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Review Article

Internal jugular phlebectasia: A systematic review

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ABSTRACT

Background: Internal jugular phlebectasia (IJP), the abnormal dilatation of internal jugular vein, is generally considered a benign anomaly. However, because IJP is uncommon, little is known about its natural history, and currently, no consensus on the best treatment modality is available.

Methods: The purpose of this article is to conduct a systematic review of available literature on recently reported IJP cases to understand the main characteristics of IJP and its most frequent therapeutic approaches. Following the preferred reporting items for systematic reviews and meta-analyses guidelines, literature search for IJP cases was conducted in the COCHRANE, PUBMED, EBSCOHOST, SCOPUS, OVID, and SCIELO databases.

Results: A total of 97 original articles were found, with a total of 247 IJP cases reported including both pediatric and adult patients.

Conclusions: To the best of our knowledge, this study is the largest systematic review analyzing all the reported cases of IJP. IJP is considered by most authors as a benign abnormality that predominantly affects the right jugular vein. It is most commonly diagnosed in children. At present, conservative treatment is preferred for pediatric but not for adult patients. Multicenter randomized prospective studies are required to further understand this rare anomaly.

Keywords: Internal jugular vein, Neck mass, Phlebectasia, Venous dilatation

INTRODUCTION

Internal jugular phlebectasia (IJP), a nontortuous dilatation of the internal jugular vein (IJV), is typically considered a benign anatomical variant of unknown etiology.[37] Clinically, its most common presentation is as a soft and painless mass in the lateral neck, which transiently appears in periods of increased intrathoracic pressure, such as when performing a Valsalva maneuver.[69] At the present time, IJP treatment is not well defined. Both a conservative approach and surgical removal are described in literature. Surgery is mostly reserved for cosmetic reasons or to prevent complications such as enlargement or thrombosis of the dilatation.[82]

In literature, multiple terms have been applied to describe IJP such as venoma, venous cyst, venous aneurysm, and venous ectasia.[69] It was first described in a case report by Harris on a 5-month-old infant with a right-sided neck mass.[38] The infant died during the surgery to remove the mass, and the only finding considered to explain the symptoms was a blood-filled cyst. It was controversial whether the original lesion was, in fact, IJP. The first definitive case report of

IJP was published by Zukschwerdt, in 1929, and the anomaly was further characterized by Gerwig, in 1952.[30] Since then, there have been multiple case reports of IJP.[37,69,82] To the best of our knowledge, however, no systematic review defining the most common presentation of IJP and treatment of this anomaly can be found in literature. The aim of this study was to describe the most common clinical presentation, method of diagnosis, and treatment of IJP in adult and pediatric patients based on published data.

METHODS

Following the preferred reporting items for systematic reviews and meta-analyses (PRISMA) guidelines,[60] literature search for IJP was conducted in COCHRANE, PUBMED, EBSCOHOST, SCOPUS, OVID, and SCIELO databases up to October 11, 2018. As search terms, we used "phlebectasia" or its synonyms, "venous cyst," "aneurysmal varix," "venoma," "venectasia," "venous aneurysm," and "venous ectasia" in the title and/or abstract, with the word "jugular" included in all fields, excluding articles that had in the title the words "external" or "anterior." Only papers written in English or Spanish were considered for this review. The search was not limited by date of publication. This search strategy resulted in 211 unique articles. Titles and abstracts of the articles were screened. Potentially suitable studies for IJP were read in full by three independent reviewers. A total of 114 of the original articles were excluded. Defined variables (patient characteristics, clinical presentation, IJP characteristics, method of diagnosis, treatment, and outcome) were extracted independently by the reviewers and disagreements were solved by consensus. All the statistical analyses were performed using the statistical program MATLAB R2016a (Mathworks Inc.) the Chi-squared test was used for dichotomous variables and the Mann–Whitney *U*-test for quantitative variables, with a confidence interval of 95%. We considered $P < 0.05$ to be statistically significant.

