated that it could be secondary to subendocardial hypoperfusion, and micro-circulatory dysfunction. Abnormalities of diastolic function are common, ranging from abnormal ventricular relaxation to restrictive cardiomyopathy [3, 4].

Some studies have shown a correlation between the number of non-compacted segments and the severity of LV dysfunction [5, 6]. In contrast, one relatively large study failed to support the relation between number of non-compacted segments and the global systolic function [3]. It is difficult to compare these studies, as the imaging modalities, LVNC diagnostic criteria, and the average left ventricular ejection fraction (LVEF) differed in these studies.

The prevalence of atrial fibrillation in LVNC is reported as high as 26% [7]. Along with ventricular dysfunction, atrial fibrillation can worsen heart failure because of the loss of atrial kick and dyssynchrony between atrial and ventricular contraction. In addition to atrial fibrillation, left bundle branch block (LBBB) has been described in between 15–44% of the patients [1, 7]. Generally, patients with LBBB have lower ejection fractions when compared to their counterparts. LBBB can also be a contributor to lower ejection fraction and heart failure in (LVNC) patients.

In pediatric patients, a reduced early diastolic velocity at the lateral mitral annulus appears to be a more sensitive predictor for heart failure hospital admission when compared to the LVEF [8]. At this time, we are not sure whether or not we can extrapolate the study results to adult population as the disease spectrum is slightly different in pediatric patients when compared to adults. Even in adults, patients with a higher LVEF tend to be younger [1]. It is critical to identify LVNC and optimize medical management early in the course, since elevated left ventricular filling pressures and hospitalization for heart failure are independently associated with high mortality and the need for cardiac transplantation [1].

Thromboembolic events are common in LVNC. Deep intertrabecular recesses are presumed to be the nidus for thrombus formation [9]. It may be difficult to identify small mural thrombi in LVNC due to the presence of large, mobile trabeculations. In our case, while the initial transthoracic echocardiogram identified a left ventricular apical thrombus, we were not able to visualize a clear thrombus on trans-esophageal echocardiogram. The final autopsy revealed a small mural thrombus.

Left ventricular non-compaction by itself may not be an independent risk factor for thromboembolic events and the increased risk may be related to severe systolic dysfunction. All LVNC patients with systolic dysfunction should be anticoagulated [9, 10]. Patients with associated atrial fibrillation are at high risk for having peripheral embolic events and should be anticoagulated irrespective of their CHADS2 score index.

It is critical not only to initiate warfarin but also to maintain the INR in therapeutic range. Newer oral anti-coagulation agents may reduce the likelihood of sub-therapeutic anticoagulation, and have proven to be effective in patients with non-valvular atrial fibrillation. These agents could potentially be used in the management of atrial fibrillation in patients with LVNC, but their efficacy in the prevention of left ventricular mural thrombi is still unproven.

CONCLUSION

Ventricular non-compaction cardiomyopathy should be considered in the differential diagnosis of refractory heart failure.

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Author Contributions

Srikanth Seethala – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Srinivasa V Jampana – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

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The corresponding author is the guarantor of submission.

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Authors declare no conflict of interest.

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CASE REPORT OPEN ACCESS

Fournier's gangrene in a pediatric patient after prolonged neglected diarrhea: A case report

Sujoy Neogi, Piare Lal Kariholu, Deepak Chatterjee, Brajesh Kumar Singh, Rajnish Kumar

ABSTRACT

Introduction: Fournier's gangrene in pediatric population is rare and because of low incidence the etiopathogenesis in children is not well known. Case Report: We report a rare case of a 10-month-old male infant with severe Fournier's gangrene occurring after an episode of prolonged diarrhea. The progression of the disease was very fast and in spite of undergoing a diversion transverse colostomy, the child could not survive. Conclusion: Fournier's gangrene in children is rare and the course of the illness in our case was very rapid. The relation of this severe form of fasciitis with a common condition like diarrhea is rare and must be known to the treating clinicians.

Keywords: Fournier's gangrene, Perineal ulcer, Pediatric, Diarrhea complication, Necrotizing fasciitis

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INTRODUCTION

Fournier's gangrene is fascitis of the perineal region commonly seen in adult males with diabetes mellitus [1]. Pediatric involvement is rare. The use of nonsteroidal anti-inflamatory drugs (NSAID) have been found to be associated with Fournier's gangrene in children, besides other causes [1–6]. In the present case Fournier's gangrene was associated with neglected prolonged diarrhea, perianal excoriation resulting from diarrhea, and poor hygiene which has not been reported in literature so far. The use of NSAID may have also have contributed to the disease.

