Review article

The ‘forgotten rubber band’ syndrome – A systematic review of a uniquely ‘desi’ complication with a case illustration

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ABSTRACT

Background: Once an exceedingly rare entity, multiple cases of forgotten rubber band syndrome or the so-called ‘dhaaga’ syndrome have now been reported in the literature.**Objectives:** To conduct a thorough and systematic review of the literature for all articles reporting a chronic type of rubber band syndrome and to present an additional similar case as an illustration.**Study design:** Systematic review and case report.**Methodology:** PubMed, EMBASE and Google Scholar databases were searched for relevant articles using different combinations of the keywords till 20th June 2017. All articles reporting cases of chronic rubber band syndrome with a discharging sinus were included. Pearlring of the bibliographies of selected articles was conducted to locate articles missed by the primary database search. Data from these reports were collected on pre-defined forms and the results were analysed.**Results:** A total of 15 cases have been reported in the literature so far and all cases are from India. Thirteen of these have been reported in the wrist region. Median duration of presentation is 7.6 months after the application of elastic band. Characteristic clinical signs are a circumferential linear scar with discharging sinus (multiple sinuses noted in around one-fourth of the cases). ‘Soft tissue constriction sign’ on plain radiograph is pathognomonic for this condition. Rate of missed/misdiagnosis is very high (46.7%) and it has been confused with tubercular osteomyelitis which is endemic in India. All cases responded to surgical debridement of circumferential fibrous tissue and foreign body removal with good functional outcomes.**Conclusions:** A high index of suspicion must be maintained for this ‘syndrome’ in chronic osteomyelitis cases presenting with a linear, circumferential scar and discharging sinus in India. Soft tissue constriction sign on plain radiographs are pathognomonic.

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1. Introduction

An elastic band can have variable constrictive effects on the limb depending on its mechanical and tensile properties.¹ Two key types of presentations have been observed in the literature – acute and chronic presentations. The acute presentation is secondary to a tight, relatively less elastic band which results in compartment syndrome and gangrene and is usually picked up

early (also called as constriction band syndrome/tourniquet syndrome).^{1,2}

The chronic type of rubber band syndrome is due to a relatively more elastic band which burrows gradually into the tissue leading to distal edema, loss of function, infection and sometimes, even neurovascular damage.^{3–5} This presentation has been variously described as ‘dhaaga’ syndrome, forgotten rubber band syndrome or the elastic band syndrome; it’s often seen in young children in communities where elastic rubber bands are worn around the wrist for decorative or religious/ceremonial purposes.^{3,4} Although it has been described as rare, many articles have been published in the literature about this syndrome in the

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last two decades.^{3–13} Interestingly, all these cases have been reported from India.

We conducted a systematic review of the literature for all studies reporting cases of forgotten rubber band syndrome; epidemiologic, clinico-radiological and principles of management of this syndrome were analysed in depth. We also present a case of rubber band syndrome of the arm with chronic osteomyelitis of humerus treated at our institute as an additional case illustration.

2. Methods

The PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-analyses) checklist was used for key aspects of this systematic review wherever applicable.¹⁴ The PubMed, EMBASE and Google scholar databases were searched from 1960 to 20th June 2017 using different combinations of keywords (Table 1). PEARLing of the references of full text articles included in the review was conducted to identify articles which were missed in the primary database search.

2.1. Inclusion & exclusion criteria

We included all studies reporting rubber band syndrome of a chronic presentation with discharging sinus/sinuses. Cases where the presentation was in the form of an acute compartment syndrome were excluded. Only cases of the paediatric age group (less than 18 years) were included in the review. No language barrier was applied for the search.

2.2. Data collection & analysis

The search results were analysed by two observers independently (R.J. and A.K.) by using the study title and abstract. In cases of ambiguity, inclusion was ascertained by analysing the entire article after obtaining the full-text and consulting senior authors (NRG and PS). Duplicates were excluded; reference manager software (Zotero® Standalone software, Fairfax, VA, USA) was used to ensure elimination of duplicity. Full-text was obtained for all studies included in the final analysis. All data from the studies selected for the systematic review was collected on pre-defined, data collection forms. The following data was collected - journal name and year of publication, country of origin, demographic features of the patients, duration at presentation, clinico-radiologic features, details of management and complications

observed during the course of treatment. This data was analysed qualitatively and the results were summarised in a tabular fashion (Table 2).

3. Results & discussion

3.1. Literature search

Out of the initial 803 records identified by our search, 11 studies were identified which fitted the inclusion & exclusion criteria of the review. According to study design, two studies were case series whereas the other 9 studies were isolated case reports. Altogether 15 cases of rubber band syndrome with discharging sinus have been reported till date. A flowchart of all the studies included and excluded in the review has been given in Fig. 1. The results of these studies has been summarised in Table 1. Two cases were excluded as they reported a relatively acute type of presentation with absence of discharging sinus on presentation.^{6,15} One study reported this syndrome in an elderly demented person and was also excluded from the review.¹⁶

3.2. Demographic features

All 15 cases have been reported from India. All cases have been reported in children less than six years; mean age was 2.9 years (Range: 7 months to 6 years). Ten cases were females and five cases were males.