RESULTS

Following PRISMA guidelines, a total of 97 articles were included in the analysis [Figure 1]. These articles covered a total of 247 patients with IJP, including both pediatric and adult patients [Supplemental Table 1]. In the analyzed papers, not all searched variables were described; therefore, the number of cases described for each variable could vary in the description and statistical analyses [Table 1].

We found that IJP predominantly affected the pediatric population, with 206 patients (83.4%) younger than 18 years and only 41 adult cases ($P < 0.001$) reported in literature. The mean age of presentation was 47.8 years in adults (range: 17–74 years) and 6.4 years in pediatric patients (range: 6 weeks–15 years). The sex most affected was female in the adult group (61%) and male in the pediatric group (71%).

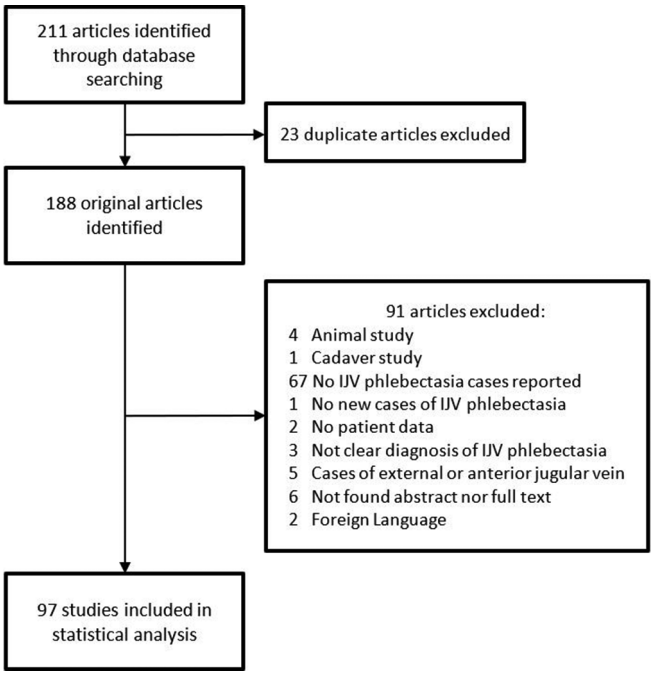

Figure 1: Preferred reporting items for systematic reviews and meta-analyses flow diagram.

The duration of symptoms before diagnosis was not significantly different between adult and pediatric patients. Adult IJP symptoms manifested predominantly as the presence of a permanent neck mass in 12 cases (29.4%), an intermittent neck mass in 11 (26.8%), an incidental neck mass in 9 (22%), voice changes and neck pressure in 2 cases each (9.8%), neck tenderness, neck pain, and hemoptysis, chest discomfort, and right shoulder weakness in one case each. Pediatric patients predominantly presented intermittent neck masses in 67 patients (57.8%), permanent neck masses in 42 cases (36.2%), voice changes in 2 (1.5%), and neck tenderness, acute venous hypertension symptoms dysphagia, Horner's syndrome, and otorrhea-otalgia in one case each. The Valsalva maneuver in both groups showed augmentation of the mass in 126 patients (92%) and no change in 11 patients (8%).

A history of head or neck trauma in both groups was reported in 97 patients. Only 4.1% of cases were related to direct trauma or surgery, and the proportion of this association in adults was higher than in pediatric patients but was not statistically significant.

IJP affected the right side in 176 patients (73.9%) and the left side in 44 patients (18.5%), and in 18 patients (7.6%), IJP showed bilateral involvement. Right-sided predominance was similar in adult and pediatric patients. IJP measurements were described in 84 of the 247 patients, of which 29 corresponded to the adult population and 55 were pediatric cases. The average dimensions are summarized in Table 2.

Table 1: Data analysis.