CASE REPORT

A 10-month-old male infant was brought by his mother to our emergency with a large ulcer in the perianal region for the last 10 days which was extending to the scrotum. The mother also complained that the child had decreased feed tolerance and decreased passage of stools. The child was ill for one month and his illness started with chest retractions, fever and diarrhea. He developed perianal excoriation following diarrhea, the intensity of which kept on increasing. The excoriation turned into a progressive ulcer including the perianal region and the scrotum. The progression of perianal disease was very fast and it reached the present

form in only 10 days (Figure 1). The child kept passing stools which contaminated the wound further. The child had received treatment in the form of antibiotic (amoxycillin) and non-steroidal anti-Inflammatory drugs (NSAID) (Ibuprofen), which were given when the child developed excoriation. On examining the child, the pulse was 150/min (low volume), respiratory rate was 35/min. General physical examination revealed left complete cleft lip with complete cleft palate. The child also had bilateral rhonchi in the chest. Local examination of the perineum revealed a large ulcer measuring 7x5 cm involving the anal canal and the perianal region. The anal canal was retracted up. There was another ulcer at the base of the scrotum measuring 3x4 cm which was superficial extending up to the dartos muscle. The floor of both the ulcers was dirty with extensive sloughing. The hematological report was as follows: hemoglobin 9.7 mg/dL, TLC $9x10^3$/ mm^3, platelet count $0.44x10^6/mm^3$, blood urea 20 mg/dL, serum creatinine 0.6 mg/dL, sodium 144 mmol/dL, potassium 2.9 mmol/dL. The pus swab culture reported the growth of klebsiella species. The child was resuscitated adequately with fluids, antibiotics (ceftriaxone, metronidazole and amikacin), platelet transfusion and ionotropes for 24 hours and then posted for emergency transverse colostomy. A colostomy was performed, the ulcer was debrided and the child was catheterized. The child was kept in the intensive care unit on ventilator owing to the poor respiratory efforts in the post-operative period. The condition of the child kept deteriorating and he expired 18 hours after surgery due to multiorgan failure.

DISCUSSION

Fournier's gangrene is a necrotizing fasciitis of the perineal region. It is progressive and life threatening if not aggressively treated. It is seen more frequently in adults than children and is associated with immunosuppressant factors [1]. Very few cases of Fournier's gangrene have been reported in children [1]. The infection in Fournier's gangrene is polymicrobial and the bacteria act synergistically to produce enzymes such as collagenase and hyaluronidase that invade the fascial planes leading to vascular thrombosis with subsequent gangrene of the overlying skin [2]. The necrotizing fasciitis commonly originates from an infection of the anorectum, urogenital tract or the skin of the genitals [3]. The predisposing factors linked to Fournier's gangrene in children, which are mentioned in literature are NSAIDs use, post varicella infection, diaper rash, prematurity, circumcision, strangulated hernia, testicular torsion, trauma and insect bite [1, 4–6]. The treatment of Fournier's gangrene in children is equivocal. On one hand some studies favor medical management or conservative surgery in children [7–9], others advocate prompt and aggressive surgical debridement [4]. In our case, the child had diarrhea with perianal excoriation which progressed rapidly to a severe debilitating ulcer of the perineum. There was also a history of NSAIDs intake which has been described as a predisposing factor for Fournier's gangrene in children. We speculate in this case that it was the prolonged and neglected diarrhea along with the poor hygiene of the child which contributed to the rapidly developing fasciitis. This case was worth reporting because Fournier's gangrene is uncommon in the pediatric population. Also, the occurrence of this condition following diarrhea is a matter of concern as the incidence of diarrhea in developing countries is very high and therefore development of perianal excoriation should be prevented and promptly treated.

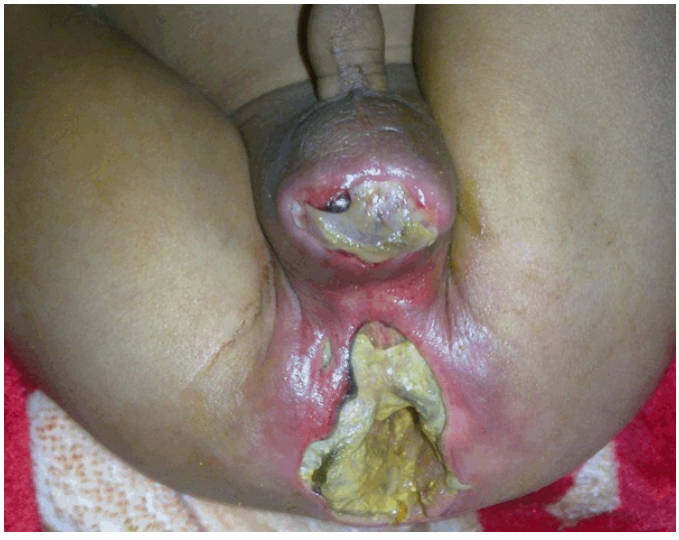

Figure 1: Clinical picture showing the perineal ulceration.

CONCLUSION

This is the first case report of severe, debilitating Founier's gangrene occurring in a child following prolonged diarrhea. Poor hygiene and improper treatment of the diarrhea have also contribute to the pathology. The role of NSAIDs cannot be confirmed, in causing this condition, although it has been given in our case for the treatment of perianal excoriation.

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Author Contributions

Sujoy Neogi – Substantial contributions to conception and design, Acquisition of data, Drafting the article, revising it critically for important intellectual content, Final approval of the version to be published

Piare Lal Kariholu – Substantial contributions to conception and design, Acquisition of data, Drafting the article, revising it critically for important intellectual content, Final approval of the version to be published

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Guarantor
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Conflict of Interest
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