3.3. Clinical presentation

Mean duration at presentation was 7.6 months (Range: one month to 30 months) for the 14 cases which have specified details. Gill et al 7 have not specified the duration after application of the band at first clinical presentation. Vast majority of cases (86.67%) reported have affected the distal forearm/wrist region (13 out of the 15 reported cases).^{3–5,7–9,12,13,17} The other two cases pertain to the arm and thigh.^{10,11} The right side was affected in 10 cases whereas the left side was affected in five cases. Multiple sinuses were noted in 26.67% of the cases (four out of 15 cases).^{3,8,10,17}

A circumferential, linear scar was consistently observed in all cases. Rate of misdiagnosis/missed diagnosis was 46.7%; seven such cases were noted - three patients were put on anti-tuberculosis chemotherapy,^{3,4} one on multiple steroid injections,¹² and three other cases on antibiotics and/or analgesics.^{7,9,13}

Table 1
Search strategy used for this review.

Database searched	Results
1. PubMed (1960 to 20th June 2017)	
a. ("rubber"[MeSH Terms] OR "rubber"[All Fields]) AND ("Band"[Journal] OR "band"[All Fields])	777 studies
b. ("constriction, pathologic"[MeSH Terms] OR ("constriction"[All Fields] AND "pathologic"[All Fields]) OR "pathologic constriction"[All Fields] OR "constriction"[All Fields] OR "constriction"[MeSH Terms]) AND ("Band"[Journal] OR "band"[All Fields]) AND ("syndrome"[MeSH Terms] OR "syndrome"[All Fields])	270 studies
c. ("rubber"[MeSH Terms] OR "rubber"[All Fields]) AND ("Band"[Journal] OR "band"[All Fields]) AND ("syndrome"[MeSH Terms] OR "syndrome"[All Fields])	27 studies
d. elastic[All Fields] AND ("Band"[Journal] OR "band"[All Fields]) AND ("syndrome"[MeSH Terms] OR "syndrome"[All Fields])	25 studies
e. dhaga[All Fields] AND ("syndrome"[MeSH Terms] OR "syndrome"[All Fields])	2 studies
f. a & b	5 studies
2. EMBASE (1960 to 20th June 2017)	
a. 'constriction'/exp OR constriction AND band AND ('syndrome'/exp OR syndrome)	382 studies
b. ('rubber'/exp OR rubber) AND band AND ('syndrome'/exp OR syndrome)	39 studies
c. dhaga AND syndrome	4 studies
d. a & b	22 studies
3. Google Scholar (1960 to 20th June 2017)	24 studies

Table 2

A concise data review of all eleven studies included for this study.

Sl no.	Author Year	Country	Age/Sex	Location	Duration (in months)	Sinus	Circumferential scar	Constriction sign (X-ray)	Plane of the band	Recovered FB	Additional comments
1.	Kumar et al 11 1995	India	1 y/F	Thigh	7	Multiple	Yes	–	Anteriorly deep to quadriceps. Posteriorly superficial to hamstrings	Rubber band	Band was used as tourniquet for CTEV surgery
2.	Aggarwal et al 5 2010	India	18mo/M	Wrist	6	Single	Yes	Yes	Cutting through tendons and neurovascular structures	Rubber band	–
	a. Case 1	India	18mo/F	Wrist	4	Single	Yes	Yes	Superficial to flexor tendons.	Rubber band	–
	b. Case 2	India	2y/M	Wrist	6	Single	Yes	Yes	Deep to neurovascular structures.	Rubber band	Misdiagnosed as TB
	c. Case 3	India	2y/M	Wrist	6	Single	Yes	Yes	Deep to neurovascular structures.	Rubber band	ATT given
3.	Kumaraswamy et al 10 2013	India	5y/F	Arm	9	Multiple	Yes	No	Subcutaneous plane	Rubber band	Applied by osteopath
4.	Anil Agarwal et al 4 2013	India	3y/F	Wrist	3	Single	Yes	Yes	Cutting through tendons and neurovascular structures	Rubber band	MRI used to diagnose buried rubber band
5.	Arora et al 3 2014	India	18mo/F	Wrist	6	Single	Yes	Yes	Superficial to tendons	Ceremonial thread	Misdiagnosed as TB
	a. Case 1	India	4y/F	Wrist	3	Multiple	Yes	Yes	NV involvement present	Ceremonial thread	ATT given
	b. Case 2	India	4y/F	Wrist	3	Multiple	Yes	Yes	Superficial plane only	Ceremonial thread	–
	c. Case 3	India	4y/F	Wrist	30	Single	Yes	Yes	Buried below FCR tendon	Ceremonial thread	Misdiagnosed as TB
6.	Sreekanth et al 12 2014	India	4y/F	Wrist	16	Single	Yes	Yes	Cutting into flexor and extensor tendons.	Elastic hair band	ATT given
											C/S positive for streptococcus (post-op)
											Missed diagnosis initially-steroid injections given
7.	Gill et al 7 2015	India	6y/M	Wrist	NS	Single	Yes	–	Bone deep with positive obstruction sign	Rubber band	–
8.	Garg et al 13 2015	India	1y/F	Wrist	1	Single	Yes	No	Superficial plane only	Rubber band	–
9.	Goyal et al 9 2016	India	5y/F	Wrist	5	Single	Yes	No	Superficial to ulnar nerve	Rubber band	–
									FCU cut		
10.	Gupta et al 8 2016	India	3y/M	Wrist	12	Multiple	Yes	Yes	Superficial plane only	Rubber band	–
11.	Ojha et al 17 2016	India	1y/F	Wrist	6	Multiple	Yes	Yes	Cutting through tendons	Rubber band and ceremonial thread	–

Fig. 1. PRISMA flowchart showing the study design and the details of inclusion/exclusion of articles.