Number of cases	Adult (%)	Pediatric (%)
	41 (16)	**206 (83.4)**
Gender		
Female	25 (61)	38 (29)
Male	16 (39)	93 (71)
P value	0.05	<0.001
Clinical presentation		
Neck mass (intermittent/permanent)	23 (56.1)	109 (94)
Others	18 (43.9)	7 (6)
P value	0.46	<0.001
Increasing with Valsalva		
Yes	20 (69)	106 (98)
No	9 (31)	2 (2)
P value	0.004	<0.001
Symptoms duration		
<1 year	9 (47.4)	31 (49.2)
>1 year	11 (52.6)	32 (50.8)
P value	0.53	0.86
Trauma		
Neck trauma (direct or surgery)	3 (10.3)	1 (1.5)
No trauma	26 (89.7)	67 (98.5)
P value	0.9	0.98
Affected side		
Left side	13 (31.7)	31 (15.7)
Right side	28 (68.3)	148 (75.2)
Bilateral	0	18 (9.1)
P value	0.002	<0.001
Primary diagnosis study		
Ultrasound	20 (54.1)	88 (77.8)
CECT	13 (35.1)	9 (8)
Other	4 (10.8)	16 (14.2)
P value	0.02	<0.001
Secondary diagnosis study		
CECT	9 (45)	16 (53.3)
Others (US, MRI, and barium swallow)	11 (55)	14 (46.7)
P value	0.71	0.76
Thrombosed		
Yes	7 (17.1)	3 (1.5)
No	34 (82.9)	203 (98.5)
P value	0.85	0.99
Treatment		
Conservative	19 (54.3)	66 (64.7)
Surgical resection	16 (45.7)	32 (31.3)
Endoscopic resection	0	1 (1)
Surgical wrapping	0	2 (2)
Endovascular angioplasty	0	1 (1)
P value	0.47	<0.001

CECT: Contrast-enhanced computed tomography, MRI: Magnetic resonance imaging, US: Ultrasound

In just 150 of 247 cases, the primary diagnostic study was reported. Ultrasound (US) was employed for 108 patients (72%) as the primary study, a percentage that was higher in the pediatric population compared with that in adult patients. A contrast-enhanced computed tomography (CECT) was performed on 22 patients (14.6%), venography on 8 patients (5.3%), angiography on 4 patients (2.7%), magnetic resonance imaging (MRI) on 3 patients (2%), xenography on 2 patients (1.3%), and cinefluorographic studies, plain X-ray, and scintigraphy 99mTc-fibrinogen on 1 patient each (0.7%). The most common secondary study was CECT in 25 cases, MRI in 13 cases, US in 7 cases, barium swallow in 4, and unenhanced CT in 1 (2%). Secondary invasive studies were venography in 22 cases and angiography in 10 cases.

The treatment most frequently reported was a conservative approach in 85 patients (62%); however, the proportion of patients treated conservatively in the pediatric population was greater than that in the adult population. In the adult group for whom the treatment was described, no complications were found during conservative treatment, and complications from surgical resection were reported in 4 (11.4%) patients ($P < 0.001$): massive hemorrhage in 2 cases and paralysis of the left vocal cord and incomplete surgical resection in one case each. In the pediatric population, 102 patients had their treatment described. No complications were found during conservative treatment, and two (2%) patients had surgical complications ($P < 0.001$). The complications were subjective congestion of the left side of the head for 24 h after surgery and a postoperative transient increase in blood pressure. One patient treated conservatively died 14 months later due to Menkes disease complications.

DISCUSSION

IJP remains an infrequently diagnosed vascular anomaly. Although it is becoming increasingly recognized, partly due to improved diagnostic techniques,[44] medical guidelines have not yet been established. Clinical decisions for the diagnosis and treatment of this condition are based almost exclusively on personal experience. In this study, we analyzed clinical data gathered from literature on 247 patients with IJP.

IJP is classically found unilaterally, although a few bilateral cases have been reported. In this review, bilateral IJP was only found in a few pediatric cases. IJP is most commonly found incidentally during a physical examination as a mass in the neck that increases with common efforts such as talking, coughing, or swallowing. Other symptoms include voice alterations, paralysis of the vocal cords, and/or dysphagia, all of which are caused by the proximity of the vagus nerve and other lower cranial nerves to the IJP.[44,63,82]

Multiple pathologies affecting the neck are considered to be differential diagnoses, especially laryngocele, branchial cyst, cystic hygroma, hemangioma, and paraganglioma.[6,7]

Table 2: Average IJP dimensions.