Fig. 2. Clinical pictures at presentation in pediatric orthopaedic out-patient clinic. Note the two sinuses with the volar sinus showing protuberant, sprouting granulation tissue.

Fig. 3. Plain radiographs in AP and lateral views showing bony indentation by the rubber band and soft tissue constriction sign.

3.4. Radiographic features

All cases were diagnosed clinically with supplementary plain radiographs except in two cases.^{5,8} Agarwal et al 5 used magnetic resonance images to confirm the presence of the buried rubber band in their report. Gupta et al 8 used ultrasonography (USG) to confirm the diagnosis.

The soft tissue constriction sign described first by Aggarwal et al 4 in 2010 was noted in 78.57% of the cases (10/13). The sign was absent in only three cases.^{9,10,13} In two studies, radiologic signs were not reported.^{7,11} The incidence of osteolytic or bony indentation signs was 23.1% (3/13 cases).

3.5. Management & complications

All cases were managed surgically (debridement, sinusectomy and foreign body removal). The foreign body retrieved was a rubber band in ten cases, ceremonial thread in three cases and an elastic hair band in one case. Ojha et al 17 reported double bands - a missed rubber band under a ceremonial thread, which was removed at the initial setting leaving the missed rubber band *in situ*, only to be removed later. Tendons were damaged in 53.3% of the cases (8/15 cases). Neurovascular structures were damaged in three cases.^{4,5} All cases healed uneventfully after surgery with the mean time to healing of sinus being 3.9 weeks (only 10 out of 15 cases reported data on healing time for sinus).

4. Discussion

The most striking demographic feature of this review is that all cases have been reported from India. This is because of the prevalent practice of tying ceremonial rubber bands on the wrist in many religious societies and communities in India.^{3,4,17}

All cases reported are in infants and children less than six years of age. The rubber band tends to stay hidden in the wrist crease; especially in infants and young children who tend to have chubby forearms and hands.^{3,4} There is gradual, painless penetration of the rubber band through the soft tissue due to the rapid growth in this age group; re-epithelisation of skin over the buried rubber band makes a retrospective diagnosis difficult.^{3,4,17} These patients often present late due to neglect by parents and also due to the young age of the affected children.

A circumferential linear scar associated with a discharging sinus is pathognomonic for this syndrome.³ Multiple sinuses or ulcers can also be seen at presentation.^{3,4,8,10,17} This syndrome can be easily missed if the clinician doesn't have a high index of suspicion and patients can be mistreated for tuberculosis (which is endemic in India).^{3,4}

Soft tissue constriction sign is a reliable radiological sign to diagnose this syndrome.^{3,4,17} Although MRI and ultrasound can be used to supplement radiographic findings, these are not a necessity.^{5,8} Surgical debridement and removal of the rubber band followed by antibiotic course was successful in all cases.

Fig. 4. Intra-operative picture showing the rubber band which was discovered during surgical exploration.

Complete recovery is the usual norm after debridement. Although, three cases also had neurovascular involvement, none of the cases required a formal neurovascular repair or graft reconstruction.^{4,5}

5. Case illustration

A 4-year-old male child presented with chronic osteomyelitis of humerus with active, discharging sinus and sprouting granulation tissue along with a circumferential scar on the arm (Fig. 2). Soft tissue constriction sign was seen on plain radiographs (Fig. 3). Operative debridement revealed a rubber band cutting through the muscle but sparing neurovascular structures (Fig. 4).

On probing further, parents gave history that child had applied rubber band on the arm while playing about 6 months ago. This rubber band had gradually cut through the skin, muscle and even bone and was the root cause of osteomyelitis. The child had excellent recovery post-operatively with full return of all functions by 12 weeks.

6. Conclusions

The presence of a linear circumferential scar with discharging sinus and osteomyelitis must always alert the physician/surgeon about the possibility of a deeply embedded elastic band which has been subsequently covered due to re-epithelisation of skin over the band. A high index of suspicion must be maintained to pick up this condition. As this syndrome has been exclusively reported from India, it is very easy to misdiagnose this condition as chronic tubercular osteomyelitis which is endemic in India. Look for the soft tissue constriction sign on the radiographs which is invariably present in these cases. Prompt surgical exploration and foreign body removal along with excision of circumferential fibrous tissue leads to a speedy recovery and good outcome in almost all cases. This review provides level IV evidence but the same is sufficient considering the rarity of the condition.

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