Affected side	Adult			Pediatric		
	Average	Maximum	Minimum	Average	Maximum	Minimum
Right sided						
Diameter	3.95 cm	5.1 cm	1.1 cm	3.35 cm	12 cm	1.1 cm
Length	3.18 cm	7.0 cm	1.1 cm	3.43 cm	7.0 cm	1.0 cm
Left sided						
Diameter	4.73 cm	10 cm	3.0 cm	3.59 cm	5.5 cm	2.0 cm
Length	3.98 cm	6.9 cm	1.5 cm	3.96 cm	7.0 cm	2.8 cm

IJP: Internal jugular phlebectasia

In this review, we found some cases with initial inaccurate diagnoses that varied depending on the age group. In the adult population, four patients had other causes of the neck mass including globus pharyngeus, an infectious process, aneurysm of the subclavian artery, and an ovoid-shaped neuroma. In the pediatric patients, the erroneous diagnoses included goiter, laryngocele, adenopathy, adenoidectomy, and aerocele.

Although the etiology of IJP is not completely understood, some authors have suggested previous direct neck injuries or medical procedures such as central venous catheterization, positive-pressure ventilation, neck surgeries, or tumors.[11,56,63,88] In this review, no definitive association with these variables was found.

Histopathological changes in those cases treated with surgical resection include a thinning of the muscular and elastic layers; however, the great majority of cases showed no significant changes compared with the normal venous wall.[17,92] Several authors have found this abnormality in patients with neurofibromatosis type 1 and in Ehlers–Danlos syndrome.[18,39,49,67] In the present review, there was no significant association with these pathologies.

IJP most commonly presents as a unilateral right-sided anomaly.[91] In this review, the right-to-left ratio was 4:1. Although no clear etiology has been elucidated for IJP, multiple hypotheses have been formulated to explain the right-sided predominance.[71] LaMonte first proposed that, given the right innominate vein is in close contact with the right apical pleura, an increase in intrathoracic pressure would be transmitted to the right IJV and thus predispose an individual to unilateral phlebectasia. The left IJV, anatomically located more medially, does not receive such stress.[54] More recently, Paleri and Gopalakrishnan hypothesized that intrathoracic pressure could be easily transmitted to the right jugular bulb due to several anatomic factors including the following: (1) the fact that the right IJV valves, which are involved in preventing retrograde blood flow, are located more cephalad than their left-sided counterparts; (2) the larger diameter of the right IJV compared with the left side; (3) the direct continuity of the superior vena cava with the right brachiocephalic vein; (4) the higher number of valves in the left brachiocephalic vein in comparison with that of the right side; and (5) the higher number of competent valves in the right subclavian vein compared with that of the left side.[71] However, not all reported cases agree with the theory proposed by Paleri,[61] and more studies are needed to fully understand the side predominance of IJP.

The image study most frequently used for the diagnosis of IJP is US, with color Doppler as the study of choice to confirm the flow. This study continues to be useful because it is safe, feasible, sensitive, and of low cost. Other studies, such as MRI or CT, are usually helpful in complementing the diagnosis. Invasive studies such as angiography and venography are less commonly used, with only 10 (4%) and 22 (9%) cases, respectively, diagnosed by such methods.

Classically, IJP has been considered a benign entity; however, a serious possible complication resulting from abnormal vascular flow is thrombosis. In this review, the presentation of this complication was more frequent in adults, with 7 (17.1%) cases reported in literature, than in pediatric patients, with only 3 (1.5%) cases. Some authors describe Horner syndrome[46] as another complication. However, no significant association with that complication was found in this study.

Conservative treatment was most frequently reported for IJP in the pediatric population, given it is classically considered a benign vascular abnormality. In adults, surgical resection was selected in almost the same proportion as conservative treatment. Alternative treatments consisted of endovascular angioplasty, surgical wrapping, and endoscopic resection, but these were only performed on the pediatric population. In general, the clinical outcome in both groups was described as good, although the follow-up was not described in a standardized manner. Descriptions of surgical treatment revealed a complication rate of 6.9% in the pediatric population and 11.4% in the adult population. In the cases treated conservatively, no complications were reported in either group. In general, conservative treatment was a safer option than surgical treatment in both groups.

CONCLUSIONS

Following PRISMA guidelines, we analyzed a total of 247 patients with IJP, including pediatric and adult patients. Due to the low incidence of this abnormality, there are not enough original prospective studies to perform a meta-analysis. In the analyzed papers, not all the studied variables were described, and the heterogeneity of the reports prevented the homologation of the variables. However, we conducted a systematic review, in which we compiled all the available literature at the present time including all reported cases in the English and Spanish literature. IJP is considered by most authors to be a benign abnormality, is most frequently found in children, and it affects predominantly the right IJV. The clinical outcome in most cases was benign regardless of the treatment, which was either conservative or surgical. Conservative treatment is preferred for children but not for adults. To the best of our knowledge, this is the largest review of IJP to date. Future prospective multicenter studies that study diagnostic and treatment options are necessary to be able to develop guidelines on approaching this vascular abnormality.

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Conflicts of interest

There are no conflicts of interest.

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SUPPLEMENTAL

Supplemental Table 1: Articles included in data analysis.

Author	Year	Country of journal	Author origin	#of cases
Chua *et al*.[14]	2018	England	England	1
Nasiri *et al*.[64]	2018	USA	Saudi Arabia	1
Nedumaran *et al*.[65]	2018	India	India	1
Delvecchio *et al*.[18]	2017	USA	USA	1
Krstačić *et al*.[51]	2017	Italy	Croatia	1
Baker *et al*.[3]	2017	USA	USA	1
Bhattacharya *et al*.[5]	2017	India	India	1
Phookan *et al*.[74]	2017	USA	USA	1
Patel *et al*.[73]	2016	Romania	England	1
Raut *et al*.[77]	2016	USA	India	1
Sundaram *et al*.[91]	2016	USA	India	1
Soares-Medina *et al*.[85]	2016	USA	Spain	1
Yaadhavakrishnan and Navaneethan[98]	2015	India	India	2
Malik *et al*.[57]	2015	India	India	1
Khashram *et al*.[50]	2014	USA	Australia	1
Daley and Colliver[16]	2014	USA	USA	1
Tanigawa *et al*.[93]	2014	USA	Japan	1
Hiraki *et al*.[39]	2014	USA	Japan	1
Huang *et al*.[42]	2013	England	China	1
Eksioglu *et al*.[20]	2013	USA	Turkey	21
Nagata *et al*.[62]	2013	USA	Japan	1
Liu *et al*.[55]	2013	Germany	China	23
Czyżowski *et al*.[15]	2013	Czech Republic	Poland	1
Rha *et al*.[79]	2013	Korea	Korea	1
Bora[7]	2013	India	India	1
Chakraborty *et al*.[11]	2013	Egypt	India	1
Bindal *et al*.[6]	2012	India	India	1
El Fakiri *et al*.[21]	2011	France	Morocco	1
Aydoğan *et al*.[2]	2011	USA	Turkey	1
Thulasiraman *et al*.[94]	2010	India	India	1
Ogbole *et al*.[69]	2010	England	Africa	1
Chang *et al*.[12]	2010	USA	China	1
Gundlach *et al*.[35]	2009	USA	Netherlands	1
Hopsu *et al*.[40]	2009	USA	Finland	1
Wen *et al*.[97]	2009	USA	China	4
Haney *et al*.[37]	2008	Netherlands	USA	1
Momoo *et al*.[61]	2008	Scotland	Japan	1
Hung *et al*.[44]	2008	USA	Canada	1
Price *et al*.[75]	2007	Ireland	USA	3
Fazilah *et al*.[24]	2006	Malaysia	Malaysia	1
Jianhong *et al*.[49]	2006	USA	China	39
Grange *et al*.[34]	2005	USA	USA	1
Hu *et al*.[41]	2005	USA	China	29

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Supplemental Table 1: *Continued*

Author	Year	Country of journal	Author origin	#of cases
Rajendran *et al*.[76]	2004	India	India	1
Gerek *et al*.[29]	2003	USA	Turkey	1
Erdem *et al*.[22]	2002	England	Turkey	3
Jeon *et al*.[48]	2002	Korea	Korea	3
Fernando *et al*.[25]	2002	Sri Lanka	Sri Lanka	1
Sakallioğlu *et al*.[81]	2002	USA	Turkey	1
Reed and Grewal[78]	2001	USA	USA	1
Fitoz *et al*.[26]	2001	USA	Turkey	2
Yoon and Messner[101]	2001	England	USA	1
Sommer and Forte[87]	2001	Canada	Canada	1
Paleri *et al*.[71]	2001	Ireland	India	2
Rossi and Tortori-Donati[80]	2001	Germany	Italy	1
Singh *et al*.[84]	2001	India	India	1
Ng *et al*.[66]	2000	China	China	1
Kwok *et al*.[53]	2000	Australia	China	3
Fan *et al*.[23]	2000	USA	China	1
Sander *et al*.[82]	1999	USA	Turkey	8
Lubianca-Neto *et al*.[56]	1999	USA	Brazil	2
Gürpinar *et al*.[36]	1999	USA	Turkey	1
Chao *et al*.[13]	1999	England	China	8
Indudharan *et al*.[47]	1998	England	Malaysia	1
Sugiyama *et al*.[90]	1998	Japan	Japan	1
Al-Dousary[1]	1997	Ireland	Saudi Arabia	1
Nopajaroonsri *et al*.[67]	1996	USA	USA	1
Bosshardt *et al*.[8]	1996	USA	USA	1
Hussein *et al*.[45]	1996	Germany	Germany	1
Inci *et al*.[46]	1995	Germany	Turkey	1
Mickelson *et al*.[59]	1995	USA	USA	1
Calligaro *et al*.[10]	1995	USA	USA	3
Balik *et al*.[4]	1995	Germany	Turkey	1
Walsh *et al*.[95]	1993	Ireland	England	1
Gorenstein *et al*.[33]	1992	USA	Israel	1
Kuo *et al*.[52]	1992	China	China	2
Walsh *et al*.[96]	1992	England	England	2
Shimizu *et al*.[83]	1992	Japan	Japan	1
Spiro *et al*.[88]	1991	USA	USA	1
Dhillon *et al*.[19]	1991	Singapore	Malaysia	1
Yokomori *et al*.[100]	1990	USA	Japan	2
Nwako *et al*.[68]	1989	USA	Nigeria	1
Zohar *et al*.[102]	1988	Scotland	Israel	2
Hughes *et al*.[43]	1988	England	England	1
Bowdler and Singh[9]	1986	Ireland	England	1
Matsuba *et al*.[58]	1985	USA	USA	1
Som *et al*.[86]	1985	USA	USA	1
Yashiro and Iio[99]	1984	USA	Brazil	2
Furukawa *et al*.[27]	1984	Germany	Japan	1

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Supplemental Table 1: *Continued*

Author	Year	Country of journal	Author origin	#of cases
Stevens *et al*.[89]	1982	USA	USA	1
Passariello *et al*.[72]	1979	Germany	Italy	4
LaMonte *et al*.[54]	1976	USA	USA	2
Gordon *et al*.[32]	1876	USA	USA	2
Gilbert *et al*.[31]	1972	USA	USA	2
Okay *et al*.[70]	1970	USA	USA	1
Garrow *et al*.[28]	1964	USA	USA	1
Gerwig Jr.[30]	1952	USA	USA	1